Rheumatic heart disease in pregnancy: gaps and facilitators of care

Geraldine Ann Vaughan

The Australian Centre for Public and Population Health Research (ACPPHR)

Faculty of Health

University of Technology Sydney

A thesis submitted for the degree of Doctor of Philosophy

at the University of Technology Sydney

Submitted July 2019

Certificate of original authorship

This thesis is submitted in fulfilment of the requirements for the award of Doctor of Philosophy in the Faculty of Health at the University of Technology Sydney.

This thesis is wholly my own work unless otherwise referenced or acknowledged. In addition, I certify that all information sources and literature used are indicated in the thesis.

This document has not been submitted for qualifications at any other academic institution.

Funding support

This research is supported by an Australian Government Research Training Program. Additional research support is gratefully acknowledged from the following awards:

- Australian Government National Health and Medical Research Council (NHMRC) postgraduate scholarship 2017-18 #1132944
- University of Technology Sydney Chancellor's Award 2018
- END RHD NHMRC Centre of Research Excellence #1080401, Telethon Kids Institute, University of Western Australia Top-up Scholarship

Geraldine Ann Vaughan

Production Note:
Signature: Signature removed prior to publication.

Date: 31/7/2019

Acknowledgements

Thank you very much to the qualitative study participants who gave their time, sharing their knowledge and considered thoughts.

My primary supervisor, Elizabeth Sullivan, helped create vision and possibility to make this PhD happen, and provided insights and guidance along the way. Thank you.

I'm enormously grateful to Professor Angela Dawson's critiquing of my work, which has been given so generously, with patience and humour.

Thank you also to Professor Michael Peek whose pithy comments (and quick turnaround!) always carry valuable insight and encouragement.

Professor Jonathan Carapetis, apart from academic critique, continues to provide inspiration and a big picture reminder that the point of this work is to help improve outcomes for a disease we should not have.

I also gratefully acknowledge the support from Professor Lisa Jackson Pulver in the early stages of my doctorate, leading to the first publication in the thesis. Lisa emboldened me to carve out my own space in this research arena.

There is a vast community of practitioners, researchers and advocates in the RHD landscape who have directly and indirectly shaped this research - in particular, the co-writing group for the Women and RHD chapter in the 3rd Edition Australian Guidelines. Similarly, AMOSS site coordinators over the years – and particularly during the RHD study – made the AMOSS studies possible, which in turn informed this doctoral study.

I acknowledge the women who journey with rheumatic heart disease and who tell their stories.

I've been fortunate to be surrounded by good colleagues and friends at UTS Sydney who inspire, give perspective and with whom to share laughs.

Lastly, thank you to my family and friends who - while mostly bemused why I should be doing this doctoral study - understand that it matters. They ground and support me and have helped make it happen.

Thesis format

This thesis comprises two published papers in peer-reviewed journals, and a third qualitative study as part of the thesis.

Published work

Research outputs

Study one

Vaughan G, Dawson A, Peek M, Carapetis J, Sullivan EA. *Standardising clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis*
Birth: Issues in Perinatal Care 2019 00:1-14 [early view]

Study two

Vaughan G, Tune K, Peek M, Jackson Pulver L, Remenyi B, Belton S, Sullivan EA. *Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia*. International Health. 2018;10(6):480-9.

Other work - national guideline development

Vaughan, G (lead author) of the **Women and RHD** chapter in the 3rd Edition Australian Guidelines for Rheumatic Fever and Rheumatic Heart Disease currently in development.

As the lead author of the Women and RHD chapter of the 3rd Edition Australian Guidelines for Rheumatic Fever and Rheumatic Heart Disease[1] currently under development, I have coordinated the structure of the writing group, working with the selected core writing (Dr Miriam Wheeler and Linda Bootle) and broader review groups, alongside coordinator Sara Noonan. Notably, the Chapter was reviewed and informed by community-based and senior Aboriginal leaders Vicki Wade and Karrina Demasi. I have led the development and structure of the chapter and co-written, co-edited and reviewed content. This collaborative work was substantively a new addition; the previous Edition had been a short section in the Guidelines focused on pregnancy only.

The content and direction of the chapter has been informed by (and in turn informs) this thesis. It is an important translational outcome of the research and is in part a response to recommendations based on research findings of the Australasian Maternity Outcomes Surveillance System (AMOSS) NHMRC #1024206 RHD in pregnancy project. It reflects the

increasing recognition that effective models of care must be grounded in respectful, collaborative systems with a life-course approach, working with women and communities.

Statement of contributions to jointly authored works contained in the thesis

Chapters 4 and 6 have been published in peer-reviewed journals. For each of these manuscripts, I have been responsible for deciding the research question, conducting the analysis and drafting the manuscript.

Specific contributions to each published paper and author signatures are listed in Appendix 7.

I take responsibility for the accuracy of the results presented in these manuscripts.

Preface

The principal motivations that have guided this work include:

- The belief that skilled, appropriate, respectful health care during pregnancy is a right for all women;
- Provision of this quality health care for pregnant women with RHD requires integrated services and information systems;
- The experience of implementing the AMOSS study of rheumatic heart disease in pregnancy (RHD-P), which highlighted several challenges and gaps in providing integrated services for women with RHD across multiple disciplines and health services;
- Those gaps in providing integrated care generated inefficiencies and potentially compromised optimal outcomes for mother and baby;
- Conversely, pregnancy provides an ideal time to engage (or re-engage) with women to review cardiac status and ensure that optimal care is provided to maximise best outcomes through the life-course.

Table of Contents

Figures and tables

List of tables

List of figures

Indigenous and First Nation peoples: terminology

The recommendations from 'Communicating Positively, A guide to appropriate Aboriginal terminology' [2] and 'The Australian Indigenous Health*Info*Net guidelines for Aboriginal and Torres Strait Islander terminology' [3] have guided reference to Aboriginal and Torres Strait Islander peoples in this thesis.

Terms such as Aboriginal peoples, Aboriginal and Torres Strait Islander peoples and Indigenous have all been used in the literature. I have used the terms non-Indigenous and non-Aboriginal interchangeably when referring to all other non-Indigenous Australians.

The term Indigenous is capitalized as a form of respect. When making international comparisons I have used the term Indigenous in general; when quoting from other sources I have used the term used in that material.

The term Māori refers to Indigenous peoples of New Zealand (NZ) and Pasifika refers to people living in New Zealand (or Australia) who have migrated from the Pacific Islands or who identify with the Pacific Islands because of ancestry or heritage [4].

Abstract

Background

In the twenty-first century, rheumatic heart disease (RHD) persists in low- and middle-income countries as well as vulnerable populations in high-income countries, particularly Indigenous peoples. RHD in pregnancy (RHD-P) is associated with an increased burden of maternal mortality and morbidity, poorer perinatal outcomes, and compromised care pathways. There is inadequate knowledge regarding models of care for women with RHD-P. This research identifies and examines gaps and facilitators of optimal care for women with RHD-P with a focus on Australian health services.

Methods

The mixed methods research employed a transformative parallel design lens, providing a mechanism with which to address the political and social complexities of research in the RHD-P arena and in turn establishing a framework for change.

The study comprises three elements. The first involved a systematic review and qualitative synthesis of the peer-reviewed literature, that explored approaches to care provision for women with RHD-P and examine reported measures. It applied content analysis to examine models of care and clinical care reporting measures.

The second element entailed a descriptive qualitative study that explored 19 health professionals' perspectives of care pathways for women with RHD-P. The semi-structured interviews were analysed thematically.

The final study conducted a process evaluation of the implementation of a population-based study of RHD-P in Australia. It examined the operationalising of the research project, and evaluated strategies developed to strengthen reporting and improve awareness of the impact of RHD during pregnancy among health services.

Findings

The studies identified gaps related to health systems, health workforces and health information that impacted on effective models of care.

The systematic review found that key reporting measures in studies that refer to RHD-P were poorly recorded.

The qualitative study of health professionals' perspectives of RHD-P identified a constellation of factors that challenged the provision of cohesive women-centred health care. Themes included *conduits of care* - helping to break down silos of information, processes and access; *'layers on layers'* – reflecting the complexity of care issues; and *shared understandings* – factors that contributed to improved understandings of disease, informed decision-making, and the inclusion of family and community members.

The process evaluation of the population level study of RHD-P exemplified several of the themes arising from the previous two studies. Effective reporting was negatively impacted by a lack of diagnostic certainty; incompatible health information systems and varying clinical awareness among health professionals.

Discussion

This research found that, despite often complex care requirements, pregnancy for women with RHD provides a unique opportunity to strengthen health system responses, improve care pathways, address whole of life health and ultimately reduce the burden of RHD for women. To respond effectively, structural and cultural changes are required to improve health system agility and capability. This includes enhanced investment in education and capacity building – particularly in maternal health – to support a better informed and skilled workforce; and improved information systems and reporting of core indicators to more accurately benchmark care pathways, outcomes and burden of RHD-P.

Conclusion

Central to the provision of informed, respectful collaborative care for women with RHD is timely diagnosis, access to health services and continuity of care. The research recommendations based on study findings aim to better achieve these goals and, in doing so, ensure the needs of vulnerable women with RHD are better met.

Chapter 1 Introduction and overview

This chapter provides context for my research and identifies rheumatic heart disease (RHD) in pregnancy from a health services perspective as a potential area of investigation. It briefly describes the underlying aetiology of RHD and its impact in pregnancy. It then outlines global strategies and interventions to combat RHD and describes how RHD in pregnancy (RHD-P) has become increasingly central in the RHD public health landscape of interventions, strategies and policy.

It introduces some of the issues that challenge optimal care for women with RHD-P, defines the overall objectives of the research and briefly describes each chapter.

This background helps provide a framework for the aim and scope of the research and define its research questions.

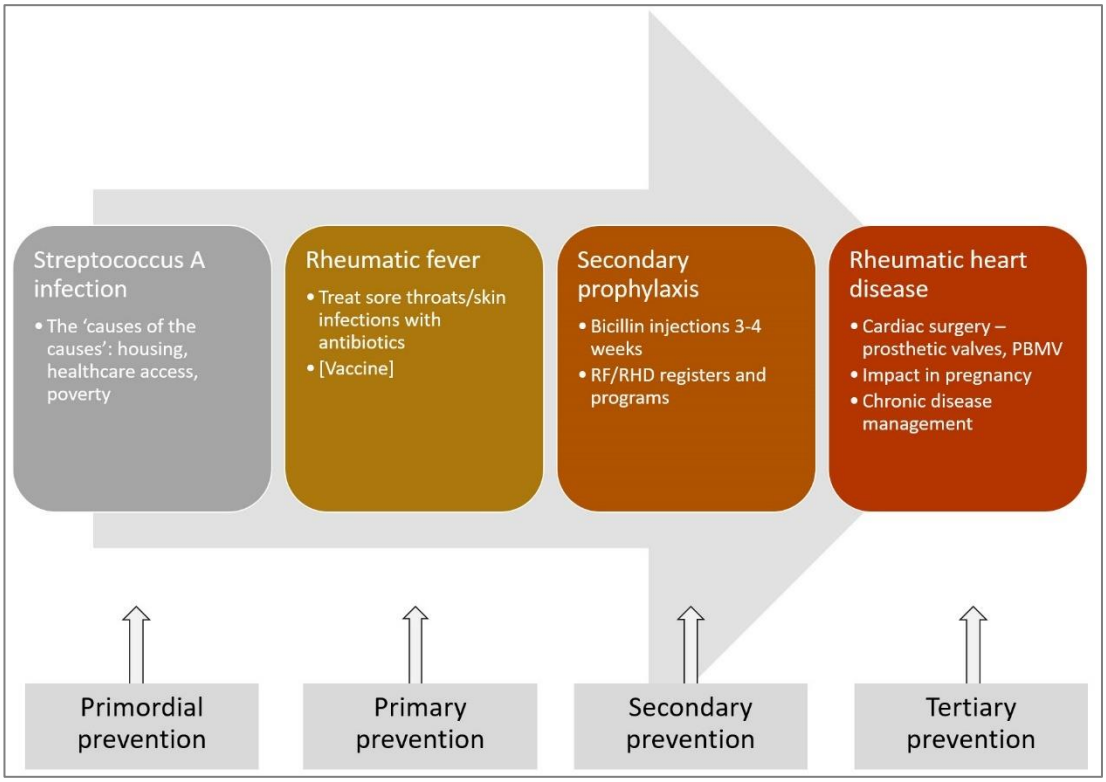
The thesis structure is outlined at the end of the chapter.

Background

The global burden of rheumatic heart disease (RHD) varies enormously according to country, geography and demographics. It is a preventable, chronic sequelae of (usually repeated) episodes of rheumatic fever, in turn caused by an immunological response to Streptococcus A Spectrum infection. Rheumatic fever (RF) and RHD are sentinel markers of inequity and preventable at multiple tiers (Figure 1-1).

The cause of this disease is rooted in a lack of access to adequate housing and health services: a result of social, environmental and economic conditions that predominantly occur in situations of poverty [5].

Figure 1-1: RHD pathway of disease and stages of prevention
(*Adapted from Australian RHD Guidelines* [6])



Secondary prophylaxis is antibiotic therapy used to prevent further attacks of RF and development of the sequelae of RHD. The treatment regimen involves 3-4 weekly injections of (usually) benzathine penicillin G (BPG or bicillin L-A) for a period of ten or more years [7]. Initiation of secondary prophylaxis and the establishment of early care plans is critical in the management of RF to prevent repeat episodes and subsequent development of RHD [8]. Yet implementing these preventative strategies is often hampered by constraints that underlie the 'causes of the causes' related to health service infrastructure and access including the accessibility of BPG [9], particularly in resource-challenged environments.

In the 21st century, RHD persists in low- and middle-income countries as well as vulnerable population in high-income countries, particularly Indigenous peoples [10].

RHD is up to four [11] times more prevalent in women according to country and region, although most studies find it around twice as common in females than males. The impact of RHD-damaged heart valves is amplified in pregnancy, where an increased haemodynamic load can exacerbate existing RHD or unmask previously undiagnosed RHD. Particularly for women with mechanical heart valves or severe mitral stenosis who require anticoagulation,

the results can be catastrophic for them and their babies [6,12]. Mortality rates escalate up to 37% [13] in pregnant women with mitral stenosis from resource-poor countries.

In Australia, RHD is found predominantly among Aboriginal and Torres Strait Islander peoples, with other populations disproportionately affected including Māori and Pasifika peoples, migrants from resource-poor countries and refugees. It had become a forgotten disease in the latter half of the twentieth century in Australia [14] which was concomitant with a decline in awareness of the epidemiology, clinical impact and lived experience of RHD-P.

Overall extremely rare (4 per 10,000 women giving birth) [15,16], in remote Aboriginal communities this escalates dramatically, with an estimated 2-3% of pregnant Aboriginal women each year in the Northern Territory [16].

Despite this increased risk and impact, no information on RHD in pregnancy has been routinely collected [16]. Targeted prevention policies and advocacy initiatives developed to address RHD in Australia and New Zealand were primarily focused on vulnerable children and adolescents with little specific reference to pregnancy until the late-2000s.

The last decade has seen a groundswell of research and advocacy initiatives to position RHD in the global health arena and recognise its impact in pregnancy. The 71st World Health Assembly (WHA) statement on RF/RHD (adopted mid-2018) marked its recognition as a global health priority, and noted the disproportionate burden among pregnant women and Indigenous populations of this preventable disease known to affect at least 33 million individuals and cause over 300 000 deaths annually [17]. The resolution was affirmed within the context of the Global Strategy for Women's, Children's and Adolescents' Health [18], advocating for Member States to include specific RHD interventions (such as RHD registers and control programs) in the Strategy's operational plan [19].

During 2012-2016, an Australian National Health and Medical Research Council (NHMRC)-funded project (#1024206) utilising the Australasian Maternity Outcomes Surveillance System (AMOSS) as an umbrella system to conduct two studies of the impact of RHD-P. A population-based observational study across Australia and New Zealand examined the clinical impact of RHD during pregnancy and the early post-partum [16]. A second study explored pregnant women's lived experiences of RHD in the Northern Territory [20,21].

In 2015, the END RHD NHMRC Centre for Research Excellence commenced a series of projects across several disciplines of research to provide an endgame approach to RHD, with an overall aim to bring the prevalence of RHD for Aboriginal and Torres Strait Islander Australians down to the same level as non-Indigenous Australians by 2031 [22].

Against this backdrop of escalated global strategies and policy and research initiatives, some of the particular challenges of providing optimal care for women with RHD-P are emerging, as are opportunities and scope for change. Clinical care and policy related to RHD-P extends across several health spheres – maternal and cardiac health, Indigenous and primary health, chronic diseases, public health and policy. Emerging findings of the AMOSS [16,20] and other studies highlight systemic and cultural deficiencies in health services responses to a preventable disease among vulnerable populations that in turn create escalated risk in pregnancy.

The increasing calls for health services to take a more integrative, comprehensive approach to RHD is congruent to approaches for other chronic conditions such as diabetes and malaria. This shift is expected to accelerate, spurred by global efforts to improve universal health coverage and integrated, people-centred health services [23], particularly within the framework of achieving the Goals and targets of the Sustainable Development Goals [24].

The need for models of care that address the whole-of-life for women with RHD is evident in both the low rates of preconception care for women with RHD, late diagnosis of RHD and the high rates of perinatal morbidity and mortality (including postpartum mortality beyond the standard reporting timeframe).

A lack of strategy and coherency of care persists for pregnant women with RHD, where a complex interplay of socio-demographic and systemic barriers and issues of equity compound challenges to the provision of optimal care [20].

Yet in these challenges lie opportunities. Pregnancy provides the potential to work with women, developing a whole-of-life approach to a chronic condition, improve cardiovascular and perinatal outcomes and overall health. To do so demands an integrated health care response from an informed and skilled health workforce.

This research explores themes arising from the AMOSS study relating to health services: the barriers and facilitators to optimal care for pregnant women with RHD. It aims to use

findings to inform decision making-policy and practice and improve health care and services for pregnant women with RHD.

In the course of placing a lens on models of care for women with RHD, the research takes a global perspective, particularly in the background and the Study 1 systematic review. In Studies 2 and 3, an Australia-specific focus is taken, with Aboriginal and/or Torres Strait Islander women representing nearly 80% of the burden of RHD-P in this country[16]. Precepts of woman-centred care must be shaped by local community needs and culture which are likely to significantly vary for other vulnerable populations, whether Maori women living in south-east Queensland, a refugee woman from Sudan, or a recent immigrant from Nepal (or, indeed, between Aboriginal and Torres Strait Islander populations). Such variation will equally apply in other country or region-specific settings. However, it is suggested that broad principles underlying the research recommendations related to women with RHD may be applicable in other contexts.

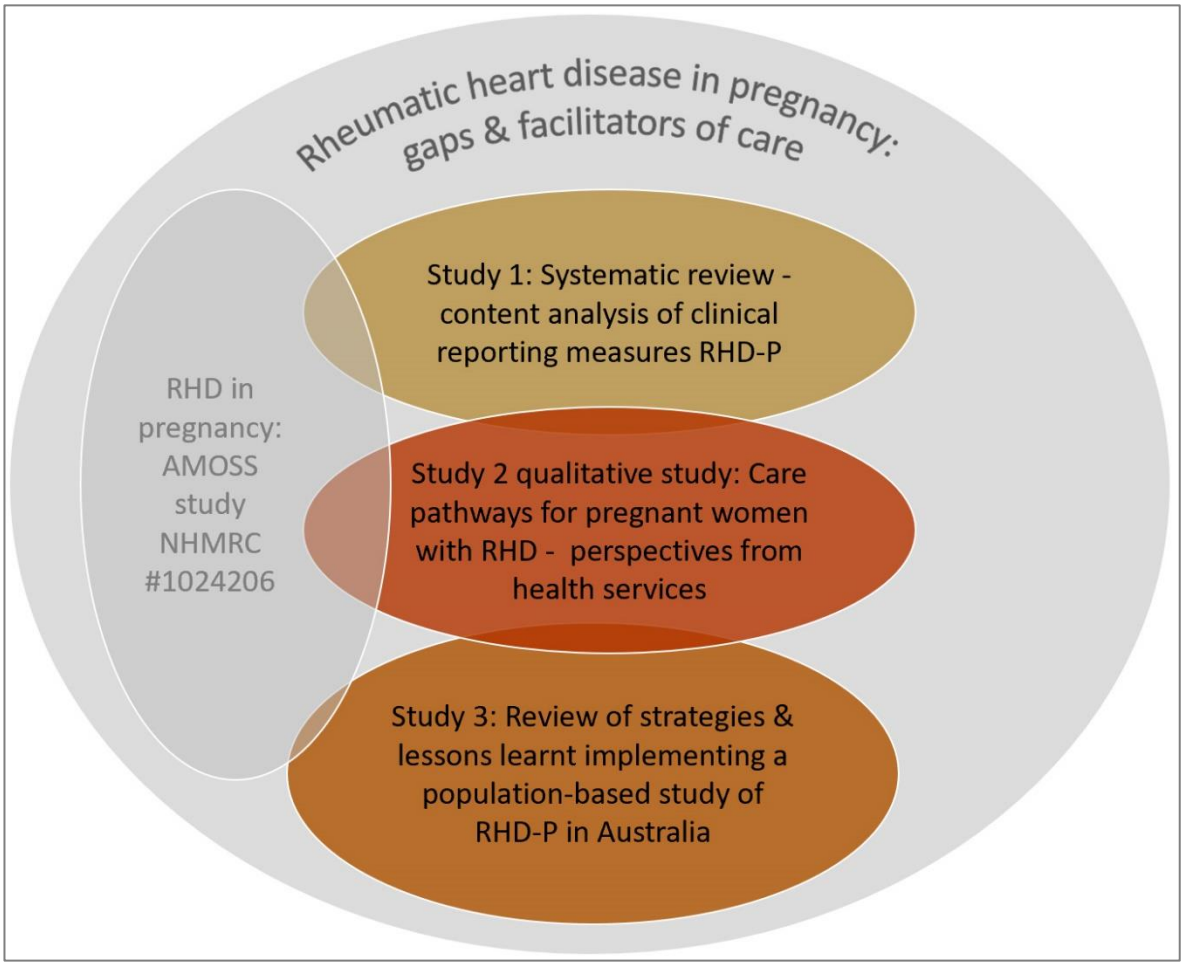
Aim and objectives

The overarching aim of this doctoral research is to investigate the factors that impact on the planning and provision of optimal care for pregnant women with RHD, with a focus on health services. Three studies were conducted to support this aim and answer the following research questions:

1. What guidelines and reviews inform or have relevance for models of care for pregnant women with RHD? (Literature review and Study 1)
2. How are attributes of care for women with RHD-P reported in the literature and how do these align with outcomes described in evidence-based guidelines? (Study 1)
3. What levels of knowledge, expertise and awareness exist amongst health professionals regarding care pathways for pregnant women with RHD and its burden? (Studies 2, 3)
4. What do health professionals perceive to be the barriers and facilitators to the provision of optimal health care for women with RHD? (Study 2)
5. What policies and strategies do health professionals suggest are required to more effectively meet the needs of these women? (Study 2)

The figure below visually depicts the three studies and their relationships to each other. They form a series of interrelated explorations that were informed by and in part arose from the AMOSS study.

Figure 1-2: Overview of studies and how they relate



Thesis structure

This PhD thesis is in a compilation format. Each of three studies is reported in a thesis chapter. Chapters 4 and 6 have been published. Chapter 5 is being prepared for publication post-doctorate.

The thesis has seven chapters, each of which is briefly described below:

Chapter 1: Introduction

This chapter introduces the thesis, including background, rationale and research objectives.

Chapter 2: Literature review

This chapter examines the literature on rheumatic heart disease in pregnancy at a more granular level than the broad overview given in Chapter 1. It introduces an historical context, examining models of care and impact of RHD-P from a public health perspective and exploring possible lessons to be learnt from medical history. The review describes the changing epidemiological landscape of RHD including its shifting global burden and examines its relevance in pregnancy particularly. Barriers to optimal care for women with RHD-P are discussed.

Based on this review, an examination of guidelines with relevance to models of care for pregnant women with RHD follows. This examination extrapolates core principles of care against which selected studies including pregnant women with RHD are assessed in Chapter 4.

Chapter 3: Research methods

This chapter explains the methods used for the research.

Chapter 4: Study one

'Standardising clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis'

A systematic review that aims to improve understanding of reporting of attributes of care for women with RHD-P and how these align with outcomes described in evidence-based guidelines.

Chapter 5: Study two

'Perspectives of care for women with RHD-P: a qualitative study'

This chapter presents findings from a qualitative exploration of care pathways of women with RHD from the perspectives of health service providers. During the thematic analysis of interview data, I found a broad range of factors which participants identified as potential barriers and facilitators to the care pathways for women with RHD. The research reflected the demand for a richer understanding of the contextual realities of providing care for pregnant women with RHD.

Chapter 6: Study three

'Review of strategies and lessons learnt implementing a population-based study of rheumatic heart disease in pregnancy in Australia'

This paper reviews processes developed to identify pregnant Australian women with RHD during a two-year population-based observational study using the AMOSS. It evaluates strategies developed to enhance reporting and discusses implications for patient care and public health. The review is sequenced as the third study, illustrating many of the issues arising and themes presented from the previous chapters, and, in turn, informing parts of Studies 1 and 2 (Figure 1-2).

Chapter 7: Discussion, recommendations and conclusion

This chapter summarises the results of Chapters 4 to 6. My research findings are contextualised within two frameworks - the Sustainable Development Goals and a chronic care model - to provide a lens with which to understand the emerging themes from this study. This gave direction to shape the recommendations arising from my research.

Chapter 2 Literature review

This chapter gives context to the thesis, including:

- An historical global perspective of RHD-P, outlining initiatives and strategies that have been developed to address the condition
- An examination of historical public health responses to RF and RHD
- A brief description of the clinical impact of RHD-P, introducing some of the complex factors that challenge provision of optimal care
- A discussion of the shifting global burden of RHD and changing advocacy landscape in the twenty-first century
- A particular focus on Australia
- A detailed examination of Guidelines with relevance to models of care for pregnant women with RHD. This review extrapolates core principles of care against which selected studies including pregnant women with RHD are assessed in Chapter 4.

Background and context

A history of broken hearts: rheumatic heart disease in pregnancy

The temporal focus of this chapter is mainly from the latter part of the twentieth century to the present: reflecting the current (and shifting) epidemiological profile of RHD and associated landscapes of research and advocacy. However, I was also interested to explore both change and continuity in relation to RHD-P: what was the evidence of burden of disease over the millennia? What principles have informed the care of women during pregnancy with RHD? What changes in treatment strategies have occurred over time; and conversely, what issues and approaches to care have remained broadly the same? Why are RHD advocacy initiatives and policy-shaping so integral to improving maternal outcomes for women with RHD-P? How have these themes played out in Australia? I was also curious where there were gaps: what can we learn from the *absence* of reference to pregnancy in the context of clinical care and research of RHD?

The documented history of RHD-P over the centuries, while limited, provides a compelling tale. It demonstrates the impact of the condition from 2,500 years ago to the current day and reflects a little of the changing landscape – as well as constant themes - in relation to the burden of RHD-P, outcomes and models of care. In looking backwards, a context is given [25] that provides a useful tool to strengthen advocacy. While health systems across the

centuries and across the world may differ hugely according to developments in knowledge, technologies, economies, and social structures, three underlying themes inform much of the literature of RHD-P: the association of RHD with poverty, recognition of the often-complex care requirements during pregnancy that includes several health domains; and demands for integrated care models to optimise perinatal outcomes.

Evil effects of the pregnancy in disturbing the cardiac equilibrium: **risk through the millennia for pregnant women with RHD**

Knowledge of the impact of a compromised heart valve in pregnancy due to RHD is not new. Nearly 2,500 years ago, Hippocrates discussed the classic symptoms of rheumatic mitral stenosis in a pregnant woman:

> *"Harpalida's sister, in the fourth or fifth month of her pregnancy, had watery swellings in her legs, swellings in the hollows of her eyes, and her whole body puffed up… Sometimes she was so near to suffocation that she was obliged to sit up in her bed without being able to lie down; and if she tried to sleep it was in a sitting position. Yet there was not much fever. For a long time the foetus did not move, as if it were dead. . . ." Epidemics VII VI (Littre, 1839-1861; Clifton, 1734) in* [26]

In the 1800s, several monographs and books noted the poor outcomes of pregnancy complicated by cardiac disease, which was overwhelmingly caused by RHD in the overcrowded urbanised centres of the UK, Europe and North America. The death of a young woman in labour due to mitral stenosis complications was a key driver in renowned 19th century Scottish cardiologist James Mackenzie's work to improve the clinical management of heart disease [27].

> *My attention was early directed to the subject of Heart Disease and Pregnancy by a tragic experience which occurred … about forty years ago. I attended a young woman, thirty-five years of age, for a miscarriage at the third month of her first pregnancy. I discovered then that she had a presystolic murmur… I had a vague notion that mitral stenosis was a serious embarrassment to the heart in pregnancy, but of the source of danger and its nature I had no knowledge.*
>
> *The woman left my neighbourhood for a year, at the end of which time she returned … seven months pregnant, and with a considerable oedema of the legs. … As the labour proceeded little progress was made; the suffering was*

very great, and there was marked distress in breathing…Moreover, so profound was the sense of suffocation that attempts to give chloroform were resisted. Two practitioners of much experience saw her, but after many hours of suffering she died undelivered, from extreme exhaustion [28].

Around the same time, fellow Scotsman and obstetrician Angus Macdonald wrote his textbook on cardiac disease in pregnancy that was used in obstetrics for the next 50 years. There was little on congenital heart disease in this work: in the nineteenth century, women did not tend to survive this spectrum of disorders long enough to become pregnant. The majority of his focus was on mitral stenosis of rheumatic origin:

We have thus nine cases out of the fourteen [pregnancies], or 64.4%., fatal, which indicates a tendency to death in the combination of mitral stenosis with pregnancy which is surely sufficiently grave… [29].

However, despite such outcomes, clinicians such as McKenzie, Macdonald and others [30] did not proscribe pregnancy altogether, but counselled according to lesion and individual clinical status, developing a risk stratification in clinical care.

… given that one of our patients is the victim of a special cardiac lesion,—we should be able to predict what are the special additional risks, if any, to which the pregnant, parturient, and lying-in conditions expose her, and what are the prophylactic or therapeutic measures we are bound to adopt, so as, if practicable, to avoid or diminish such risks. Such a result, I opine, can only be attained by a careful and unbiased investigation into the physiological and pathological conditions involved, and perhaps even then can only be imperfectly secured [29].

Macdonald concluded that…

These good results encourage us to believe that judicious and skilful management of similar cases, both throughout the pregnancy and at delivery, might greatly lessen the risks associated with lesions of this nature [mitral stenosis] *[29].*

He also mused on the professional and ethical responsibilities of obstetricians to advise women on pregnancy choices (with marriage seen as a proxy), which he argued were largely unconsidered among physicians:

> *I know of no English writer who has striven to put obstetricians in a position to answer with intelligence the question that is every now and again asked of us by a patient who knows she has heart disease, — Should I marry? or do I run great risk in marrying? … What is more, I think I shall be able to show in the sequel, that not only have physicians not written on this subject, but that a great amount of ignorance exists in the professional mind upon the matter...* [29]

The first iteration of the New York Heart Association (NYHA) functional classification of cardiac disease based on clinical severity and prognosis [31] was published in 1928. It was swiftly adopted as a risk stratification in pregnancy [32-36]. The first known longitudinal study of maternal outcomes over consecutive pregnancies in 169 women used the NYHA class to mark the progression of cardiac disease [33].

> *Of recent years emphasis has shifted more and more away from the actual lesion present and towards the recognition of the great importance of judging the likely outcome during pregnancy from a simple knowledge of the degree of activity which the patient can achieve. It is one of the first duties of the cardiologist to place his [sic] patient in one of the four categories suggested by the New York Heart Association (1939)* [36].

This article drew on research of others from the 1940s [33,35,37] to conclude that RHD of itself was not an indication for caesarean section which in fact should be avoided unless indicated by obstetric reasons or severe cardiac compromise [36].

An earlier comparative study of deaths in men and (nulliparous and parous) women with RHD concluded that …*"while pregnancy should be avoided in the severer grades of rheumatic heart disease … one or two children may be borne without detriment by the majority of cardiac women"* [38]. Notably, autopsies from all cases in that study showed a degree of mitral stenosis.

The cramping effects of too-rigid specialism: care across disciplines

There have been calls for collaborative cardiac care for pregnant women between professional specialisations from the late nineteenth century. In 1878, Macdonald admonished his British obstetric colleagues to devote more professional attention to cardiac disease in pregnancy … *"commensurate with its importance"* in order to better *"…differentiate the effects of the special cardiac lesions, and to define in any way their individual bearings upon either pregnancy or parturition".*

Obstetricians' *"rigid specialism"* he declared, compromised effective management of women under their care, and led to much of the *"disfavour and affected hauteur with which obstetrical medicine has frequently been treated by pure physicians and surgeons"* [29].

Macdonald also referenced good nursing care which was … "*Under no conditions … more needed, or likely to be better rewarded, than in warding off the exciting causes of pulmonary disturbances in connection with pregnancy complicated with various cardiac lesions"* [29], but disciplinary references outside cardiac and obstetric specialisations were largely absent.

However, the next half century did not bring much evidence of better general awareness of the impact of cardiac disease in pregnancy in maternal health, nor improved collaborative care. Some forty years later, McKenzie observed with concern that knowledge of cardiac disease among obstetric physicians was generally poorer than when Macdonald had written his 1878 treatise [28].

The objective of MacKenzie's 1921 textbook was to foster enhanced knowledge in the cardiac discipline for obstetric physicians. He spoke with insight of the fear of consequences for women and the need to provide informed and educated advice:

> *"These disastrous [cardiac] happenings surround a natural process with dread and mystery, for the dread is aggravated by the fact that the source of danger is not clearly realised. As a result all sorts of signs are looked upon with suspicion—signs innocent as well as signs grave. Needless to say, this obscurity does a great deal of harm. Many women are subjected to unnecessary alarms and restrictions when pregnant ; others have to suppress the natural desire of motherhood…"* [28]

The joint cardiac-obstetric clinic established in 1928 at the Edinburgh Royal Infirmary/Royal Maternity Hospital saw cardiac deaths as a percentage of cardiac cases (94% of which had underlying rheumatic pathology) drop from 6.3 to 0.9% by 1947 [33].

Across the Pacific around this period, the collaborative care and services that Macdonald and McKenzie had called for were being developed in North American centres. Similar trends were described by Hamilton, although the maternal mortality was double that of Haig and Gilchrist's study. When a cardiac clinic commenced at Boston Lying-In Hospital in 1921, the maternal mortality rate was 20% with the underlying cardiac condition in 93% of

these women due to RHD. This mortality dropped to under 5% in the ensuing three years [39]. Women were divided women into 'favourable' and 'non-favourable' cases, with Hamilton finding that …

> *"A pregnancy, then, has cost the favourable cases little, if any, more risk than their risk of death in one year of living, but it has cost the unfavourable cases a risk nearly three times greater, and those that have auricular fibrillation* [atrial fibrillation, particularly secondary to rheumatic mitral stenosis], *a risk that is four times greater" [39].*

Treatment (rather than the rheumatic condition itself) was the largest factor in prognosis for pregnancy in this study. Regular antenatal review during pregnancy was stressed as an essential component of care [39]. This was the first known statistical reporting of the impact of care in determining outcomes.

A 1936 Canadian study called for the… *"establishment of a combined prenatal and cardiac clinic which has permitted personal consultations between cardiologist and obstetrician and a continuity of observation of all heart cases"* [40].

From the 1930s, similar studies in London and Dublin found that, with adequate collaborative cardiac-obstetric care the need for medically advised terminations of pregnancy was reduced significantly [41]. McIlroy (the first UK female obstetrician and medical professor) detailed the mutual benefits of obstetric-cardiac collaboration (and comprehensive antenatal care), including earlier diagnosis of cases that had previously been recognised when the woman entered labour [41].

She recognised the overwhelming burden of RHD in cardiac pathology:

> *"By far the largest number of cases, however, are those of chronic valvular disease, which is practically always rheumatic in origin even though there may be no definite rheumatic history" [41].*

A prospective study of 539 pregnancies of women with RHD commencing in Dublin in 1948 found that … "*early diagnosis and adequate supervision can virtually eliminate the immediate hazards of pregnancy in cases of rheumatic heart disease"* [42].

A review of women with RHD-P at two UK sites over a 28 year period (1942-1969) concluded that *"routine medical examinations at about 10, 15, and 20 years of age with*

appropriate management would render pregnancy virtually safe for every patient with rheumatic heart disease" [43].

However, such outcomes were challenged by a lack of interdisciplinary collaborative approach to care which persists to the current day:

> *Another difficulty arises from the different way in which the cardiologist and the obstetrician regard the problem. The cardiologist considers the pregnancy as complicating the pre-existing heart disease, but the obstetrician considers that his [sic] patient's pregnancy is complicated by her cardiac lesion. Another cause for ignorance is that the obstetrician rarely studies his patients once the puerperium has been successfully passed, and if further pregnancies do not ensue he may never see the patient again. On the other hand, the cardiologist rarely follows the same patient through several pregnancies unless he is particularly interested in the subject, and his opportunities for observing closely the heart in normal pregnancy are few [36].*

A properly conceived public health program: rheumatic fever/RHD strategies

These initiatives and findings were against a backdrop of better understanding of disease burden: rheumatic fever (RF) and RHD had become part of the public health discourse. Its magnitude and impact were severe in the early-mid twentieth century: Swift reports that in 1938 New York, there were five times more deaths from RF/RHD than the combined total of six common reportable diseases, and it was on par with tuberculosis [44].

There was a growing recognition of the association of RF/RHD with poverty:

> *"No disease has a clearer cut 'social incidence' than acute rheumatism [rheumatic fever] which falls perhaps thirty times as frequently upon the poorer children of the industrial town as upon the children of the well-to-do …The incidence of acute rheumatism increases directly with poverty, malnutrition, over-crowding and bad housing (Glover 1930 quoted in [45]).*

By the 1940s, peak bodies in the USA and Britain were grappling with how to best address RF and RHD. From the London Rheumatism Scheme, to the various cardiac clinics in the USA and initiatives in Ireland: most of these public health measures focused on RF and RHD in children, with much reference to school programs [44,46-48]. Rutstein, a public health physician, advocated strongly for coordinated approaches to RF/RHD straddling cardiac, nurse, and occupational therapy, although curiously he did not include maternity services

despite referring to a study which found that half the women with RHD had no recollection of the condition [49].

The shifting global burden of RHD: country, fiscal and maternal

The epidemiology of RHD generally mirrors the evolution of nation economies and associated changing health profiles. By the 1960s, the overall prevalence of RHD was waning in high-income countries [50-52], driven by improved medical care, living conditions, economic development, antibiotics and the possibly altered virulence of circulating Streptococcal strains [53-55].

While RHD was still the most common cause of maternal deaths in some high-income settings [56], studies of the impact of cardiac disease in pregnancy in these countries began noting the increasing proportion of congenital heart disease compared to the decrease in RHD in studies of maternal morbidity and mortality trends [57,58].

Unsurprisingly, this decline was not shared by low-income countries, and the 1970s brought reports of newly identified high-prevalence regions such as India, Africa, and the Pacific [59]. The upper-middle income country of South Africa has some of the highest rates in the world, with its World Bank classification [60] offset by major inequalities of income distribution [61]. Reflecting the association of RHD with poverty and deprivation, the reduced burden of disease has varied *within* high-income countries [10,62-65], with continued high prevalence among American Indian [66] and Inuit populations [10,67] as well as Australian Aboriginal and Torres Strait Islander and New Zealand Māori and Pasifika peoples.

The overall global burden of RHD remains high - and likely underestimated. An estimated thirty-three million people live with RHD globally [68]. They are young (with a median age of 28 years in a two-year follow-up of the global rheumatic heart disease registry in low-middle income countries). They die young (with a two-year case fatality rate of 16.9%) [69]. Recent estimates of 314,600 (CI 302,300 to 328,700) [52] all-age deaths per year globally due to RHD are likely under-ascertained for a variety of reasons. The burden of RHD has not been matched by proportionate investment in public health responses. RHD mortality is comparable to that of rotavirus, and about 50% of that of malaria [70], yet only 0.07% of global funding is directed toward RF, and less again for the treatment and prevention of RHD [71].

Systematic differences in the reporting of - and diagnostic approach to - RHD exist, reflecting differences in local experience and disease patterns. Underreporting is due to many factors: a lack of high quality (or in some cases any) data from low-resource high-prevalence countries and regions; methodological differences between studies [72], clinical (under)diagnosis [65] and – until recently - a lack of uniform definitions and diagnostic definition of RHD on echo [73-79].

The most recent Global Burden of Disease Study places RHD in the leading 30 Level 3 causes of total years of life lost (YLL)s in low-middle income countries according to Socio-Demographic Index (SDI) grouping, yet it is absent in the low SDI country grouping [52], (although 'other cardiovascular diseases' are listed in this group), suggesting RHD is highly likely to be part of this category.

In 2012, the World Heart Federation (WHF) echocardiographic criteria for diagnosis of RHD was published, aiming to make the diagnosis of RHD on echocardiogram consistent, reproducible and based on the best available evidence [74].

Even ignoring the critical social justice issues of RF/RHD, its neglect in public health and funding arenas [71,80] makes for poor policy in economic terms: it costs $5 to treat a sore throat, compared to lifetime valvular surgery costs upwards of $29,000[81]. In his doctoral thesis, Schoon found (in his "probably grossly underestimated" costings), that heart disease with life-threatening complications in South African pregnant women requiring emergency referral to his tertiary institution was eight times more expensive than uncomplicated heart disease. The treatment costs for women who did not attend clinics and who subsequently developed complications was almost 200 times more than those without complications. Unsurprisingly, the most expensive group to treat were found to be women in whom the diagnosis was made only after presenting with heart failure [82].

The burden of RHD is borne disproportionately by women, with up to four times more females than males [11], although most studies suggest rates around twice as high [69,83-86]. Similar to overall RHD rates, these are no doubt underestimated: a high prevalence of subclinical RHD (2.3%) was found in one of the few echocardiographic screening studies that focused on pregnant women in the low-income setting of Eritrea [87], while a more recent Ugandan longitudinal study found a 1.5% community prevalence of confirmed RHD 1.5%, with only 3.4% known prior to pregnancy [88].

A study of the clinical and economic burden of RHD in India and Uganda found that women accounted for the majority (71-80%) of the disability-adjusted life years (DALYs), bearing a disproportionate clinical burden from pregnancy-related complications; in Uganda, women bore 75% of the total cost [86]. When the indirect costs of economic impact are taken into account – family, community, work – this increases exponentially.

History of RHD and RHD-P in Australia: burden, trends, and strategies

Australia and New Zealand shared similar epidemiological RF/RHD profiles in the early-mid twentieth century to other European and North American regions [89,90]. In Australia, the rates of all-age all-female deaths from RHD in 1931 were 53 per 100,000 [91]. A 1937 review stated that NSW occupied an 'intermediate' position of risk of cardiac rheumatism, compared to known data from the UK, northern USA and from other Australian jurisdictions (noting an apparent low rate of incidence of rheumatic fever in the tropics [92], based on Clarke's assertions [93]).

Aboriginal identification is not specified in this review: until the 1967 Australian Referendum when Section 127 of the Constitution was repealed, Aboriginal people were excluded from census data collection [94]. Maddox did note the 'similar incidence' among Aboriginal children, based on notifiable cardiac conditions in 766 examinations in schools attended only by Aboriginal children [92].

The wonderfully titled and methodologically flawed '*Morbidity in the Australian Housewife'* gave an estimated rate of 2 per 1000 of women with RHD each in both married and unmarried categories (not broken down by age groups) during 1962-63, based on the review of a twelve month National Morbidity Study [95].

There was a steady overall Australian decline of RF/RHD from the 1940s: all-age all-female deaths from RHD dropped to 9.3 in 1950 and 1.3 per 100,000 in 2000 [91]. This was reflected in very limited RF/RHD reference in Australian mainstream medical literature from the 1970s to late 1990s and less so of its impact in pregnant women.

A 1976 paper that compared East Africa and Australia cardiac disease based on personal experience contrasted the 30% of RHD-caused cardiovascular disease in Uganda to that of Australia where it had significantly reduced *"…except in underprivileged groups such as Aboriginal children in the tropical areas of the continent and the former Territory of Papua*

New Guinea". The paper highlights the particular risks that include *"… a large number of the younger female patients [who] risk deterioration of their cardiac status in pregnancy."* [96].

A retrospective study in the Kimberley WA in the 1980s found the annual incidence of RF up to 4.7 times higher than that of 1938-48 Melbourne, Victoria. It estimated that up to 89% of cases were in Aboriginal patients in this region, with an all-case incidence twice as high in females, and three times as high for recurrence [97], although there was no discussion of the consequence this held for women during pregnancy.

Apart from the Kimberley region, particularly high rates of RHD in the Northern Territory (NT) and far north Queensland (Qld) have been documented since the 1990s. In the NT, 92% of people with RHD are Aboriginal and/or Torres Strait Islanders, of whom 85% live in remote communities and towns. The documented all-age prevalence of RHD in the NT has steadily increased from around 1.3-1.7% in 2002 [14] to almost 2% of the Aboriginal population (19.4/1000), with a prevalence of 3.2% in the 35–44 year age group in 2008 [55,98] (likely to be partly due to improved diagnosis and data collection [98]).

The prevalence in the 5–14 year age group in 2008 was 8.5 per 1000 [55]. These NT data compare to an estimated RHD prevalence of 7.7/1000 in 5-14 year-olds in 1937 London [54] and to the 5-15 year old prevalence of 5.7/1000 in Sub-Saharan Africa [70]. A 2018 screening study conducted in Arnhem Land, Northern Territory found one in 20 school age children had RHD [99,100].

By 2011, RHD was among the top 20 causes of death in Aboriginal and Torres Strait Islander women. Its burden represents the largest relative difference between Indigenous and non-Indigenous Australians of any cardiovascular disease with an age-standardised rate ratio of 6.6 [101].

While these data show the burden of RHD is overwhelmingly among Aboriginal and Torres Strait Islanders in high-risk remote and regional Australia, it is also disproportionate among other vulnerable populations, particularly Māori and Pacific Islanders and migrants (especially refugees) from resource-poor countries [6]. In neighbouring Timor Leste, a recent screening study showed that 3.5% of school-aged children had definite or borderline rheumatic heart disease, with girls over-represented in the numbers [102].

It is difficult to ascertain trends and distribution of the burden of RHD-P across Australia, with no population-based studies on pregnant women with RHD until recently. A 1990

review of maternal mortality trends noted the downward trend of death due to RHD between 1970-1986 [103] and compared to a Victorian report of 1953-67 where 32% of women who died due to heart disease had underlying RHD [104]. It cautioned of the increasing number of women with "residual rheumatic damage" who were immigrants from the Eastern Mediterranean and South East Asia [103].

Neither of these reports referred to Aboriginal and Torres Strait Islander women. Total confinement data for Indigenous women were not available Australia wide until 1991 [105]. Indigenous identification continues to be under-ascertained [106]: a long-term concern that affects the accurate monitoring of RHD and associated health outcomes for Aboriginal and Torres Strait Islander peoples [107].

We do know that cardiovascular conditions are among the leading contributor to maternal deaths, which have stubbornly remained up to three to five times higher than non-Indigenous women [108,109]. The age-standardised maternal mortality ratio (MMR) for Aboriginal and Torres Strait Islander women between 2012-2016 was 31.6 deaths per 100,000 confinements, around 4.6 times higher than the ratio of 7.1 per 100,000 for non-Indigenous women [109]. This report excluded Northern Territory data.

In 2013, a two-year observational study of the prevalence, management and outcomes of RHD-P across Australia and New Zealand under the umbrella of the Australasian Maternity Outcomes Surveillance System (AMOSS) [110] was conducted. While overall rare (4.3 per 10,000 pregnancies in Australia) in the NT this escalated to 211 per 10,000 in Aboriginal women [16].

Registers, Control Programs and data collection systems

The most effective approach for direct control of RF and RHD is secondary prophylaxis, which is best delivered as part of a coordinated control program (Figure 1-1). However, along with the variable data on the Australian RHD burden outlined in the previous section, the lack of acknowledgement at the national level of the growing impact of RF/RHD in Aboriginal populations [111] until the early 21st century precluded a systematic program approach. Responses to RF/RHD among Aboriginal peoples until the late 1990s were isolated and usually community driven [112]. The NT drove the first regional (Top End and Centre) programs [53,113].

The early 2000s saw a shift in policy and political will, no doubt galvanised by increasing advocacy and locally-driven initiatives. By 2016, there were five jurisdictional-based programs (NT, Qld, WA, South Australia (SA) and New South Wales (NSW)). RF is a notifiable disease in five jurisdictions; however RHD is not notifiable [114] except in NSW for under 35-year olds [115].

In 2006 the National Heart Foundation of Australia (NHF), Heart Foundation of New Zealand (HF) and the Cardiac Society of Australia and New Zealand (CSANZ) convened health experts to develop strategies for the treatment and control of RF and RHD. The resultant review and guidelines developed in both Australia and New Zealand (hereafter Australian RHD Guidelines and NZ RHD Guidelines respectively) address factors contributing to diagnosis and management of RF/RHD in these countries [6,116,117].

In 2009, RHDAustralia was established to provide a national, coordinated approach to RHD programs. Its development of a central repository for the collection and reporting of RF/RHD data against a recommended clinical data set and key performance indicators are now located within the Australian Institute of Health and Welfare's MetaData Online Registry [114]. There were recommended data items on pregnancy and estimated date of birth included in this recommended dataset, however these were not mandated [6,118]. From 2018, with its subsequent shift in focus to working with health systems to achieve focused prevention activities in high-risk communities [114], as well as updated guidelines and education tools, RHDAustralia is emphasising the impact of RHD in women with a suite of initiatives including advocacy [119], multiple-language films [120] and midwifery curricula materials [121].

Of course, the most desired outcome is the prevention of RF/RHD through primordial measures, which demand addressing the socio-economic determinants underlying the disease: the 'causes of the causes' [122].

Still neglected? RHD and RHD-P in the twenty-first century

This changing landscape of Australian awareness of - and responses to - RF/RHD has been matched world-wide. Since the early 21st century, a strengthened wave of advocacy for RHD [71,123-126] has emerged, supported by increasing evidence of its global burden. The 2013 World Heart Federation (WHF) position statement on RF/RHD put forward a strategic goal of a 25% reduction in premature deaths from RF/RHD among individuals aged <25 years by the year 2025, encompassing five key targets: comprehensive register-based control

programs, global access to benzathine penicillin G, identification and development of public figures as 'RHD champions', expansion of RHD training hubs, and support for vaccine development [74].

The last decade has seen initiatives in the primary and secondary prevention of ARF/RHD [127], advancements in pathogenesis and biomarker research as well as an increased number of country registers [128] and steady progress in vaccine development [129] with increased funding commitments [130]. Improved diagnosis has been supported by technological advances in handheld echocardiography equipment, telemedicine, task-shifting for non-experts and standardised training [131].

An important development has been the increased focus on (and working with) people living with RHD and frontline health workers delivering essential RHD services [119,132-134].

At the 2013 World Health Assembly, RHD was anchored in the non-communicable diseases agenda, with inclusion in the Global Action Plan [123]. In 2018 the Executive Board of the World Health Organization (WHO) recommended the Resolution on 'Rheumatic Fever and Rheumatic Heart Disease' for adoption. This landmark event represents the first global policy on RHD to be endorsed by all governments [17]. Reflecting a shifting recognition of its impact in maternal health, the resolution was situated in the context of maternal health and access to services [17].

Significantly, these initiatives and policy frameworks recognise the impact on pregnant women, and the imperative to situate RHD strategies within the context of maternal health [19]. As the principal heart disease seen in pregnant women in endemic regions, it is arguable that the targets for Sustainable Development Goal 3 of reducing the global maternal mortality ratio to <70/100 000 live births by 2030 cannot be met if RHD is not properly addressed for women.

Improved access to reproductive health services for women with RHD and other non-communicable diseases (NCD) is one of the seven key priority actions called for in a roadmap to eliminate RF and eradicate RHD in Africa [124]. Conversely, RHD has been included as one of 32 causes in risk-standardised mortality rates in the Universal Health Coverage Index as a marker of quality of health care [135].

Barriers to optimal care for women with RHD

The above description of the overall RHD landscape and its impact in pregnancy - with a particular lens on Australia - all highlight several challenges to provision of optimal care. While barriers can be broadly categorised – access to health services, geographical distance, jurisdictional divides, transfer of women, health workforce – they form a complex matrix of contributing factors that can impact directly and on each other. Further, underpinning each of these categories are the policies, governance, provision of care and practical decisions that shape the promotion – or hindering – of a culturally safe model of care.

Access to health services; workforce; tyranny of distance and jurisdictions

There is a lack of cardiac service resources including equipment and trained personnel for the diagnosis and management of RHD in remote areas [136,137]. This impacts on a number of levels. A 2007-2010 national study of RF in children through the Australian Paediatric Surveillance Unit (APSU) and led by the Menzies School of Health Research (MSHR) found several delayed referrals in remote areas across Australia that were due to lack of available specialist services [138].

Cardiac surgery is not available in the NT, so surgical intervention usually involves travel thousands of kilometres away from community. Calls to improve the health workforce capacity, availability of specialist care and systems for delivering that care are argued as essential to improve outcomes for RHD [139].

Whilst there are guidelines and some outreach capabilities, the ability to service the need according to recommended review schedules is limited. Portable echocardiographs enable women in remote communities to receive an individual risk assessment as early as possible in pregnancy (and ideally before conception). However, these are expensive, and cardiology outreach services to remote areas in Australia are very limited [137], thus the continuity of care prescribed by best practice [6] is often absent. There have been increasing calls for the specific targeting of appropriate health care Aboriginal peoples as a priority for RHD [140], including designated surgical units for Aboriginal patients with RHD [141], more Aboriginal cardiovascular health workers, better education and training for non-Aboriginal health workers and integrated collaborative research [140,142-144].

In addition to issues of clinical management and intervention, Australia is impacted by reduced capacity and capability of care and support for all pregnant women in remote regions [145].

There are major geographical barriers for women who live in remote areas of the country, with attendant difficulties (public transport, moving outside of community) in access to major health centres for specialist cardiac services, which are often hospital based. There are several flow-on effects of the impact of high transfer rates of women with RHD who may travel thousands of kilometres to give birth, raising issues of continuity of care. The direct and indirect costs include financial, family, community, and of course the often distressing experience of giving birth away from home in suboptimal conditions [146].

The high turnover of health staff in rural and remote regions hinders ongoing quality care. This lack of care continuity is compounded by mobility, where visits to other communities may impact on diagnosing/monitoring cases, availability of medical history and monitoring of warfarin therapy [137]. Other factors include the cost of medication for those who live in rural and urban centres, and insufficient access to specialist cardiology outreach services and echocardiography [136].

Health Information Systems

The ability of care providers to be able to store, share and retrieve relevant information related to the history of pregnant woman with RHD is integral to the provision of cohesive and integrated care. Yet the multiple cardiac/maternity care pathways are matched by information systems with varying degrees of integration according to jurisdiction and region. They are often fragmented, with a lack of capacity to share and are on multiple platforms (both electronic and non-electronic).

Kritharides et al talk about the impact of gaps in health information systems, where specific problems in health care delivery are not being documented and shared. Identification of these problems requires detailed information from each health service. These can range from availability of medication (i.e. resourcing), to lack of escalation of therapy where indicated by guidelines (i.e. medical decision/education), to personal choice (i.e. patient decision/education). They argue that information is best obtained when auto-populated electronically as part of health care service delivery, where it can also be used as a performance indicator for health care delivery [142]. Present fragmented health information systems – particularly in remote Australia - often do not permit this.

Issues of integrity related to the collection of perinatal data in Australia are widely recognised [147-152]. These include gaps in consistency across jurisdictions, lack of consensus on nationally consistent definitions for common maternal conditions, and lack of data on maternal characteristics and risk factors [151]. These issues are not particular to Australia [153,154], but tyrannies of distance, jurisdictional and policy divides exacerbate such challenges to continuity of care [155].

These gaps in health information systems impact on quality of surveillance data [156], with compromised data integrity contributing to under-reporting of RHD.

Health workforce

Pregnant women with RHD may be variously supported by and encounter an enormous range of health services during the course of their pregnancy, including Aboriginal health workers, midwives, obstetricians, cardiologists and cardiac nurses, echo technicians, remote area nurses and doctors, RHD Control Register staff – all of them potentially in different locations. Apart from the logistical issues outlined above, challenges to care for women may include lack of training or awareness among health staff and transient health professional staffing in remote areas [139]. These issues were emphasised particularly in the AMOSS study of women's journeys with RHD during pregnancy [20].

Successful models?

There are no known programs that have specifically addressed needs of pregnant women with RHD in Australia.

The fundamental drivers of successful cardiac care programs have incorporated engagement, recovery interventions, capacity building and self-governance. There are small numbers of programs which have addressed these, such as the provision of outreach cardiac health care services directly to rural and remote communities across Queensland [157]. The cardiac nurse co-ordinator is the primary resource for medical and nursing staff at local, intra/interstate hospital, remote centres and GP's at a Central Australia based program which establishes appropriate follow up after investigations with a care plan in place [158]. Other initiatives are informed by building relationships and widespread community involvement particularly with Aboriginal health workers [159-161].

The last ten years has seen a number of maternity care initiatives that attempt to provide better models of care for Indigenous women, particularly in remote and regional Australia.

The Mums and Bubs program (QLD), Aboriginal Maternal Infant Care program AMIC (SA), Aboriginal Maternal Infant Health Service AMIHS (NSW) and Midwifery Group Practices (MGP) in NT[1] all have worked to improve uptake and quality of antenatal care. Successful programs incorporate an Aboriginal workforce (doctors, midwives, health workers, allied health professionals) that is integral to the maternity services team, including support for training positions [146].

A twelve month review of one MGP in the NT found increased engagement with women who give birth, with improved access to maternity care [162]. This study showed that there is no magic bullet in the effective addressing of complex issues related to maternity services *("It's not a perfect system but it's changing")*. However, it highlighted some of the small yet meaningful steps that are being taken to instigate profound change and better outcomes. These initiatives provide a useful base to explore what could work to provide improved and integrated care for pregnant women with RHD.

Cultural safety

The specific socio-economic determinants that impact on Indigenous women demand more than simply a clinical response to management [146,163].

There is indisputable recognition that good effective health care cannot be delivered without containing the principles of cultural safety [146,155,164-170]. A wholistic approach with continuity of care have been identified as important determinants of adhering to secondary prophylaxis for RF and RHD [171] and this is likely to apply to anticoagulant regimens for women with mechanical valves [172]. Lawrence stresses the imperative to generate mechanisms to address cultural impediments to uptake of Western medical care [139].

[1] Aboriginal Mother and Baby (AMB) health services are Australian jurisdiction-based programs where midwives and Aboriginal health practitioners/workers partner to provide culturally-appropriate support and care throughout pregnancy. They are known by various names across states: NSW Aboriginal Maternal Infant Health Services (AMIHS); SA Aboriginal Maternal and Infant Care (AMIC); NT Midwifery Group Practice; QLD Mums and Bubs and Birthing in Our Community Service. In this thesis, the services are generically referred to as Aboriginal mothers and baby (AMB) services.

There is often tension between providing appropriate medical care for pregnant women with RHD and the cultural, personal and community disruption of birthing away from Country. Nappaljari Jone talks of this disruption at a collective level contributing to breakdown of community, and consequent impact on risk factors for women in pregnancy and labour [173]. Several papers refer to disruption and negative impact of evacuation from community to tertiary centre. Continuity of caregiver, involvement in decision making, and choices related to presence of partners, family, and social support: all impact on the birthing experience and are often limited in these settings [155,162,173-177].

There are vexed questions about the provision of necessary medical care for women with RHD - who are at higher risk of other co-morbidities - but careful clinical assessment and continuity of care should underpin any maternity services [146]. Kruske refers to improved perinatal outcomes in maternity services that are used by significant numbers of other Aboriginal women as an argument for the imperative of Aboriginal-specific maternity services [146].

Arnold et al suggest that, while aiming for 36 weeks' gestation for transfer is medically appropriate for women travelling from remote Far North Queensland to Cairns Base, the long periods of separation of women from family and friends have detrimental social, cultural and financial consequences. They call for the reopening of maternity units in towns serving the Cape communities in order to reduce the number of women from the region having to travel to Cairns for pregnancy care and birth [176].

Several studies of birthing practices in Inuit remote communities have demonstrated good outcomes for mother and baby [174,178-181] who birth in community rather than transfer, where local birthing services are supported by effective expertise, good clinical judgement and health resources. Developed in response to criticisms of the policy of evacuating women from the region in order to give birth in hospitals in southern Canada, the midwifery service is integrally linked to community development, cultural revival, and healing from the impacts of colonisation [182]. Of note is the broader conceptualisation of perinatal risk that guides principles of safe care in this remote setting. Risk screening is seen as a social, cultural, and community process in addition to biomedical factors. The Nunavik initiative recognises – in accordance with the Ottawa Charter [183] - that health is regarded as more than the absence of disease, and includes that of the individual's physical, mental,

emotional, and spiritual aspects, in addition to health in the family and the community as a whole [182].

The above historical perspective of women with RHD and public health responses to RHD with a focus on the Australian context illustrates some of the themes and issues central to care pathways for women with RHD-P: diagnosis, pregnancy planning, risk assessment, often-complex care needs, continuity of care, access to woman-centred integrated health services, and information systems. These physical and structural imperatives are against a backdrop of often-resource challenged environments that test optimal care and outcomes for mother and baby.

Guidelines and reviews

The second part of this literature review examines guidelines with relevance to models of care for pregnant women with RHD; and extrapolates core principles of care against which selected studies including pregnant women with RHD are assessed.

Models of care informed by evidence-based clinical guidelines are essential to optimal health outcomes[184,185]. There is a lack of evidence base of the effect of different models of care and strategies on the course of pregnancy complicated by RHD and associated best practice to optimise outcomes [6,186]. Guidelines addressing RHD-P are mostly based on case series and observational studies, often part of broader studies of all-valvular or all-cardiac disease. While many recommendations of clinical guidelines for pregnant women with valvular disease or all-cardiac disease are relevant for women with RHD, the epidemiology of this disease and associated social determinants requires examining other factors that may impact on care and outcomes.

This section answers research question 1:

1. What guidelines and reviews inform or have relevance for models of care for pregnant women with RHD?

Methods

A structured search of 13 databases was conducted. Standards of quality care were identified for the care of women with RHD-P in English language guidelines and reviews between 1995 and 2018.

Search protocol

The search strategy incorporated a combination of free term text items and Medical Subject Headings (MeSH) of the following:

("rheumatic heart" OR "rheumatic fever")

AND

("pregnancy" OR "pregnancy complications" OR "pregnancy, high-risk" OR "pregnancy complications, cardiovascular" "maternal" OR

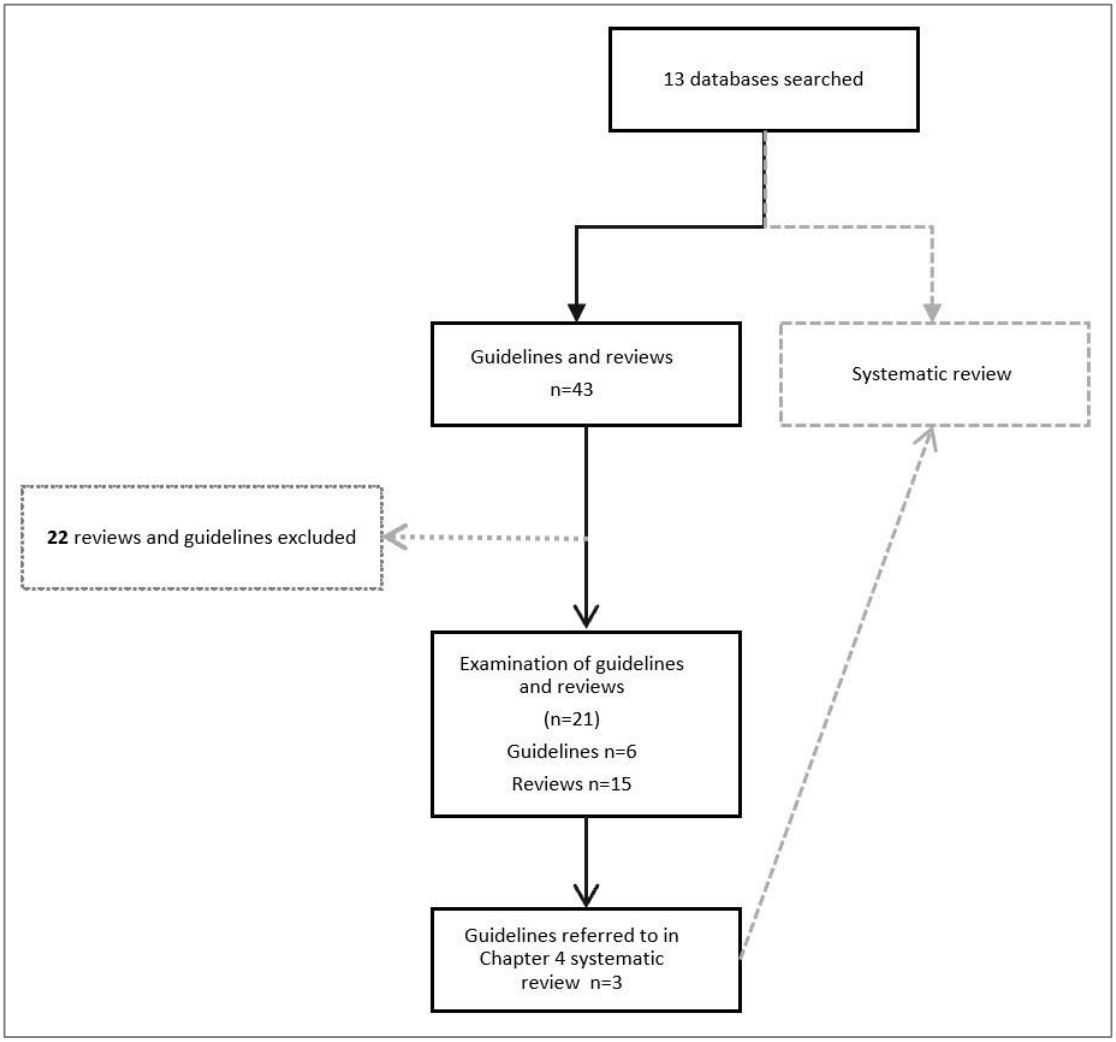
AND

("models of care" OR "guideline*" OR "health service" OR "maternal health services" OR "Primary health care" OR "practice guideline" OR "guideline adherence" OR "Health services accessibility" OR "health care")

Primary searches were conducted on PubMed, Medline, EMBASE, CINAHL, Nursing and Allied Health Database, Cochrane Library, Aboriginal and Torres Strait Islander Health (ATSIhealth), Indigenous Collection (Informit), Rural and Remote Health Database (Informit), ETG Complete, ISI Web of Science All Databases (ISI), PLoS (Public Library of Science), Trip Pro Database.

The search for Grey literature included Clinical Practice Guidelines and reports. Guidelines and reviews were included if they referenced maternal care and RHD or all-cardiac disease.

Figure 2-1: Search methods: Guidelines/Standards and Reviews



Findings

Forty-three reviews and guidelines were identified as having relevance to broad principles of care relevant for pregnant women with RHD. Twenty-two of these were subsequently excluded due to lack of specificity to RHD and/or maternal health, leaving six guidelines/standards and 15 reviews (see Figure 2-1).

Thirteen of the guidelines/reviews (indicated with asterisk in Appendix 2 table) substantially addressed RHD - defined as containing a minimum chapter or section specifically on RHD - as well as pregnancy and maternal health. Others had some reference to RHD but were within a clinical context that focused on valvular heart disease or all-cardiac disease in pregnancy. Two reviews did not refer to RHD explicitly but were included because the clinical features discussed were strongly associated with the condition, and recommended care standards were relevant [187,188].

Clinical practice guidelines are defined as *'statements that include recommendations intended to optimize patient care that are informed by a systematic review of evidence and an assessment of the benefits and harms of alternative care options'* [184,189], providing *'well-balanced information reflecting established evidence-based knowledge on a specific subject and systematically developed recommendations for diagnosis and treatment for practitioners'* [190]. Clinical care standards are defined as … *' a small number of quality statements that describe the care patients should be offered by health professionals and health services for a specific clinical condition or defined clinical pathway in line with current best evidence'* [191]. However, in this study, a broader definition of standards is used (adapted from the ESSENCE essential service standards) which … *outline elements of care and service delivery that should be accessible to all [women with RHD] regardless of their location, ethnicity, economic circumstances or gender. The standards reflect a strong mix of service activities, evidence based clinical care, quality of care and process of care indicators, alongside systematic approaches to improve the delivery of these standards to all people in need* [192].

Guidelines and Standards for RHD-P referred to in this study include the 1) American Heart Association(AHA)/American College of Cardiology (ACC) for valvular heart disease, with a section reference to pregnancy [193] and a brief reference in the context of anticoagulation in

its 2017 update [194]; 2) Australian RHD Guidelines [62] and 3) New Zealand RHD Guidelines [116]; 4) Essential Service Standards for Equitable National Cardiovascular Care for Aboriginal and Torres Strait Islander People (ESSENCE) [192]; 5) European Society of Cardiology (ESC) Guidelines on the management of cardiovascular diseases during pregnancy [196]; 6) RCPSG Standards of good clinical practice in the shared obstetric and cardiology care of women of childbearing age [187]; the 7) RCOG Cardiac Disease and Pregnancy Good Practice No.13 [197].

Some professional Colleges provide direct reference to specific RHD-P guidelines: for instance, the Royal Australia and New Zealand College of Obstetrics and Gynaecology lists a direct link under its Guidelines and Statements section to the RF/RHD Australian Guidelines [198].

Key attributes of quality care relevant in the care of women with RHD-P were identified from the guidelines/reviews and categorised into themes. These included care priorities and standards identified in RHD and reproductive health care during the life-course, with overarching principles of care to underpin systems as adapted from the ESSENCE standards [192]. Some themes were repeated, reflecting the varying impact, consequences and care needs of RHD according to life-course and degree of severity.

Attributes of quality care: themes

1. Over-arching principles of care
2. Diagnosis of RHD
3. Preconception care and planning
4. Antenatal care
5. Labour and birth
6. Postpartum and interpregnancy care

Aspects of care within each theme are defined below.

[2] The 3rd Edition of the Australian Guideline currently under development has a substantially revised and extended section on Women with RHD to incorporate a whole-of-life approach, including: a new section on transition to adult care, reproductive health, and preconception care; an updated discussion around the need and strategies for a well-planned pregnancy and delivery; an updated discussion about anticoagulation during pregnancy; and a more detailed section on other medications. This new chapter, along with the rest of the Guideline, has been shaped and informed by a review that emphasises respect and cultural safety (195.Noonan S, Wade V. Integrating Cultural Safety Into Disease Management Guidelines. *Heart, Lung and Circulation.* 2019;28:S48-S49.

Attributes of quality care: sub-themes

1. Over-arching principles of care

1.1. Primordial prevention

Addressing the 'causes of the causes', comprehensive primary health care articulating with maternity health

1.2. Primary prevention

Identify and manage risk; awareness and education of workforce (particularly maternity) and community in high risk areas; impact of RHD for women of child-bearing ages

1.3. Secondary prevention

Education, multidisciplinary care, secondary prophylaxis; dental care; reproductive health and management

1.4. Health systems and access to services

Articulation of RHD control programs with maternity services [199]

1.5. Woman-centred, culturally safe care

Recognising heterogeneity of communities, integrated with both remote and tertiary services [146,200,201]

An Australian project by Brown et al defines a set of standards developed through a consensus process which include a series of foundational **service elements** with relevance to the prevention, management and mitigation of cardiovascular conditions (including RHD) for Aboriginal and Torres Strait Islander peoples. These overarching principles have a particular focus on reducing population levels of risk factors including targeting the social determinants of health and improving primary health care frameworks [192].

The **epidemiological and regional variances** of a disease that has its genesis in deprivation were not addressed in several guidelines/reviews, including reviews in journals with a world-wide focus [12,188,202].

Yet, apart from burden of disease, describing such underpinnings matter: access to simple preventive measures such as secondary prophylaxis may be constrained by availability [203] in

resource-challenged environments, let alone access to tertiary level review and high-level services including surgery and other interventions [203-206].

2. Timing and method of diagnosis of RHD

2.1. Timing

The **timing of RHD diagnosis** – whether pre-pregnancy, antenatal or postpartum – has a significant bearing on perinatal and maternal outcomes, with escalated risk after 20 weeks' gestation and post birth. The diagnosis of RHD in late pregnancy or postpartum is generally associated with severe complications including cardiac decompensation.

2.2. Method

The **method of diagnosis of RHD** is most accurately determined using standard guidelines developed for echocardiographic diagnosis, based on which heart valves are involved and the type and severity of damage to each valve [73].

3. Preconception care (PCC) and planning

3.1. General principles, evaluation, risk assessment

3.2. Contraception

3.3. Prosthetic heart valves, anticoagulation and other high-risk factors

3.4. Transition to adult cardiac care

The World Health Organisation (WHO) supports pre-conception care (PCC) towards improving health and pregnancy outcomes. PPC entails **a continuum of promotive, preventative and curative health and social interventions**, and identifies current and potential medical problems of women of child-bearing age towards strategising optimal pregnancy outcomes [207]. This should be underpinned by **discussion with women and families** in order to develop a shared understanding of risk, planning and decisions.

Pregnancy planning and preconception care

Ideally, women with known RHD would be properly assessed before pregnancy occurs. If they are already symptomatic due to significant RHD, serious consideration should be given to intervention prior to pregnancy. In patients with moderate or severe mitral stenosis, percutaneous balloon mitral valvuloplasty (PBMV) should be considered, because of the high risk of maternal and fetal complications during pregnancy.

Early (pre-conception) echocardiographic assessment is particularly important for pregnant women, in order to diagnose and grade the severity of RHD and attendant pathologies, and in turn to inform effective care [65,202,208-211].

Women with mechanical valves who are on warfarin should be given appropriate contraceptive advice and counselled about the risks to mother and fetus with pregnancy [212,213]. Moreover, there are significant issues of quality of clinical management - and, by implication, equity - where young women of child-bearing age are given mechanical valves as opposed to other surgical options. These clinical dilemmas are compounded by the increased rate of re-surgery required of bioprosthetic vs mechanical replacement.

Care when describing the consequences of RHD and pregnancy is required. Many women who have effective multidisciplinary care have reasonable or good perinatal outcomes even in low-resource environments [214]. Descriptions that imply the consequences of RHD-P are inevitably devastating can be misleading to clinicians - and frightening for women [199].

The need for **a formal, individualised step-wise approach** to prepare young patients for successful transfer to an adult healthcare is well-documented for congenital heart disease [215-217], but was documented poorly in the guidelines and reviews examined for RHD.

Sliwa et al present algorithms for heart valve surgery for aortic and mitral lesions in women of child-bearing age which they suggest is relevant in high-income and low-and-middle income countries, where the available expertise required for surgery or valvuloplasty is factored in [218].

The AHA/ACC guidelines state that 'All patients referred for a valve operation before pregnancy should receive pre-pregnancy counselling by a cardiologist with expertise in managing patients with valvular heart disease during pregnancy about the risks and benefits of all options for operative interventions, including mechanical prosthesis, bioprostheses and valve repair' [193]. The report from the 2016 UK maternal deaths comments that, whilst this recommendation is helpful, an obstetrician should also be involved in the consultation [219].

4. Antenatal care

4.1. General principles: multi-disciplinary, women-centred care

4.2. Review and risk assessment; reference to Guidelines/Standards; access to services; co-morbidities

4.2.1. Risk assessment

4.2.2. Care for women assessed as low-moderate risk

4.2.3. Care for women assessed as high risk: discussion of surgery, PBMV, other interventions, and care for women with existing prosthetic heart valves

4.2.4. Medication: discussion of general principles, teratogenicity, anticoagulants, other medications

Management (and diagnosis) of RHD in pregnancy

RF and RHD have a defined clinical progression: diagnosis and management at each stage can help avoid progression to the next more serious phase. However, often-limited cardiac services due to under-resourced health services and/or remote locations combine with lack of reporting of symptoms, communication difficulties and cultural barriers that may mask even advanced valvular disease [137]. Zollner et al comment that, while women with congenital heart disease are usually aware of their diagnosis, this is often not the case in acquired heart disease such as RHD, although many have significant cardiovascular risk factors [209].

Subclinical RHD may thus become apparent for the first time during pregnancy, particularly during the second trimester [6,209,220].

It is crucial to identify women at high risk early in pregnancy to offer multidisciplinary care if indicated and to ensure that these women are investigated appropriately when they present with cardiac symptoms [210].

In low-middle income countries, RHD jostles with other more prevalent conditions such as tuberculosis, malaria and (particularly in South Africa) AIDS [221,222].

There are a number of issues specific to women with RHD-P [65,208,209]. Increased cardiac demands, treatment interventions such as mechanical valves and anticoagulant regimens [223], and increased co-morbidities all combine to produce a higher risk of morbidity for mother and fetus [6,224]. The overall scarcity of evidence-based research on RHD-P results in an associated lack of coherent management [14,141].

The gap in evidence-based research has contributed to a lack of international consensus on preferred model(s) of care for pregnant women with RHD [6,117,225]. Apart from isolated references to specific features of RHD-P [12,194,202,226] key published studies on the impact of all cardiac diseases on pregnancy emphasise the importance of pre-conception proper assessment and counselling before pregnancy occurs, with early echocardiographic assessment and individual risk assessment antenatally to diagnose severity and develop a management plan [6,13,65,82,208,222,227-237]. Siu derives a clinically useful risk index for mother and fetus based on key clinical parameters [212,236] and anticoagulant treatments are discussed [212,223,233,234]. The risk index developed by Siu et al has been cited as prognostic factors in lesion-specific studies [212].

An Australian paper on management of obstetric patients with RHD profiles two case histories and emphasises that underlying RHD should be considered in pregnant Indigenous women from remote regions[238], and a subsequent retrospective study by the same author discussed these clinical management issues in more detail, adapting the Siu index for the Australian context [239].

However, combined factors in Australia such as limited cardiac services in remote areas, high turnover of health staff, and delayed attendance at clinics due a lack of trust and cultural safety may result in even advanced valvular disease being masked [136,137]. There have been increasing calls for the specific targeting of appropriate health care for Aboriginal peoples as a priority for RHD [140], including designated surgical units for Aboriginal patients with RHD [141], more Aboriginal cardiovascular health workers, better education and training for non-Aboriginal health workers and collaborative research [140,143].

There is no single **risk score** for valvular heart disease. Risk assessment models to estimate maternal cardiac and offspring risk associated with pregnancy in women with cardiovascular disease include the NYHA functional classification [31], World Health Organisation (WHO), CARPREG (CARdiac disease in PREGnancy) from a large single high-income country cohort [212], ESC modified WHO classification (mWHO) [196,240] which is based mainly on expert consensus [241] and ZAHARA (Zwangerschap bij Aangeboren HARtAfwijkingen, pregnancy in congenital heart disease) [242]. A Registry Of Pregnancy And Cardiac disease (ROPAC) evaluation of the modified (mWHO) risk classification suggested that it is a useful tool for predicting cardiac events during pregnancy in women with established cardiac disease in advanced countries, but less effective in emerging countries

[241]. Hameed [217] and Sliwa [243] present algorithms to guide stratification and initial evaluation of symptomatic or high-risk pregnant or postpartum women. The 3rd Edition of the Australian RHD Guideline (under development) provides an adapted risk algorithm to guide care pathways [1].

The need for **collaborative multidisciplinary care** was emphasised in most guidelines/reviews [187] but was generally limited to cardiac-obstetric reference, with some also calling for anaesthetic/maternal fetal/neonatologist specialist care [205]. Few studies referred to disciplines such as midwifery/nursing, or those outside tertiary care settings (such as primary health services, community midwifery/nursing or general practitioners, Aboriginal health practitioners, specialised nursing staff and allied health services according to country and region) [192]. Apart from multidisciplinary care, a strengthened health workforce is identified as a critical component in delivering optimal care. Human resource deficits have been identified as one of the eight roadblocks in successfully addressing RHD [199].

The **impact of co-morbidities** such as obesity, diabetes, HIV/AIDs and chronic kidney disease vary according to country, political and economic factors. In South Africa, 32% of 2014-16 maternal deaths from cardiac disease were in HIV positive women (where status was known) [244], consistent with an earlier single site study of cardiac disease in pregnancy that found 33% of women to be HIV positive with RHD being the most common aetiology (81%) [245]. The prevalence of chronic kidney disease (CKD) in pregnancy is rising [246]; in Australia, end stage kidney disease is over four times more prevalent among Indigenous 30-39 year old women [247]. Generally speaking, the social determinants underlying RHD are the same as for many other morbidities that will potentially (and often exponentially) increase risk in pregnancy [219,248-251], again highlighting the need for collaborative care with relevant specialists.

Secondary prophylaxis regimens – safe in pregnancy where prescribed – were referred to in four of the six guidelines.

5. Labour, birth and early post-partum

5.1. Mode of birth – general principles

5.2. High risk; anticoagulation, mechanical heart valves

5.3. Antibiotic prophylaxis, sepsis, endocarditis prevention

5.4. Lactation and medication

Labour and birth

Labour and birth result in a further increase in cardiac output, which such factors as type of valvular lesion, method of delivery and maternal position being associated with significant cardiovascular changes. Vaginal delivery is recommended with a few exceptions: caesarean section increases the risk of haemorrhage, postpartum infection, pulmonary co-morbidity, and puerperal fluid shifts and metabolic demands [6].

Surgery

Recommended interventions vary according to severity of the disease and pathology, and include valve repairs, and mechanical and bioprosthetic valves, all of which have associated risks for mother and fetus [6,137]. Women of childbearing age who require surgical intervention should have valve repair as a first preference, with bioprosthetic valves if repair is not possible [6].

The often-delayed diagnosis of RHD impacts on management: more conservative interventions of valve repair or valvuloplasty are less suitable for advanced valve disease, and consequently replacements are required [137]. Bio prostheses are not as durable as mechanical prostheses, but women with mechanical valves have a higher rate of thromboembolism and 10-year mortality than those with bio prosthetic valves [234].

Cardiac surgery during pregnancy is associated with a high-risk of fetal loss (~20%) and morbidity, including late developmental delays in the child, and should be avoided unless indicated by severe maternal cardiac valvular disease that cannot be managed by medical therapy alone. The decision must be made on an individual basis in consultation with relevant specialists in centres with expertise. Beyond 28 weeks, delivery before surgery should be considered [196,252]

Anticoagulation in pregnancy

The surgery chosen and consequent anticoagulation regimens have a significant impact on adolescents and women of child-bearing age.

A mechanical heart valve (MHV) replacement for RHD requires lifelong anticoagulation to prevent valve thrombosis and thromboembolism but the choice of anticoagulant during pregnancy is controversial [6,213,220,225,234,253-256]. This creates clinical and personal dilemmas for women requiring anticoagulation (whether for MHV or other indications such as atrial

fibrillation) and the clinicians caring for them. Vitamin K antagonists (VKA) such as warfarin, give clear maternal benefit with lower thromboembolic events [196,257] compared to low molecular weight heparin (LMWH) and is favoured in the ESC Guideline [196,254]. However, VKAs cross the placenta and are teratogenic, causing warfarin embryopathy and high rates of late fetal loss due to fetal intracranial haemorrhage. On the other hand, LMWH, whilst safer for the fetus, is a less effective anticoagulant [254] and may not prevent maternal thrombotic complications [6].

Women require specialist care and monitoring preceding and during pregnancy to address often vexed decisions surrounding anticoagulant risk; however there are inconsistent approaches to management [254], with clinicians often having a lack of awareness of the complexities and high risk associated with anticoagulant use in pregnancy [254,258].

Women may know that VKAs can have adverse effects in their unborn child and avoid it through pregnancy. This poses the highest risk to a woman and her infant.

The ESSENCE standards call for access to routine and appropriate monitoring of anticoagulation as close to home as possible, including point-of-care monitoring for patients in rural and remote settings [192].

Lactation and medications. While breast-feeding is generally recommended for women (including for women on anticoagulation [6] and those with other co-morbidities such HIV-positive/AIDS) [259], cardiac medications may contraindicate this [260]. The ESC Guideline [240] outlines general principles, summarising pertinent drugs and their potential use during pregnancy and breastfeeding.

6. Postpartum and interpregnancy care

6.1. Short-term, long-term care pathways

Postpartum

Pre-discharge conception planning

Discussion with women regarding cardiovascular health, future pregnancy risk and inter-pregnancy planning should take place before discharge in high-risk women and followed up in the primary health setting. A shared understanding about risk and preference promotes informed decisions about pregnancy planning and contraception. The date confirmed for the woman's next secondary prophylaxis treatment (if applicable) should be checked.

Where a mother has RHD, her children will often have an increased risk of rheumatic fever/RHD.

Post-discharge

A vital aspect of preconception care is the post-partum and inter-pregnancy periods. High-risk women should be referred to tertiary care centres with required expertise [196].

Early involvement of primary care services is crucial to ensure a smooth transition post-partum [217]. Information about the woman's treatment, medication, future management plans and conception planning should reach her specialist and primary care provider(s) and referring hospital (where relevant) within 48 hours of discharge [192]. This includes clear information regarding RHD diagnosis, treatments and interventions, pregnancy and birth, routine recall plans and specific information about non-routine care requirements. All women without a designated GP or primary care provider should be integrated into a community program for home- or centre-based therapy and education following hospital discharge and be assisted to access appropriate primary care services [192].

The need for **improved postnatal care** was emphasised in UK and South Africa maternal deaths reports [219,244] and other reviews [192,217]. Gaps in postnatal care, in particular too-early discharge of and lack of follow-up of patients were found to be a major problem in the most recent South African maternal deaths report: approximately 20% of maternal deaths occurred outside of healthcare facilities, with causes similar to the causes of maternal deaths in facilities [244]. In South Africa, more than 36% of maternal deaths occurred in the postpartum period (2014-16) and a …*"considerable proportion of women who died were discharged home after delivery with abnormal vital signs. … Assessors believed that 226 (48.4%) of deaths were potentially preventable" [244].* Of note in this report, 165 of 467 maternal deaths in the 'medical and surgical disease in pregnancy' (35%) were due to all-cardiac disease. The breakdown of cardiac disease was not specified for these deaths - nor was RHD referred to in the 2018 Report - but earlier Reports note that the significant number of cardiac cases due to rheumatic heart disease [261,262]. The 2016 UK Maternal Deaths report with focus on cardiac deaths 2009-14 noted that, as well as the 153 deaths reviewed, there were 36 late cardiac deaths identified from the records of the Office for National Statistics (ONS) and National Records of Scotland (NRS) for which no information was available [219].

Sliwa et al call for improved clinician training, joint obstetric-medical-cardiac clinics with earlier referral pathways to avoid the persistent (and growing) numbers of women with late maternal death due to cardiovascular disease [243].

Brown et al detail required features of post-discharge clinical communication and handover including expeditious sharing of information about birth, treatment, medication, any complications and future management plans with the woman's primary care provider(s) such as early child and maternal care, and referring hospital. These clinical communications should include clear information regarding the woman's cardiac status, routine recall plans and specific information about non-routine care requirements [192].

Hameed emphasises the need for primary care, Emergency Department providers, and obstetricians to maintain a high index of suspicion for underlying cardiovascular disease when a woman presents with symptoms, signs, and risk factors concerning for heart disease for as long five months postpartum [211].

Frameworks of care: domains

In reviewing the literature, it became evident that these models of care were often (and necessarily) located within other frameworks that gave a critical context for women with RHD-P. Thus, themes were further categorised according to which of four contextual domains relevant to care of women with RHD-P were addressed (and according to whether primary or not). The primary category comprised domains A and B in all included guidelines and reviews. Domains C and D were potentially listed in the primary or 'other' category according to focus.

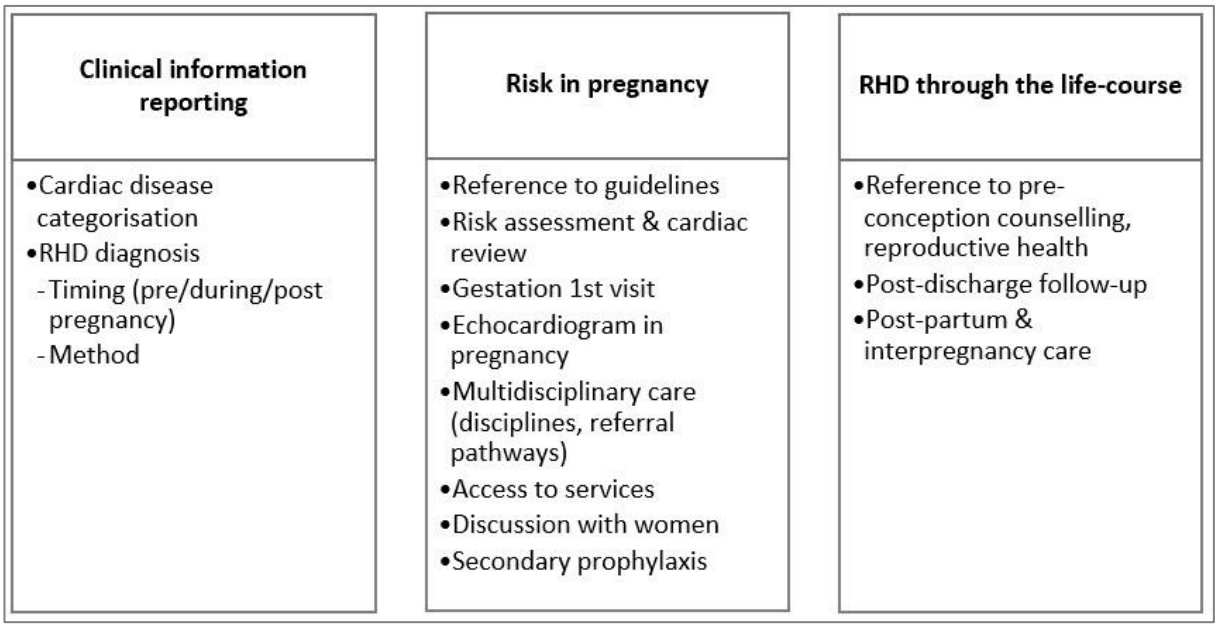
A. RHD or all-cardiac care framework: reviews and guidelines that define standards of cardiac care and include specific reference to RHD.
B. Pregnancy and maternal health framework: reviews and guidelines that define standards of care within a reproductive health context, with reference to RHD.
C. Indigenous health and vulnerable populations: reviews and guidelines that address RHD in the context of Indigenous health and vulnerable populations.
D. Health systems: reviews and guidelines that situate RHD within a health systems context, addressing structure, access, politics, policy and social determinants in an over-arching framework that specifies models of care relevant for women with RHD.

The extrapolated key attributes of care and the contextual domains from the guidelines and reviews are listed in the table in Appendix 2.

Core attributes of care for women with RHD: a framework of reporting measures

From the list of attributes of quality care outlined above, a subset of core attributes was identified for inclusion in the Chapter 4 content analysis of studies that referenced women with RHD (see Figure 2-2 below and explanation):

Figure 2-2: Framework of reporting measures



- Categorisation of cardiac disease: (specifying number of women with RHD in all-cardiac studies or studies of specific lesions such as mitral stenosis)
- Diagnosis: timing (breakdown according to whether diagnosed pre-pregnancy, antepartum, postpartum); method (clinical assessment; echocardiogram)
- Reference to Guidelines: (statements and recommendations based on independent review of the available evidence by professional societies such as American Heart Association or European Society of Cardiology, RHD-specific including pregnancy such as the Australian RF/RHD guideline, or hospital-specific guideline on RHD or all-cardiac care in pregnancy.)
- Antenatal risk assessment and reviews: (reference to risk assessment and risk score; frequency and scope of review)
- Discussion of access to surgery and other interventions
- Reference to anticoagulation protocol (and reference to discussion with women)

- Discussion of disciplines involved: (multi-disciplinary care cardiac-obstetric; other disciplines)
- Conception counselling: a) conception planning, access to reproductive health services and risk (medications, surgical choices) discussed; and b) reference to discussion with women and family in the study
- Postpartum and interpregnancy care, continuum of care

Discussion

Section one of this literature review describes the history of the burden of RHD/RHD-P over the last 150 years and its shifting epidemiological profile. It provides an interesting counterpoint to addressing this disease of inequity in the twenty-first century. So, too, do the descriptions of evolving management and preventative strategies during that time.

The initiatives and programs developed in the 1920-30's on both sides of the Pacific that so dramatically reduced (mostly RHD) cardiac maternal mortality [33,37,39] were based on a collaborative approach with joint cardiac-obstetric care. These and other historical writings on RHD-P on risk (particularly stratification according to NYHA) provide context and an important framework of reference [25] for present day strategies related to clinical care, policies, education and advocacy. Sadly, the lessons from a century ago have not been universally adopted, and the call for multi-disciplinary care has even more relevance today, with models including midwifery, primary and Indigenous health as well as specialist medical and surgical care.

As relevant, too, are the underlying 'causes of the causes' [122] that are so definitive in precipitating each preventable stage of the RF/RHD disease trajectory. Its shifting geographic and demographic burden in turn reflects nation (and populations-within-nations) health and housing deficits.

Yet, in contrast to the late nineteenth century, the clinical and epidemiological evidence exists to inform appropriate care for people living with RHD and to prevent new cases of RHD [68]. We know that the cost of effective treatment of RF (including its underlying causes) is miniscule when compared to emergency-driven clinical care of RF and RHD which can have such devastating impact in pregnancy. We know that RHD effectively becomes an inter-generational disease, with the disease cycle repeating across generations. We also know that RHD can be controlled.

The Guideline review in the second section of this Chapter identified core attributes of quality care relevant for women with RHD-P were identified and categorised into themes.

Core outcome sets (COS) are minimum collections of outcomes with standardised measurement and reporting. They represent a minimum data set of outcomes prioritised by stakeholders, including healthcare professionals, researchers, and patients [263]. Since 2014, when the Core Outcomes in Women's Health Initiative began, it has been adopted and further developed in over 50 journals [263-268]. This initiative has a number of aims, which include encouraging researchers to develop COS in the field of women's health, organising robust peer review and facilitating effective dissemination of manuscripts [267].

The last decade has seen a refinement and strengthening of Guidelines with relevance to women with RHD. The 2018 European Society of Cardiology has enforced the mWHO classification of maternal risk, refined anticoagulation protocols, strengthened recommendations of tertiary cardiac-maternity care for high-risk women and introduced the concept of a pregnancy heart team, and introduced advice on contraception and termination of pregnancy in women with cardiac disease [196]. A revised CARPREG risk score highlights late first antenatal visit (>20 weeks) as an independent risk factor [269].

However, while many of the general and specific principles of care apply to women with RHD, there are no Guidelines that specifically address this group of women, who often require additional aspects of care to be addressed in the context of the socio-economic determinants that underpin RHD. Defining core principles of care within a life-course approach to health required extending the review beyond clinical guidelines to include other guidelines and reviews that addressed determinants of health, risk and outcomes for women with RHD. This was conducted within a framework of four intersecting and contextual domains: RHD or all-cardiac care; pregnancy and maternal health: Indigenous health and vulnerable populations; and health systems.

Conclusion

The literature and guideline reviews in this chapter define and address key principles of care for pregnant women with RHD.

The broad themes that have emerged are explored in more detail in the ensuing studies, which will take a more granular approach to exploring barriers to delivery of optimal care for women with RHD-P within health services.

Chapter 3 Research methods

This chapter describes the underlying methodology that informed and shaped the studies in this thesis. It also details the methods of the qualitative study 2 including information on data analysis and ethics approval.

Introduction

The overarching aim of this doctoral research was to investigate the factors that impact on optimal care for pregnant women with RHD, with a focus on health services.

The study sought to answer the following research questions. Three studies were conducted to support this aim and answer the research questions.

1. What guidelines and reviews inform or have relevance for models of care for pregnant women with RHD? (Literature review and Study 1)
2. How are attributes of care for women with RHD-P reported in the literature and how do these align with outcomes described in evidence-based guidelines? (Study 1)
3. What levels of knowledge, expertise and awareness exist amongst health professionals regarding care pathways for pregnant women with RHD and its burden? (Studies 2, 3)
4. What do health professionals perceive to be the barriers and facilitators to the provision of optimal health care for women with RHD-P? (Study 2)
5. What policies and strategies do health professionals suggest are required to more effectively meet the needs of these women? (Study 2)

Methodology

An overarching mixed methods approach was established as a methodological framework for the research in the course of an iterative process. The research takes a transformative lens [270].

Mixed methods research that is reflective of the transformative paradigm is … *"identified by adherence to a social justice agenda; explicit acknowledgment of factors that are culturally based in the definition of what is perceived to be real; recognition and challenging of power differences in relationships in the research context and wider society; and the need to develop methodological approaches that are responsive to the aforementioned complexities. A transformative model for mixed methods research suggests the need for community involvement, as well as the cyclical use of data to inform decisions for next*

steps, whether those steps related to additional research or to program changes" (Mertens, 2007, 2009 in [270].

The community in this instance were health service providers for women with RHD. My research journey was threaded with conversations – deliberative, serendipitous and opportunistic – in addition to the formal interviews in Study two. These were two-way interactions: conversations served an educative function for health services providers (predominantly in maternity units) to learn more about aspects of care for women with RHD, and I learnt about issues that both directly and indirectly impacted on care pathways for women with RHD. In the first of several workshops with an Aboriginal Mothers and Babies program where it had previously been stated there was little RHD, a young Aboriginal health worker commented quietly afterwards 'Yeh, I've got that RHD' and a senior colleague described her childhood having secondary prophylaxis injections. That group then advised on the content and messages in promotional material that was co-developed as part of the AMOSS RHD-P project and over subsequent years became strong ambassadors for raising RF/RHD awareness among other colleagues.

In-service sessions under the auspices of the AMOSS project had a formal educative purpose, but also served to bolster collaborative partnerships: linking midwives with local RHD programs in one high prevalence region, and an obstetrician with the cardiac care nurse in another. These linkages had an immediately practical benefit in helping support better notifications for the AMOSS study. It was invaluable in my research journey both in learning of the importance of forging collaborative structures and hearing many stories of the 'what works, what doesn't' from a broad range of health service providers.

Similarly, ongoing conversations with a large proportion of 250+ AMOSS data collectors active during that quantitative arm of that study helped shape research questions and the direction of my research.

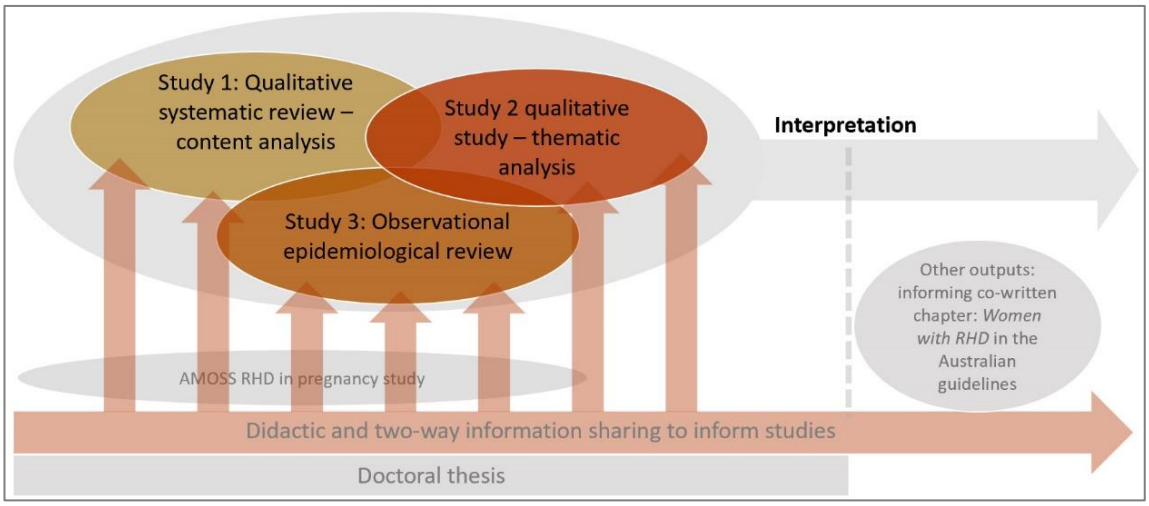
This process of engaging with professional communities helped elicit themes and questions that were explored in more detail in the qualitative Study two and informed Study three.

The work was thus defined as a transformative parallel design. This involves the use of both quantitative and qualitative methods at essentially the same time during the study.

There were elements of both fixed and emergent aspects to the design. Emergent mixed methods designs generally occur when a second approach (quantitative or qualitative) is

added after the study is underway because one method is found to be inadequate [271]. My original doctoral research proposal included a systematic review of the literature and a quantitative study that reviewed surveillance methods and information pathways of data collection and reporting of RHD in pregnant women in the AMOSS RHD-P study. The Study one qualitative synthesis systematically searched for research on models of care that referenced women with RHD-P, aggregating findings to develop themes [272]. However, in the course of planning and implementing these studies, the design of the qualitative arm (Study 2) was refined. See Figure 3-1 below:

Figure 3-1 Mixed methods study: a transformative framework



The explicit connection between the process and outcomes of research and the furtherance of a social justice agenda [270] in this transformative framework is illustrated by the doctoral outputs in turn informing content in the 3rd Edition of the Australian RF/RHD Guideline. The collaboratively-driven process to develop the chapter maps social determinants against clinical risk and management.

The detailing of research methods for Study two is outlined below. Previously published journal articles (Studies one and three) describe research methods in chapters 4 and 6 respectively.

Study one: Qualitative systematic review – a content analysis

The methods of this study are presented in chapter 4 as part of the published manuscript.

Study two: Care pathways for pregnant women with RHD – perspectives of health professionals

Background

The genesis of this research evolved during the process of developing and conducting the AMOSS RHD-P research project and the findings that arose from its studies.

The experiences of Aboriginal women with RHD and their families in the Northern Territory (NT) were explored as women journeyed through pregnancy in a qualitative arm of the AMOSS research. Its findings illustrated deficiencies of health services particularly in addressing health literacy needs and women's understandings of RHD. The study called for language- and culturally-appropriate health education that promoted a shared understanding relevant to the gender, life- stage and social context of women with RHD [20,21].

The process of implementing the AMOSS RHD-P study [15], along with reviews of the literature and models of care provided for pregnant women with RHD [273] highlighted the need to develop a richer understanding of health services' perspectives in Australia.

Theoretical perspectives and conceptual framework

Themes arising from the previous chapters underlined the need for a richer understanding of the contextual realities of care provision for women with RHD during pregnancy.

I was interested in learning more about perspectives from health services: to tease out discussion points that had been identified in other sections in this thesis as well as the AMOSS study of RHD-P, but also to explore possible themes arising previously not identified.

My contextual review of the literature and the systematic review related to models of care (Chapters 2 and 4), as well as the insights provided by a review of the implementation of the AMOSS RHD-P study (Chapter 6) supported an emerging understanding of key concepts, patterns and associations related to pregnant women with RHD and enabled me to place my research within its theoretical framework.

A qualitative framework was indicated to best explore these themes. Hearing stakeholder perspectives and analysing through a qualitative theoretical lens would permit a nuanced approach and support the development of contextual meaning [274]. I was curious, too, to

explore possible disconnects between knowledge and praxis. For instance, features of care pathways that promote optimal outcomes for pregnant women with RHD including collaborative care were identified nearly 150 years ago [29] and implemented with demonstrable success by the 1920s [37,39]. Why are aspects of these proven measures not adopted as a matter of course? A qualitative approach would allow me to better 'look between the lines' [274].

The qualitative approach was informed by social constructionist theory, a methodological frame of reference that asserts that knowledge is social in origin and not predetermined by some natural order [275]. This attempts to make sense of reality through a relativist orientation: knowledge claims made from constructivist research are not purported to be absolute or universal realities and thus have the dynamic tendency to evolve over time [275]. This fit well with the research questions of my study.

In-depth semi-structured interviews were thought to provide the best fit to address this framework of objectives in order to provide good insight. An interview structure acknowledged constraints (resource burden, access and time) to participants whilst enabling perspectives from a broad base. Interviews with health providers and other stakeholders in relevant policy and RHD service domains were conducted.

Interview data were analysed using a thematic analysis method. This approach promoted the identification of emergent themes generated through the thoughts and experiences of participants.

Methods

Case inclusion

a) Health care providers including midwives, Aboriginal maternal and infant care workers, obstetricians, cardiologists, cardiac care nurses, Aboriginal health workers, remote health providers, RHD health services, health professionals working with other vulnerable populations; and

b) Non-clinical services who are key stakeholders, including RHD control program managers.

Study period

The interviews were conducted between June 2014-January 2018, when data saturation was achieved.

Data collection and analysis research tools

Interviews were held in person (where possible) and by phone. Interviews were all taped and then transcribed. Both audio and transcribed versions were subsequently imported and stored in the n-Vivo program, where coding of theme 'nodes' was conducted.

This process was carried out both individually by two researchers (myself and my primary qualitative supervisor) and in a workshop format to confirm member agreement. Data visualisation was then carried out in the Tableau application, providing a visual framework for the themes and sub-themes.

Lastly, a schema was created that pulled together all themes and sub-themes in a single cohesive representation of the issues and themes related to women with RHD-P.

Recruitment and sampling

The 'logic and power' [276] of purposive (or purposeful) sampling has been described by Patton as lying in *"…selecting information-rich cases for in-depth study. Information-rich cases are those from which one can learn a great deal about issues of central importance to the purpose of the inquiry...Studying information-rich cases yields insights and in-depth understanding*" (p264 [277], quoted in [278]).

Thus, predetermined criteria were created for selecting participants who were best able to answer the research questions (Creswell & Plano Clark 2011; McCann & Clark 2005). The participants in this study included clinicians who provided care for pregnant women with RHD – spanning levels of service, location and professional role. It also included other key stakeholders, predominantly working in Australian RHD programs. Their backgrounds included maternity, cardiac, Aboriginal health-specific services, RHD-specific initiatives, policy shapers; across several domains – urban/regional/remote locations, community/(mid-level)/tertiary level services, government.

Maximum variation sampling [279] was employed in this study with the aim of providing meaningful and rich perspectives on aspects of models of care for pregnant women with RHD from heterogenous samples of those involved in the provision of that care across clinical, policy and strategic domains.

Selection was based on the following factors:

Prevalence: There was an anticipated higher prevalence of RHD in the jurisdictions of NT, WA, western NSW, western and south-western Sydney, far north Qld).

Location: A broad distribution of urban, regional and remote locations (ranging from tertiary urban centres to regional centres and small remote maternity units).

Models of care: Varied models of maternity care ranging from hospital-based high-risk tertiary antenatal care to midwifery group practices; Aboriginal health services; primary health care; and cardiac care.

Role: Individuals were invited to take part based on their experience of providing care for pregnant women with RHD, and/or their professional involvement in RHD at program and/or policy levels.

Invitation to participate occurred in various ways. Initial contact and requests were often face-to-face in meetings or at conferences. Other initial contact was by email. In all cases, this was followed up by a phone call to discuss the study in more detail, and then a formal request by email with the participant consent form.

Twenty-one people were invited to semi-structured in-depth interviews. Of these, nineteen participated.

Interview structure

The study protocol determined in-depth semi structured interviews (in person and/or by phone) that explore perspectives, knowledge and experience of health service professionals (health providers and other stakeholders) working in selected centres and groups across Australia.

Interviews lasted between 60 and 90 minutes, were conducted at a time convenient to participants, and were digitally recorded.

Interview questions

The interviews were guided by questions related to the following:

1. Professional role and location (maternity, cardiac, Aboriginal health, remote health, RHD Control program staff).
2. Knowledge and awareness of ARF/RHD: disease, treatment, management (general and in pregnancy). Knowledge of access to resources.
3. Experience of provision of care for pregnant women with RHD: when diagnosed, what the woman was told, her knowledge and understanding of the disease and treatment. Health professionals' experience and understanding of secondary prophylaxis,

echocardiography, issues about anticoagulation. Experience and understanding of surgical or other interventions.

4. Health care service provision for pregnant women with RHD: logistics, access, strategies, policies, resources. Challenges and gaps. Identifying what works and what doesn't.
5. Urban and regional/remote differences in health (general, cardiac and maternity) care access and services.
6. Health information systems and surveillance: what degree of coordination exists between systems to support optimal and timely sharing of information? What gaps exist? How integral is the RHD Control register perceived as being in the provision of care for women with RHD? What gaps exist, and what strategies have been successful in promoting timely effective sharing of health information?

See Appendix 4: Interview guide for detail.

Hearing and reading the messages; the transcribing process

After conducting interviews, I listened and re-listened to these either in part or all, during the process of reading transcripts and of coding themes and sub-themes.

Interviews were transcribed by a paid external service: a woman who was not involved directly in this research, but who had conducted a considerable amount of transcribing for other qualitative studies in health. Interviews were listened to at several levels for meaning, supported by specific re-listening where words were unclear.

Listening and re-listening to the interviews; and reading and re-reading the transcripts helped to immerse myself in the data, in order to make sense, best interpret and understand the interviews. This commenced from the first interview in 2014 and shaped development of codes and themes during subsequent interviews and during the analysis.

Audio interviews were then imported into the n-Vivo program, where both audio and transcribed versions were stored.

I re-listened to each interview in the process of coding themes, which served to both re-familiarise myself with the content and context, check nuance and clarify words. This provided additional tiers of understanding and analysis; as well as a tool for me to learn from and reflect on the way I approached and conducted the interviews.

Data analysis and interpretation

Thematic analysis of semi-structured interviews was undertaken, in order to identify patterned meaning across the dataset. The inductive approach permitted coding and theme development that was directed by the content of the data.

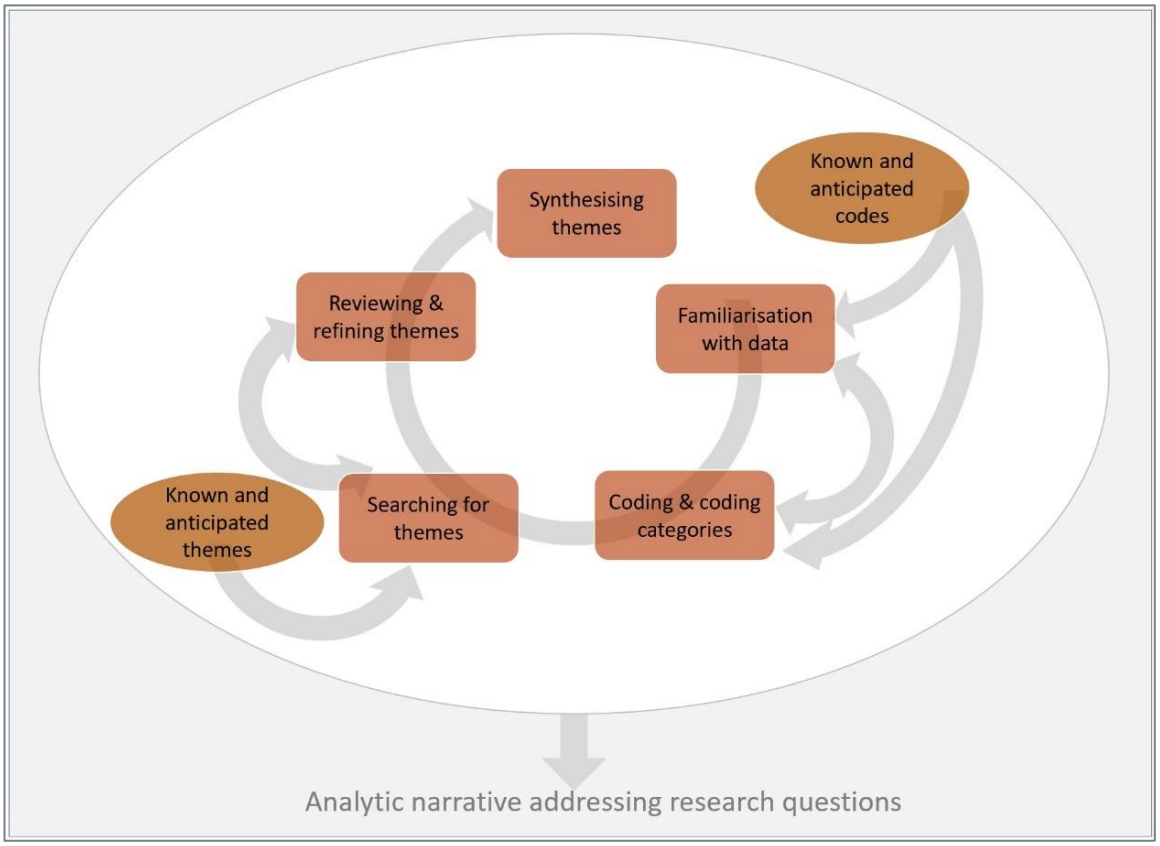
Interview data were analysed using thematic analysis (TA). This analytic approach was chosen in order to identify and analyse patterned meaning across the dataset, provide description of those patterns and the overarching design that unites them [280] in order to generate answers to the research questions being addressed. TA is methodologically agnostic, giving it a theoretic flexibility [281] that it aligns well with the underlying methodology of social constructionism.

Data familiarisation, data coding, and theme development and revision were conducted as a recursive process (Figure 3-2). Interview audios and transcripts were reviewed multiple times as coding and themes were developed as described above. Known and anticipated codes informed the initial coding and coding categories. These were revisited during the process of data familiarisation and immersion generated new and refined codes. The inductive approach permitted coding and theme development that was directed by the content of the data.

As well as directly coding in transcriptions, this iterative process included NVivo query tools such as a word frequency report, which helped inform an additional tier of searching and cross-checking. In turn, evolving themes were reviewed and refined during the analytic process, establishing the scope and focus of each theme, determining its 'story' in order to develop the analytic narrative (see Figure 3-2 below).

The segmentation, categorisation, summarising and reconstructing of qualitative data during the thematic coding process helped unravel important concepts. These could then be used to describe the patterns which were analysed using a thematic approach [280].

Figure 3-2: Thematic analysis – a recursive process



The inductive process of thematic analysis was supported by qualitative tools of rigor including triangulation and an audit trail.

Ethics

Ethical approval was sought from the administering institutions and lead HRECs (initially University of New South Wales [reference 2014-7-36, 2014]; and subsequently ratified by the University of Technology Sydney when the primary supervisor and then myself moved to that institution [reference ETH17-1349, 2017].

Letters of support were received from participating centres. Participant information sheets and accompanying consent forms were distributed to all those interviewed, and consent was obtained before participation was confirmed.

All participants were informed, both verbally and in writing, about all aspects of the study; that they were free to withdraw at any time. They were given opportunities to ask questions about the study, and questions were actively sought to ensure that they fully understood the study process. All of the participants had time to reflect upon the study before giving consent, as there were days and sometimes weeks between participants

being informed of the study and being interviewed. The participant information sheets and consent forms were written in plain English; and all of the participants had a good understanding of English and were competent to give consent.

All participant information sheets included the telephone number for a direct contact with the Human Research Ethics committee and the protocol number of the study in case they wanted to make a complaint or to enquire about ethics approval.

Each participant was assigned a unique identifier, providing the ability to distinguish participants whilst protecting anonymity. Pseudonyms were used for any reference to a person's name or a place location by a participant that was referenced in study findings in order to ensure confidentiality and anonymity.

See 'Funding support' section for details of scholarship and other award funding. There is no conflict of interest.

Data protection

The NVivo project file, which contained audio and transcript files for all participants, was password-protected. The password is held by myself and was given to one of my supervisors. The personal computers on which data were stored have rigorous password-protection with access by a single user account only. Files stored at the administering institution UTS Sydney were similarly protected, with storage and single-user access on the home drive.

Rigour and trustworthiness

The study was guided by the underlying principle of seeking rigour and trustworthiness through a methodology "… deemed worthy of recognition and wider dissemination." Padgett 2008 p179 in [279]. It aims to carry out the research ethically and provide findings that represent as closely as possible the experiences of the participants [282].

In line with the strategies outlined by Shenton, drawing on Guba's work [283], four strategies for rigor pursued during the study include:

Credibility: I attempted to demonstrate that a *'true picture of the phenomenon under scrutiny is being presented, and that what is presented is authentic'* [283]. What was the 'fit' of what interviewees said and how I represented those views? Based on the constructivist assumption that there is no single reality, but rather multiple realities and multiple truths

(Carpenter and Suto, 2008 p149 in [279], I aimed to represent those multiple realities revealed by participants as accurately and authentically as possible through 1) member checking where meaning was felt to be ambiguous or lacked clarity with participants who were purposively selected for their knowledge and perspectives; 2) triangulation and 3) peer review with supervisors who reviewed audio and transcripts of interviews and with whom I conducted workshops to develop themes.

To promote **transferability**, I endeavoured to provide context of those who were interviewed in order to determine that findings could justifiably be applied to other settings. I describe the professional contexts and settings of participants in detail and how they were selected for the study. While my study is not 'generalisable' in a statistical sense based on how the sample was chosen, it aims to hold resonance and a capacity to stimulate thought, improve practices and policies, and provide a base for further research [282].

The meeting of the **dependability** criterion has been described as difficult in qualitative work [283] in its striving to enable a future investigator to repeat the study. However, Liamputtong [279] and Padgett [282] assert that the auditability, while they may not necessarily result in the same conclusions, but provide a logic that others can make sense of. The methods I have used are systematically documented in an attempt to achieve this.

Finally, to achieve **confirmability**, I took steps to demonstrate that findings emerged from the data and not my own predispositions [283]. Triangulation with the other two studies affirmed findings of this qualitative study, and ongoing contact, particularly with my primary qualitative supervisor.

Reflections on the interview process and structure

Interview questions referenced the guide (see Appendix 4: Interview guide) and were approached both thematically, to produce knowledge; and dynamically, to enhance interpersonal relationship in the interview [279].

The researcher: personal perspective and reflections

The motivation for commencing this doctoral research was influenced by my background, politics and work. I had retained my strong interest in women's health, infectious diseases - particularly in vulnerable populations - from my previous clinical nurse background.

This fed into subsequent academic study in public health and my role coordinating studies of rare and/or serious conditions in pregnancy. The AMOSS (Australasian Maternity

Outcomes Surveillance System) commenced in 2009 as a National Health and Medical Research Council (NHMRC)-funded research project of several conditions. Informed by AMOSS study Investigators and following discussion with stakeholders in Indigenous health and RHD initiatives, a second NHMRC (#1024206) project grant commenced, exploring the impact of RHD-P in a two-armed study. Its clinical impact was examined under the umbrella of the AMOSS system [15], while an exploration of women's journeys with RHD during pregnancy was conducted in the NT [20].

The process of working and having conversations with an extremely broad spectrum of stakeholders commenced before the funding was confirmed and shaped the operationalising of the RHD-P AMOSS project. From its inception, issues related to health services that were outside the scope of the study became evident. From this direct experience, overlaid with my strong interest and background, I became keen to explore issues of provision of care for women with RHD, particularly in pregnancy, with a focus on health services and systems.

As principal investigator, guided by my supervisors, I conceived and conducted all aspects of the studies that form this doctoral thesis.

I was conscious that my background would shape the way the research study was approached, analysed and evaluated [284]. Would it constrain findings and usefulness?

I am not an Aboriginal nor a Torres Strait Islander woman. I am not a midwife. I have not worked in clinical settings for a number of years. I have not worked in remote settings. I therefore do not have the lived experience of many of the issues faced by women who live with RHD, nor those of the health services who provide care for them. I was forced to reflect on my motivation and what I could bring to the study.

I had worked from the early outset of the AMOSS studies, establishing processes and systems to support the project. This entailed building a large network over 300 maternity sites which was extended considerably to include Indigenous and primary health settings during the RHD-P study. In developing this network, I worked at various levels, building an understanding of health services and the people in that system.

I have worked with vulnerable women both in women's health and in voluntary capacities. This and my own life experience have shaped my perspectives and given context. I bring my background into this work and acknowledge it. It is not a neutral space. I have influences of

social structures, my background, politics, language and culture, laden on my self as unique 'habitus' [285]. However, I worked to set aside the impact of my own preconceptions or assumptions throughout the study in order to minimise influencing the integrity of the research.

There was possible value, too, in being an outsider [284]. The very nature of RHD, its impact in women, and specifically in pregnancy, means that it bridges multiple health domains, sectors, professions, locations, and models of health care delivery. Perhaps my agnostic position and broad-based background could be an advantage here?

I came to the simple conclusion the experience of women with RHD and what they faced was not right. I aimed to shine a light on what worked and what did not from the perspectives of health services. I endeavoured to make sure the process of collecting and exploring the data was done with integrity and represented a true reflection of participants' views.

An underlying intent from the outset was to ensure the findings of this study and others in the research (along with findings of other relevant studies such as the preceding NHMRC project grant of AMOSS RHD-P) were usefully incorporated into tangible translational outputs such as Guidelines and inform advocacy work.

Study three: Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia

The methods of this study are presented in chapter 6 as part of the published manuscript.

Chapter 4 Models of care for pregnant women with rheumatic heart disease: content analysis

List of presentations and publication from this chapter

This Chapter is the accepted version of the article which has been published in final form at DOI: 10.1111/birt.12435 as detailed below:

Vaughan, G., Dawson, A., Peek, M., Carapetis, J. & Sullivan, E. 2019. *Standardizing clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis*.
Birth: Issues in Perinatal Care (early view), 2019: 00 1-14

About this Chapter

While there are systematic reviews of cardiac disease in pregnancy, and of aspects of management of RHD, there have been no known reviews that systematically examine approaches to care specifically for pregnant women with RHD and associated reporting measures.

Following the review of guidelines and developing a framework of reporting measures in Chapter 2, I conducted a content analysis of models of care for women with RHD-P. The review objective was to improve understanding of reporting of attributes of care for these women and how they align to guidelines as reviewed in Chapter 2.

Standardizing clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis

Abstract

Background: Rheumatic heart disease (RHD) is a preventable cardiac condition that escalates risk in pregnancy. Models of care informed by evidence-based clinical guidelines are essential to optimal health outcomes. There are no published reviews that systematically explore approaches to care provision for pregnant women with RHD and examine reported measures. The review objective was to improve understanding of reporting of attributes of care for these women and how they align to guidelines.

Methods: A search of 13 databases was supported by hand-searching. Papers that met inclusion criteria were appraised using CASP/JBI checklists. A content analysis of extracted data from the findings sections of included papers was undertaken, informed by attributes of quality care identified previously from existing guidelines.

Results: The 43 included studies were predominantly conducted in tertiary care centers of low-middle-income countries. Cardiac guidelines were referred to in 25/43 studies. Poorer outcomes were associated with higher risk scores (detailed in 36/41 quantitative studies). Indicators associated with increased risk include anticoagulation during pregnancy (28/41 reported) and late booking (gestation documented in 15/41 studies). Limited access to cardiac interventions was discussed (19/43) in the context of poorer outcomes. Conversely, early assessment and access to regular multidisciplinary care was emphasized in promoting optimal outcomes for women and their babies.

Conclusions: Despite often complex care requirements in challenging environments, pregnancy provides an opportunity to strengthen health system responses and address whole-of-life health for women with RHD. A standard set of core indicators is proposed to more accurately benchmark care pathways, outcomes and burden.

Keywords: Health Care Quality, Access, and Evaluation; Pregnancy; Rheumatic Heart Disease; Social Determinants of Health; Systematic review, Best practice

Introduction

Rheumatic heart disease (RHD) is a preventable disease of inequity. It is twice as common in women [6,69,72,286], creating added risk in pregnancy. There are many challenges to providing optimal care for women with RHD, particularly in low-and-middle income countries. Service provision is limited by poorly resourced expertise and facilities with barriers of distance and cost. There is often deficient awareness for women and health services of RHD and its impact in pregnancy.

Consequently, the higher prevalence of RHD in pregnancy (RHD-P) in low-and-middle income countries is matched by poorer outcomes than in high-income countries, with documented maternal mortality rates of up to 37% [13]. Its burden is also high among vulnerable populations in upper-income countries. In Australia, Aboriginal and Torres Strait Islander women are over five times more likely to die from RHD [287], with RHD-P rates for Aboriginal Northern Territory women up to 63 times those of non-Indigenous women [15]. Inequitable outcomes are also seen in Māori and Pasifika women[288] and First Nation populations in North America [8,67]. There are growing numbers of women with RHD in high-income countries as migration from resource-poor countries increases [289,290].

There are no known systematic reviews that describe approaches to care and associated reporting measures for women with RHD-P globally. A review of the burden of antenatal cardiac disease in South Africa has a strong focus on RHD [291]. Guidelines refer to all-cardiovascular pathologies in pregnancy[196], or are referenced in non-pregnancy-specific cardiac valvular [193,194,292] or RHD-specific guidelines [6].

Reporting measures for studies of cardiac disease in pregnancy are currently in development [265] as part of the Core Outcomes in Women's and Newborn Health (CROWN) initiative [263,293], but there is no known equivalent for RHD-P, which has specific risks related to its epidemiology.

Although clinical pathways can vary considerably according to the severity of RHD, principles of care that promote optimal maternal and baby outcomes include early diagnosis; preconception care including surgery and other interventions where required; early antenatal assessment including echocardiogram; access to specialized centers and treatment for high-risk women; and collaborative individualized care across disciplines and sectors [6,192,196].

The purpose of this study was to systematically examine descriptions of care provision and associated outcomes for women with RHD-P in order to improve the understanding of how attributes of care are reported and how they align with guidelines.

Methods

Because of the lack of internationally accepted RHD-P measures, we reviewed relevant models of care and associated reporting measures referred to in clinical guidelines to conceptualize existing measures in a framework. We found no specific guidelines for RHD-P. Guidelines were chosen that addressed all-cardiac disease in pregnancy [196] and RHD with some reference to pregnancy [6].

The scope was further broadened to include cardiovascular care standards in primary health settings for Australian Aboriginal and Torres Strait Islander peoples [192]. This guideline outlines elements of care across the continuum of risk and disease, with a focus on reducing disparity in access and outcomes: applicable for most populations where RHD is disproportionate.

Reporting measures relevant for women with RHD-P were identified and grouped in three categories to provide an analytic tool with which to interrogate the literature (Figure 2-2). These included the following: clinical information and reporting; risk in pregnancy; and RHD through the life-course. This framework served to guide the analysis of data gathered for the systematic review presented in this paper.

Data sources and search protocol

A structured search of peer-reviewed research literature identified studies that described clinical care and measures for women with RHD-P. Data were extracted from the reported results of included studies and examined using a content analytic process[294], directed by the framework of reporting measures (Figure 2-2).

The study was registered with the International Prospective Register of Systematic Reviews (PROSPERO #CRD42018059849).

Searches on PubMed, Medline, EMBASE, CINAHL, Nursing and Allied Health Database, ATSIhealth, Indigenous Collection, Rural and Remote Health Database, ETG Complete, ISI Web of Science, Public Library of Science and Trip Pro Databases; were supported by hand-searching. The search strategy incorporated a combination of free term text items and

Medical Subject Headings (MeSH): ("rheumatic heart" or "rheumatic fever" or “valvular heart disease”) and ("pregnancy" or "pregnancy complications" or "pregnancy, high-risk" or "pregnancy complications, cardiovascular" "maternal") and ("models of care" or "guideline*" or "health service" or "maternal health services" or "primary health care" or "practice guideline" or "guideline adherence" or "health services accessibility" or "health care"). Inclusion criteria included: all English-language peer-reviewed studies after 1994 in any setting or country with reference to RHD-P and attributes of care (Table 4-1).

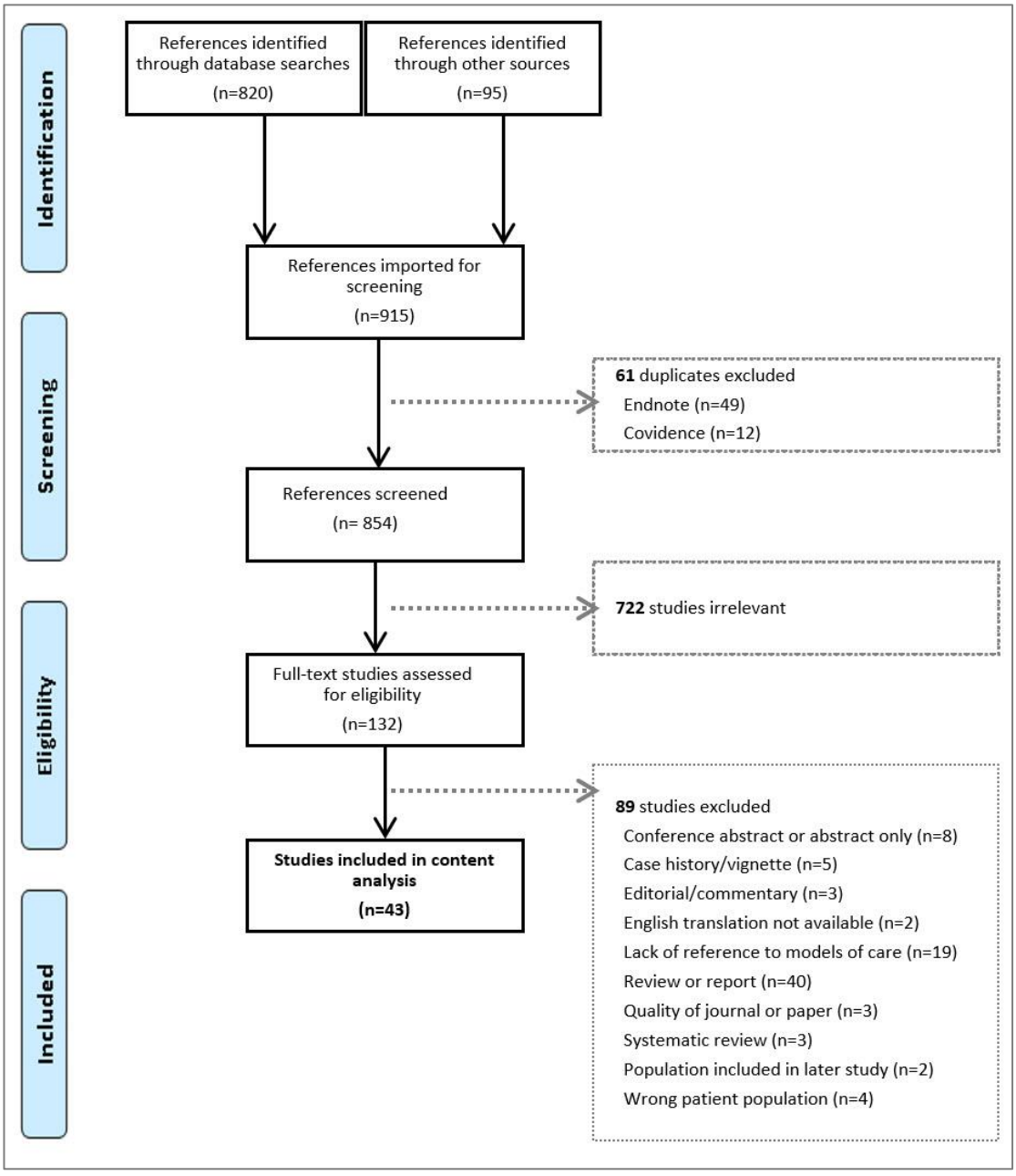
Table 4-1: Inclusion / exclusion criteria for content analysis of studies with reference to RHD-P

Included	**Excluded**
1995-2018	Pre-1995
English language	Non-English
Any setting in any country	None
Any study of women with cardiac disease with reference to RHD and pregnancy and attributes of care	Conference abstracts Opinion pieces/editorials Guidelines/reviews Systematic reviews Studies of biomedical treatment/interventions for women with RHD that do not refer to models of care in pregnancy

The PICOS framework (Population, Interventions, Comparators, Outcomes, Study design) [295] guided the review question: *In studies that reference pregnant women with RHD, what core reporting measures are used to describe models of care?*

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [296] informed the review. Screening utilized Endnote™ bibliographic and Covidence™review tools. Critical appraisal referenced CASP and JBI checklists [297,298] and the quality appraisal is summarized in Figure 4-3 as a four-tier grading. Differing judgments on inclusion were resolved by consensus, or, where no consensus was achieved, by a third reviewer. Reasons for excluding studies were clearly documented (Figure 4-1).

Figure 4-1: PRISMA breakdown of studies with reference to RHD-P



Data extraction and content analysis

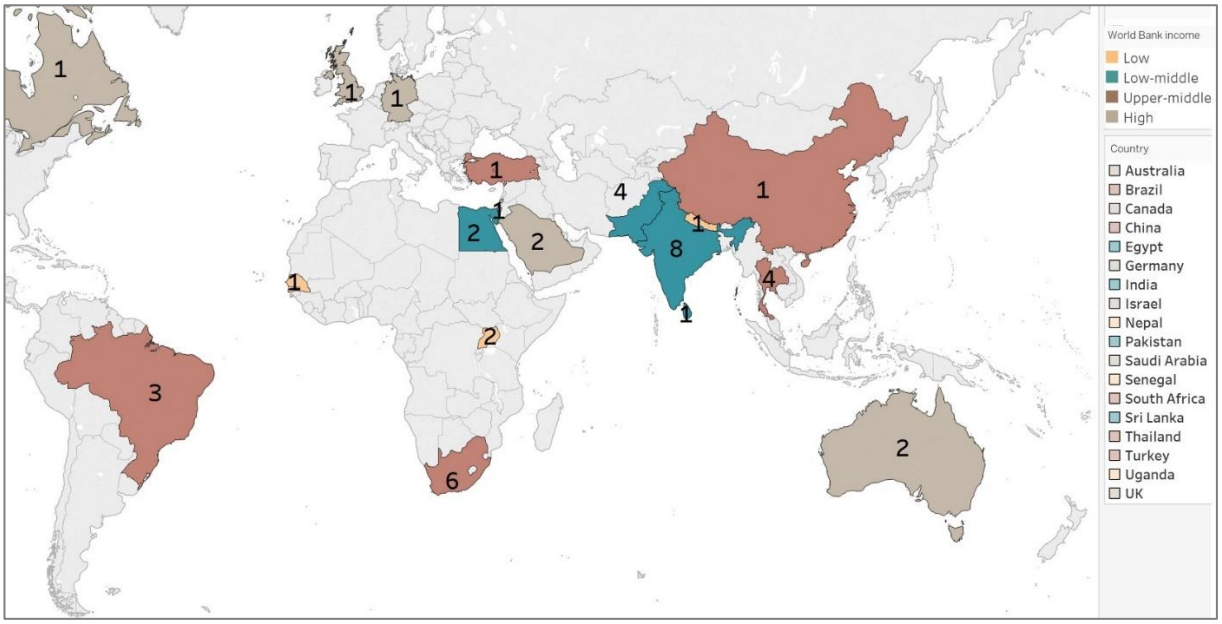
A data extraction tool was developed using Microsoft Excel™. Visual mapping used Tableau™v2018.2.0 analytic software. Study characteristics included (Table 4-2, Figure 4-2) country, World Bank income category, study design, setting/s and population, as well as documenting maternal mortality. Data were coded against the reporting framework and associated measures (Figure 2-2).

Results

General characteristics and quality appraisal

The most common types of study design were cohort (19) and case-series (20), with two qualitative [20,132] studies, one cross-sectional [299] and one longitudinal screening study [88]. There was considerable heterogeneity in the methodologies, levels of evidence and reporting measures of these predominantly retrospective studies. Individual study characteristics are outlined in Table 4-2. Reflecting the overall burden of RHD, the majority of the 43 studies from 18 countries were from India (8), South Africa (6), Pakistan (4) and Thailand (4), with one multi-country (predominantly Egypt) study (Figure 4-2). Most were published after 2004, paralleling a resurged clinical and research interest [300]. The distribution of studies by country and World Bank income category is detailed in Figure 4-2.

Figure 4-2: Number of studies referencing women with RHD-P: by country and World Bank income category



All studies were conducted in tertiary care settings with access to cardiac (or dedicated obstetric-cardiac) care, as well as primary health settings [20,88,132] and regional centers [82,88]. Maternal mortality ranged from 0% (16/42) to 37% [13]. Between 1-4% of women died in nearly half (20) of the studies. One study found significantly lower mortality rates in its index population (10%) compared to referred women (32%) [82].

Study designs impacted on quality and were subject to high levels of bias, especially the case series. Referral and other selection biases as acknowledged in several papers

[82,88,221,236,301-309] were particularly related to the predominantly single site tertiary care level settings. Study periods ranged from six months to 21 years, with five of unspecified periods [222,307,310-312]. Long study periods (from 10-21 years in 16 studies) were noted to impact on protocols which changed in response to therapeutic advances during that time [301].

Figure 4-3 provides the quality appraisal overview and maps studies against reporting measures. The studies were assessed as low (9), medium (12), medium-high (21) and high (1) quality respectively. Key reporting measures from the framework (Figure 2-2) were poorly documented.

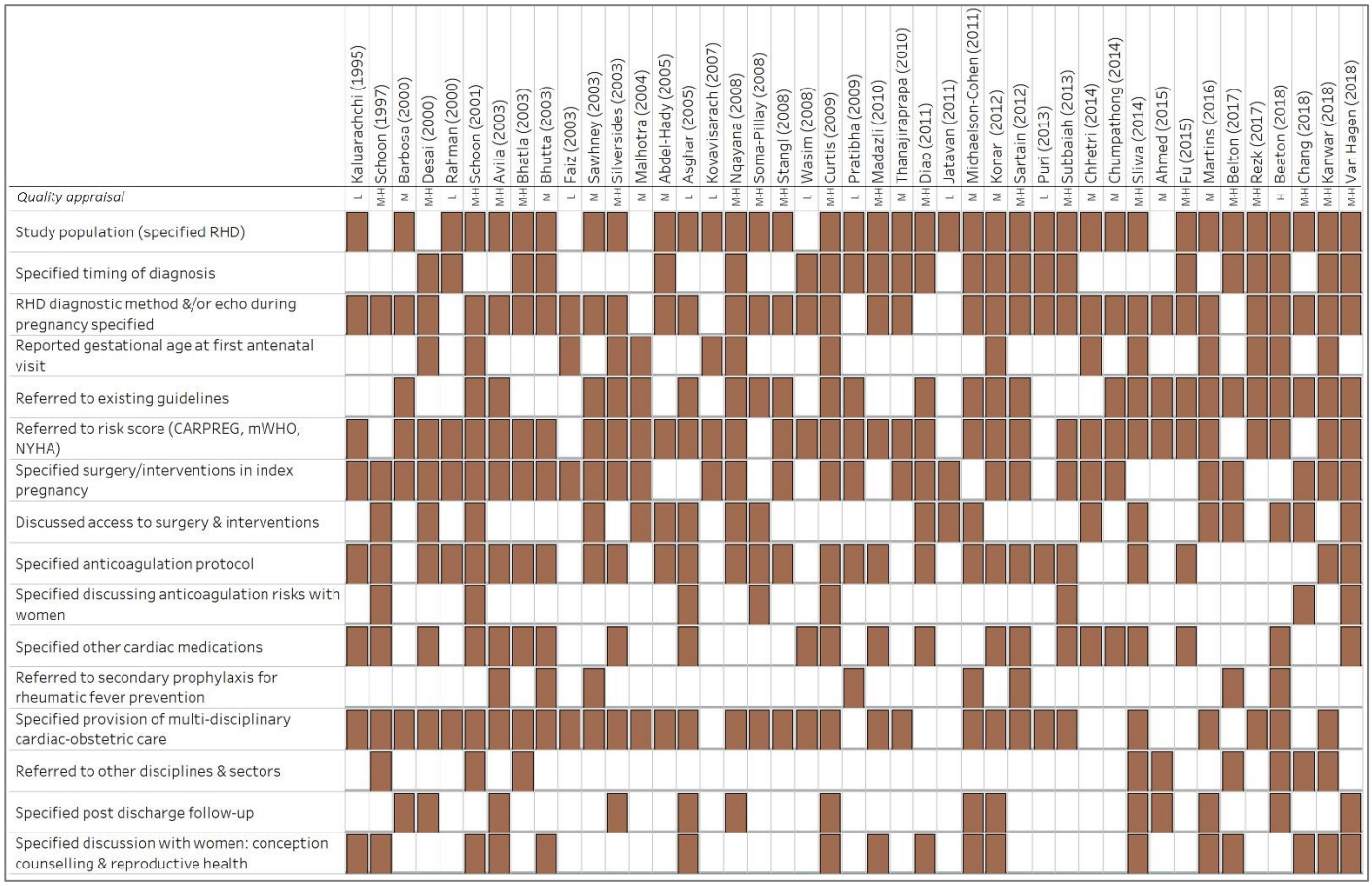
Clinical information reporting

The percentage of the study population with RHD ranged from 100% (11 studies of women with RHD or mitral stenosis) to 3% in a high-income country [289], with most comprising over 55% of the study population Table 4-2. Six studies [235,299,312-315] from countries with an otherwise medium-to-high burden of RHD did not give a breakdown of underlying pathology of mitral stenosis or all-valvular heart disease (Table 4-2, Figure 4-3). Mitral stenosis in women during their reproductive years is usually of rheumatic origin [196,316] and was used as a proxy for RHD where causation was unspecified.

Heart disease in low-and-middle income countries is commonly diagnosed in pregnancy on development of severe symptoms [13,82,291,310,317]. However, 18 of the 41 quantitative studies did not specify timing of diagnosis. Others referred to late diagnosis in the context of poorer outcomes and health system shortcomings [221,231,235,303,308,315,318,319]. Diagnosis during pregnancy/post-partum ranged from 1% [320] to 97% [88] in a longitudinal screening study, with eight studies above 20% and four above 40%. In one high-income country, four women (of 95 pregnancies) were diagnosed with RHD after developing peripartum acute pulmonary oedema [239]. One found 7% of women diagnosed post-partum [307], but this was poorly documented overall.

Figure 4-3: Studies with reference to RHD-P: core reporting measures and quality appraisal by study

***Legend**:* visual representation of specified reporting measures (outlined in figure 1 framework) according to study. Shaded square indicates the reporting measure was specified in the study; blank square indicates it was not.
Quality appraisal assessed as L (Low), M (Medium), M-H (Medium-high) or H (High).



Thirty-six studies specified echocardiographic review during pregnancy, although only four referenced diagnostic criteria [88,239,309,313]. Six studies did not specify RHD diagnosis confirmed by echocardiography, nor its use during pregnancy [13,237,312,317,321,322].

Models of care and risk in pregnancy

There was limited or no reference made to guidelines related to the care of pregnant women with cardiac disease in 16 studies.

The majority of the 36 quantitative studies that specified a cardiac risk score used the New York Heart Association (NYHA) classification (I-IV) of functional capacity [31]. Pregnancy-specific scores referenced CARdiac disease in PREGnancy (CARPREG) [212] cardiac events risk

index, modified CARPREG [239], and modified World Health Organisation (mWHO) risk classifications [196,240]. A referral algorithm was developed for suspected and known cardiovascular disease in a low resource setting [221].

Poorer maternal and fetal outcomes were associated with higher risk scores (NYHA>II [222,228,230,231,236,237,299,303,306,309,311,312,314,317-321,323-327], NYHA>I with mitral stenosis [309], mWHO>1 [221], CARPREG [232,304,318,325]/modified CARPREG [239]>0 or study-specific factors such as mitral stenosis and anticoagulation therapy leading to increased maternal risks of heart failure, pulmonary hypertension, thromboembolic episodes, atrial fibrillation and death [82,88,235,305,308,312,313,315]). The CARPREG index underestimated cardiac events in low-risk women but over-estimated it in CARPREG>0 in one study, possibly reflecting late diagnoses in pregnancy [318]. The quality of care and avoidable factors associated with near-miss morbidity was assessed in two papers [82,308], while others described gaps between guideline recommendations and clinical implementation leading to compromised care [289,309].

Late booking and/or infrequent antenatal care hampered early diagnosis and treatment [13,235,237] and was associated with poorer cardiac and perinatal outcomes [82,221,230,235,308,315,325,326], yet the gestational age at first antenatal visit was reported in only 15 of 41 quantitative studies.

Medical management (such as beta-blockers, digoxin and/or diuretics) and percutaneous balloon mitral valvuloplasty [328] (hereafter valvuloplasty) in refractory cases of mitral stenosis generally improved outcomes [231,301,302] where reported. However, studies emphasized the challenges of providing optimal care in resource-challenged environments, including appropriate clinician skills, access to medication, valvuloplasty and surgery, health system shortcomings and sociocultural factors [82,88,132,221,231,235,237,302,303,308-310,312,315,318,319], with one (where 37% of women with rheumatic valvular disease died) noting that valvuloplasty facilities were simply unavailable at their tertiary center [13]. The multi-country study found a greater number of valvular interventions in high-income countries despite more women in low-income settings having severe mitral stenosis [309].

Despite the potentially catastrophic maternal-fetal risks associated with the use of anticoagulation in pregnancy, the regimen was detailed in only half of the 28 quantitative studies that specified the number of women on therapy. Nine referred to discussion with women in the context of complex decisions surrounding choice of regimen which balanced the maternal risk of thromboembolism using low molecular weight heparin against the

increased fetal risk of warfarin use. Studies discussed late booking affecting the anticoagulation regimen [222,230,315,324] with lack of adherence to protocol (or access to treatment) detailed as an important risk factor for morbidity and mortality[222,308,315,317,320]. Warfarin embryopathy/fetopathy was likely underestimated in studies where postmortems or detailed examinations were not performed [308,323].

Secondary prophylaxis (usually 3-4 weekly bicillin injections), where indicated, prevents rheumatic fever recurrence and is safe during pregnancy. Its use was referred to in only seven quantitative studies.

Most studies emphasized the need for multidisciplinary care in discussion and/or recommendations although somewhat fewer (32) specified its provision in their study. This was highlighted in one study that found obstetric-cardiac individualized review determined according to risk promoted optimal outcomes despite its low-resource setting[221]. Others similarly pointed to early multidisciplinary evaluation and management contributing to few or no maternal deaths in otherwise high-risk women [222,228,230,231,235,239,306,314,322,323,327].

Vaginal birth is recommended for women with valvular heart disease unless contraindicated by severe cardiac morbidity [196], or obstetric complications. Caesarean section rates varied enormously from less than 10% [310,312,314,322] to 75% [326], with several studies above 40% [222,228,236,303,308,309,320] and higher again in groups stratified by risk or poorer outcomes [228,236,309,320,326].

There was limited reference to care outside the index pregnancy period. Those that did noted the continued heightened risk of morbidity and mortality [221,309], and another detailed an increased need for cardiac intervention in the first year following delivery [289].

Papers that called for improved clinician training in primary health settings to support cardiac disease detection/referral [313,315,325] were mostly (5/6) published since 2014. Gaps in awareness among primary health care nurses (and women) were associated with delayed referrals [221] and consistent with other studies that found women received contradictory advice and limited education [20,132]. Language-appropriate health education that promoted a shared understanding was largely absent for Aboriginal women with RHD [20].

RHD through the life-course

Twenty-six studies did not specify the provision of conception counselling and reproductive health in their setting, with one listing it under management standards not followed [289]. Women can perceive risk to be over with the end of pregnancy [323], underscoring the significance of postnatal counselling.

Emerging themes in a qualitative study of women's experiences with RHD included misconceptions about side-effects of contraceptives; lack of agency in reproductive decision-making; and stigma related to financial and perceived reproductive limitations [132].

Discussion

The aim of this review was to synthesize the literature and map reported measures against a framework drawn from guidelines related to models of care for RHD-P. Our study found gaps in the three framework categories of clinical reporting, risk in pregnancy and RHD through the life-course.

A recent overview of RHD strategies emphasizes the imperative for accurate, current data in order to inform policy and measure trends [286]. Poor reporting of measures related to cardiac pathology and diagnoses precludes a true assessment of the burden of RHD-P and changing epidemiology. In turn, this is limited by the capacity of health services to diagnose cases. Women with subclinical or milder forms of disease or fatal events prior to admission are likely to be missed in low-income settings [221]. The community-based screening study found less than 4% of women with RHD were aware of their diagnosis pre-pregnancy [88]. There are no known studies of the impact of RHD-P from countries that have among the highest reported rates of RHD in the world [329], including the Pacifica [330,331] and Oceanic regions [102]. A population-based study conducted in the high-income countries of Australia and New Zealand (currently under review)[3] shows similarly high rates among Māori and Pasifika women [288,332].

Reporting gaps are consistent with a South African systematic review of antenatal heart disease, which recommended minimum criteria including diagnosis, reference population, cardiac profile and outcomes [291].

[3] Now published as: Sullivan, E., et al. (2019). "The high prevalence and impact of rheumatic heart disease in pregnancy in First Nations populations in a high-income setting: a prospective cohort study." BJOG: An International Journal of Obstetrics & Gynaecology 00.

While the lack of diagnostic reference to echocardiography is partly explained by study periods, resource limitations of facilities and expertise no doubt also impact. However, increasingly portable technologies and successful screening programs strengthen the argument for earlier review in primary care settings [88,333,334]. Standardization has improved with the 2012 echocardiographic diagnostic criteria [73].

The high risk of anticoagulation in pregnancy requires better reporting in any study of women with RHD. A recent meta-analysis of anticoagulation in women with mechanical heart valves found fetal risk was similar between women taking <5 mg warfarin daily to those on low-molecular weight heparin [335]. These findings underscore the need for granularity of reporting prescribed regimens – including level of adherence and whether women had access to treatment.

Increasing calls to improve the scale-up of and access to surgery/interventions in low-income settings [336-340] reflect service deficiencies highlighted in studies.

There were few studies that followed RHD-P care trajectories and outcomes outside tertiary centers. A small but growing number of initiatives such as the landmark RHD screening study [88] harness specialist resources in community settings to improve early diagnosis of RHD and care for women. These are embedded in collaborative cross-sectoral approaches [334], drawing on successful strategies developed in other chronic disease models and supported by strengthened health systems [88,341]. They potentially obviate the need for emergency-driven, costly tertiary care [82] - and in turn support improved outcomes for this disease which is preventable at many levels [6]. Such principles can equally apply to vulnerable populations in high-income countries [342]. However, executing these models in practice is often tested by the plethora of structural, political and economic barriers to implementation that are part of the RHD landscape.

The overall lack of reference to post-discharge care (including recommended follow-up) suggests likely under-reporting of complications. Three of four RHD-related maternal deaths reported in the multi-country study were up to six months' post-partum [309], consistent with (often-avoidable) factors and risks reported in other studies of late maternal death [217,219,343].

Existing literature on preconception and reproductive health care is predominantly focused on congenital heart disease. There is a growing body of evidence of the role of

preconception care in optimizing general health and risk awareness in marginalized communities [207]: highly relevant for women with RHD [69].

There are no RHD-P-specific guidelines. Selected reporting measures were drawn from cardiac disease in pregnancy guidelines that referenced RHD [196], the Australia-specific RHD guidelines [1,6] (an updated edition of which includes a substantially enhanced section on women and RHD [1]) and cardiovascular standards for Aboriginal and Torres Strait Islander peoples [192]. Guidelines are themselves mostly based on case series and observational studies. However, we believe the included reporting measures reflect fundamental principles of care for vulnerable populations where RHD is prevalent, particularly in relation to maternal health. RHD-specific research to test the evidence is required to strengthen the rigour of recommendations, better understand the effects of pregnancy and choose the best individualized plan for ongoing care.

We propose the reviewed framework of measures (Figure 2-2) addressing the categories of clinical information reporting; models of care and risk in pregnancy; and RHD through the life course as a core outcome set for women with RHD-P, adapted to local cultural, social and economic contexts [344]. A Delphi method review [345,346] to evaluate an extended set with neonatal outcomes (with global stakeholders including health services and women in high-prevalence settings) will further strengthen recommendations for adoption.

Strengths and limitations

This review was constrained by the heterogeneity and design of included studies, with most subject to substantial bias (particularly referral) and reporting inconsistencies. Study sites were predominantly tertiary centers, providing care particularly for those with severe RHD who were able to access specialist care. However, these observational studies provide the best available current evidence and insight in determining models of care associated with optimal maternal outcomes.

What was reported (or not) may not reflect actual practice. In the absence of specific reference to a care attribute, it was assumed that it was not addressed, which may or may not be true. This may be particularly relevant for aspects such as conception counselling [289].

Conclusions

RHD has been described as providing a model for strengthening health systems to address other cardiovascular diseases in limited-resource countries. This framework is especially

pertinent for women with RHD, where best-practice models of care in a strengthened maternal health system are often congruent with those that support women with RHD.

This qualitative synthesis highlights gaps of what is reported in the literature, with consequent under-estimation of burden and weakened ability to action strategies based on findings. We propose a Delphi testing of the reporting framework detailed in this paper and adoption of a core outcome set to support data consistency, comparability of studies, strengthen knowledge and awareness of burden (clinical and social) and improve benchmarking of care for women with RHD.

Table 4-2: Characteristics of studies with reference to RHD-P

Legend:
TCC: Tertiary care center; CR: Community setting and/or regional center
PCS: Prospective case-series; RCS: Retrospective case-series; PC: Prospective cohort; RC: Retrospective cohort; PLS: Prospective longitudinal screening
CDM: Dedicated Cardiac/Maternity clinic; HRPC: High-risk multidisciplinary pregnancy clinic
CDiP: Cardiac disease in pregnancy; VHD: valvular heart disease; MVHD: mitral valvular heart disease; MS: mitral stenosis
Maternal mortality: * Asterisked percentages indicate RHD only

	Study	Setting / Type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
1	Abdel-Hady(2005)[320]	TCC-HRPC	n=86 RHD=90%	PCS	Egypt	Assess maternal/perinatal CDiP outcome.	1%*
2	Ahmed(2015)[313]	TCC-HRPC	n=101 RHD=N/S MS=100%	PCS	Pakistan	Evaluate MS feto-maternal outcomes, patient-specific management plan.	2%
3	Asghar(2005)[310]	TCC-HRPC	n=50 RHD=66%	PCS	Pakistan	Assess maternal/fetal outcome CDiP.	0%*
4	Avila(2003)[301]	TCC-HRPC	n=1000 RHD=56%	RCS	Brazil	Experiences & outcomes CDiP in referral center.	2%
5	Barbosa(2000)[302]	TCC-CDM	n=45 RHD=100%	RC	Brazil	Identify characteristics of complications MS in pregnancy.	2%*
6	Beaton (2018)	PLS, CR	n=58 RHD=88%	PLS	Uganda	Determine prevalence of maternal heart disease through active case finding & its attributable risk to adverse pregnancy outcomes.	2%*

	Study	Setting / Type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
7	Belton(2017)[20]	TCC-HRPC, CR	n=8 RHD=100%	Qualitative, Yarning	Australia	Study RHD-P health literacy; health services responses.	N/A (none)
8	Bhatla(2003)[231]	TCC-HRPC	n=207 RHD=88%	RC	India	Evaluate CDiP maternal/fetal outcome in developing country.	0%
9	Bhutta(2003)[323]	TCC-HRPC	n=170 RHD=91%	PCS	Pakistan	Determine CDiP outcomes post-cardiac surgery.	0%
10	Chang(2018)[132]	TCC	n=50 n= 25 RHD = 100%	Mixed methods	Uganda	Understand factors/attitudes towards reproductive health & disease in women with RHD.	N/A (none)
11	Chhetri(2014)[303]	TCC-HRPC	n=53 RHD=89%	PCS	Nepal	Investigate prevalence, characteristics, outcomes CDiP.	4%*
12	Chumpathong(2014)[304]	TCC-HRPC	n=175 RHD=66%	RC	Thailand	Evaluate CARPREG predicting cardiac/obstetric/neonatal complications.	3%
13	Curtis(2009)[289]	TCC-HRPC	n=177 RHD=3%	RCS	UK	Describe CDiP; review guidelines adherence, identify suboptimal management.	2%
14	Desai(2000)[235]	TCC-HRPC	n=208 RHD=N/S MS=100%	PCS	South Africa	Evaluate management/outcomes MS in pregnancy.	0%
15	Diao(2011)[13]	TCC-HRPC	n=50 RHD=92%	RCS	Senegal	CDiP maternal/foetal outcomes in a low-income country.	37%*
16	Faiz(2003)[314]	TCC-HRPC	n=126 RHD=N/S MVHD=95%	RCS	Saudi Arabia	Review MVHD during pregnancy: incidence, outcome.	0%
17	Fu(2015)[305]	TCC CDM	n=1086 RHD=15%	RC	China	Identify heart failure risk during pregnancy women with pre-existing disease	1%*
18	Jatavan(2011)[237]	TCC-HRPC	n=125 RHD=49%	RC	Thailand	Determine outcomes CDiP.	0%
19	Kaluarachchi(1995)[324]	TCC-HRPC	n=166 RHD=70%	PCS	Sri Lanka	Evaluate CDiP pattern and outcome.	2%
20	Kanwar(2018)[325]	TCC-HRPC	n=66 RHD=77%	PC	India	Identify feto-maternal CDiP predictors	6%

	Study	Setting / Type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
						complications/ outcomes ≤28v>28 weeks.	
21	Konar(2012)[311]	TCC-HRPC	n=281 RHD=69%	PCS	India	Evaluate CDiP, maternal/perinatal outcome.	1%
22	Kovavisarach(2007)[321]	TCC-HRPC	n=196 RHD=55% (period 3)	RC	Thailand	Assess prevalence, demographics, maternal/perinatal outcomes CDiP (3 study periods).	3%
23	Madazli(2010)[228]	TCC-HRPC	n=144 RHD=87%	RC	Turkey	Evaluate maternal/fetal outcome CDiP developing country.	0%
24	Malhotra(2004)[312]	TCC-HRPC	n=312 RHD=N/S VHD=100%	RC	India	Compare pregnancy outcomes of women with VHD to healthy women.	0.6%
25	Martins(2016)[318]	TCC-HRPC	n=132 RHD=62%	RC	Brazil	Determine CDiP risk factors associated with maternal/neonatal complications.	3%
26	Michaelson-Cohen(2011)[306]	TCC-HRPC	n=175 RHD=41%	PC	Israel	Assess CDiP outcome.	0%
27	Nqayana(2008)[222]	TCC CDM	n=95 RHD=81%	RCS	South Africa	Review CDiP in developing country.	0%
28	Pratibha (2014)[317]	TCC-HRPC	n=200 RHD=100%	RCS	India	Study pregnancy outcomes of RHD-P; evaluate perinatal outcomes of Percutaneous Balloon Mitral Valvuloplasty during pregnancy.	1%*
29	Puri(2013)[307]	TCC-HRPC	n=97 RHD=70%	RC	India	Assess CDiP & associated maternal/fetal complications.	3%
30	Rahman(2000)[322]	TCC-HRPC	n=274 RHD=76%	RCS	Saudi Arabia	Review CDiP outcomes.	0%
31	Rezk(2018)[326]	TCC-HRPC	n=204 RHD=100%	PC	Egypt	Assess cardiac/obstetric outcome in RHD-P & predictors of poor outcome.	0%*
32	Sartain(2012)[239]	TCC-HRPC	n=95 RHD=100%	RC	Australia	Determine maternal-cardiac complications/outcomes in patients with RHD.	0%*

	Study	Setting / Type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
33	Sawhney(2003)[319]	TCC CDM	n=500 RHD=100%	RC	India	Study maternal/perinatal outcomes RHD-P.	2%
34	Schoon(2001)[82]	TCC-HRPC	n=42index+ 25 referred RHD=33%	RCS	South Africa	Document CDiP mortality/morbidity; compare complicated vs uncomplicated.	18%
35	Schoon(1997)[315]	TCC/regional-HRPC	n=164 RHD=N/S	RCS	South Africa	Describe maternal outcome CDiP.	10%
36	Silversides(2003)[232]	TCC-CDMs x2	n=80 RHD=100%	PC	Canada	Define predictors maternal-cardiac complications in women with MS.	0%*
37	Sliwa(2014)[221]	TCC-CDM	n=225 RHD=25%	PC	South Africa	Investigate spectrum of disease & maternal/fetal outcome in CDM.	4%
38	Soma-Pillay(2008)[308]	TCC-CDM	n=189 RHD=64%	RCS	South Africa	Assess CDiP profile & maternal/fetal outcome, identify risk categories.	3%
39	Stangl(2008)[236]	TCC-HRPC	n=93 RHD=7.5%	RC	Germany	Analyze risks in low/high-risk women with CDiP.	0%*
40	Subbaiah(2013)[230]	TCC-HRPC	n=100 RHD=64%	RC	India	Analyze CDiP & maternal/fetal outcome.	0%*
41	Thanajiraprapa(2010)[327]	TCC-HRPC	n=193 RHD=69%	RCS	Thailand	Identify complications CDiP.	1%
42	Van Hagen(2018)[309]	TCCs. Multiple sites/countries	n=390 RHD=100%	RC	Multiple countries	Assess maternal/fetal outcomes in women with MVHD.	1%*
43	Wasim(2008)[299]	TCC-HRPC	n=160 RHD=N/S	Cross-sectional descriptive	Pakistan	Assess CDiP; feto-maternal outcomes.	4%

Disclosure of interests: None declared. Completed disclosure of interests form available to view online as supporting information.

Contribution to authorship: Conceptualisation, review design: GV, AD, EAS Drafting of manuscript GV, AD, EAS with input from MP, JC. Study selection and data extraction: GV and AD. Analysis and interpretation: GV, EAS, AD. Critical revision of the review for intellectual content: GV, EAS, AD, MP, JC.

Details of ethics approval: The study was exempt from ethics approval because the research was not conducted with humans or animals and used publicly available data.

Chapter 5 Findings: care pathways for pregnant women with RHD - perspectives of health professionals

Introduction

Previous chapters in this thesis focused on issues related to models of care for pregnant women with RHD and describe gaps and facilitators of optimal care pathways.

The literature review in Chapter 2 highlighted historical deficiencies in health service responses to RHD, particularly in pregnancy, including access to services, data management, clinical care, the health workforce and particularly illustrated the need for collaborative care.

The review of guidelines in Chapter 2 identified key attributes of care relevant for women with RHD and a reporting framework was developed. The findings of the systematic review in the previous chapter described gaps in the reporting of clinical measures of RHD-P. This has implications for the estimation of associated burden that affects the ability of the health system to plan effective strategies to provide – and improve – appropriate maternal and newborn care. In order to better understand the care pathways of women with RHD-P at the health service level and identify strategies that can be employed to support health professionals to improve that care, I undertook a qualitative study to explore their perspectives.

This study presents findings from a qualitative exploration of care pathways of women with RHD from the perspectives of health service providers. The study was designed to investigate issues and themes that emerged during the AMOSS RHD-P project. It explores thoughts, arguments and perspectives of those involved in the many spheres of service delivery for pregnant women with RHD. These bridge clinical and non-clinical care domains – maternity, cardiac and other specialist care; primary health; Aboriginal health-specific services, as well as RHD-specific strategies and programs. They are delivered across several domains: geographically (remote through to urban settings), levels of care (primary health through to tertiary level care) and health sectors (public and private). They are driven by - and shaped within - political and policy frameworks that in turn dictate funding and operational models.

Research questions

This study aimed to provide rich understanding of the contextual realities of providing care for pregnant women with RHD. The research questions are:

1. What is the knowledge, expertise and awareness of health professionals who provide clinical care for pregnant women with RHD?
2. How do health professionals perceive the needs of Aboriginal and non-Aboriginal women with RHD and how does this vary between services?
3. What do health professionals perceive to be the barriers to the provision of optimal health care for women with RHD-P?
4. What strategies do health professionals suggested that can be implemented to more effectively meet the needs of these women, including access, health information systems, education, counselling and clinical management of RHD-P?

Participant characteristics

The recruitment strategy of participants is outlined in detail in Chapter 3. Nineteen participants from clinical and non-clinical settings were interviewed over a four-year period. Roles included midwives, obstetricians, cardiologists, Aboriginal health workers and primary health physicians. They worked in urban, regional and remote locations in hospitals, community and policy settings. The majority of clinical practitioners worked in maternity services. There was a spread across urban and regional locations, with six including remote outreach work (Figure 5-1, Table 5-1).

Figure 5-1: Participants - professional role categories

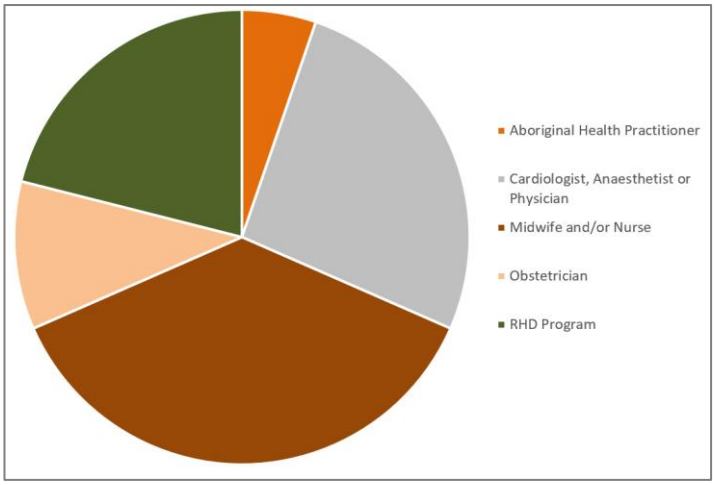


Table 5-1: Participant attributes and characteristics – sector, location, organisation

Jurisdiction	Northern Territory	8
	New South Wales	4
	Queensland	5
	Western Australia	2
Location	Regional with remote outreach	4
	Regional	8
	Urban with remote outreach	2
	Urban	5
Workplace	Hospital-based maternity	5
	Aboriginal Mothers and Babies service	3
	Aboriginal medical service or primary health	4
	Hospital - cardiac or high-risk specialist	3
	RHD program or strategy	4

The table above does not adequately describe the backgrounds of participants, whose experience brought a substantial breadth and depth to the interviews. Some had worked remotely in Australia for extended periods and were now working in urban locations. Others had strong Aboriginal community experience and were now working in hospital settings - and vice versa. RHD program staff brought a rich mix of clinical and public health expertise with them. Some participants were born and had worked in countries with a high prevalence of RHD, or had worked overseas in those regions as part of their clinical history.

These backgrounds all influenced perspectives and in turn promoted a richness of in-depth and thoughtful responses.

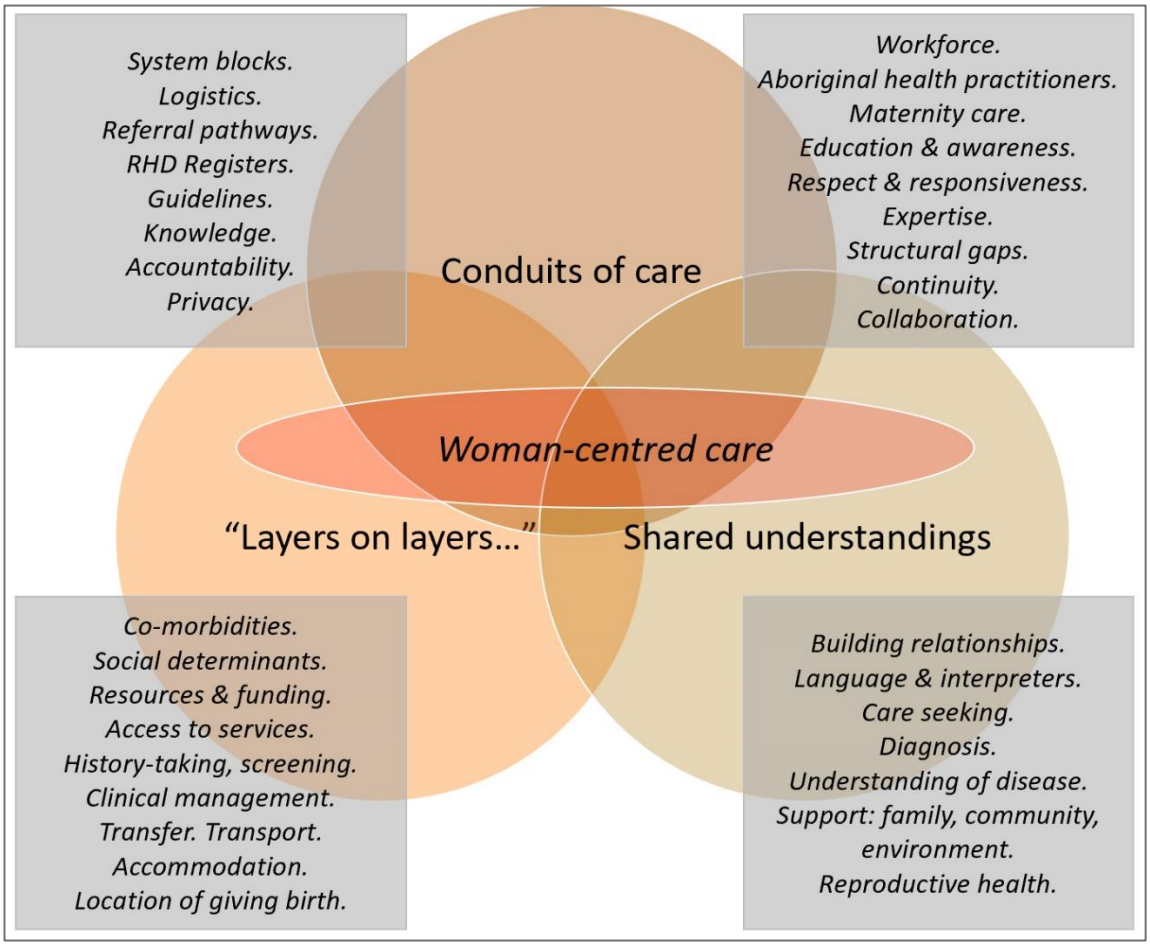
Note: *any reference to an individual's name, specific workplace or identifying geographical location has been omitted or re-named this chapter, and third person references are gender neutral.*

Themes

The process of coding and the categories arising (described in Appendix 5) during the review of interview data helped unravel important concepts. I found a broad range of factors that participants identified as potential barriers and facilitators to the care pathways for women with RHD. The thematic analysis resulted in the following framework being generated (Figure 5-2). Patterns of experience emerged of three main themes incorporating

several interconnected sub-themes. These are described below with explanations regarding their relevance to the research.

Figure 5-2: Themes related to care trajectories for women with RHD-P



Conduits of care. System blocks. Breaking down silos and joining the dots.

Providing cohesive models of care for pregnant women with RHD was challenged by a multiplicity of factors straddling system blocks, logistics, workforce, access to services, information systems, awareness and knowledge.

There were silos identified in all these domains at several tiers. Silos of information. Silos of processes. Silos of knowledge. Siloed care pathways. Siloed workforces.

Yet participants also spoke of their effort to break down these silos and the strategies they employed to do so. Some of these strategies were system-based, while others were (often creative, usually collaborative and occasionally disruptive) work-arounds.

System blocks, Logistics, Referral pathways. Workarounds.

In aiming for respectful, responsive care provision that listened to women, participants described a dance between protocol and process in imperfect systems that did not permit easy adherence to principles of good care.

There was near universal frustration expressed by participants across jurisdictions and health services in relation to health systems and managing information and communications. RHD Program staff were particularly cognizant of the barriers created by poor articulation of clunky systems that did not talk with each other.

Systemic issues that participants saw as impacting on quality of care centred around health information disconnects and referral pathway blocks. There were multiple tiers of siloed information, multiple players across public and private sectors and multiple technologies (including legacy paper-based systems). There was often no real-time access to information. These blocks all had a direct impact on delivery of care and outcomes. This was particularly evident in primary health care systems.

The impact of these siloed systems had direct consequences for women and clinicians. Participants described experiences where referrals were not actioned, and results were not received. Women who had been diagnosed with RHD in a previous pregnancy had their diagnosis 'missed' in a subsequent pregnancy. Frustration where information was not shared among clinical services created another layer of information-sharing barrier, particularly in relation to cardiac services or where there was inter(or intra)-jurisdictional transfer. Legacy paper-based data systems alongside electronic created another tier of disconnect. Both inter and intra-jurisdictional systems did not talk with each other.

The multiple facilities providing multiple services were seen as creating data deficits where there was no way to extract information about treatments, history and outcomes.[SP03]

The call for better-integrated systems was directly linked to improved outcomes for women with RHD. Interviewees referred to up to seven data systems that they had to query, including (in remote Northern Territory) two primary health information systems within communities, although one participant \ indicated that they used the MyHealth system over these to circumvent communication issues [CL07]. As well as impacting on care services for a condition that demanded multiple providers, health domains and

geographical locations, the impact of such disconnected systems was more pronounced given the likelihood that many women particularly in remote and regional Australia often moved between communities. A study of secondary prophylaxis adherence found that there were typically three homelands and three locations where a person may go [CR01].

Concerns about privacy were referenced by one participant, who continued to describe that information-sourcing was personnel rather than system-based, heavily reliant on staff knowledge and their impetus to inform. Consistent with other participants, they concluded that there needed to be support for patient information to be linked into one system.

Work-around responses were common. An obstetrician in an Aboriginal Mothers and Babies (AMB) service described system and resource-based blocks as well as initiatives they had instigated to glean a better picture of clinical histories of women under her care. They went on to describe how the service acted as a conduit between community and tertiary centre, and highlighted the role of the AMB midwife in supporting continuity of care:

> *I've instituted a teleconference [between the Aboriginal Mothers and Babies service and communities]. And I go through every single friggin' antenate with two systems open on my computer in front of me, using the hospital record system and my system. So, with my system I can say, "Hey, hang on a second, let's go back" and hopefully I've got their notes.*
>
> *That means I can look at what actually happened in a previous pregnancy. Because your discharge summary is only as good as the doctor who wrote it and some of those discharge summaries are great but some are really, really, really bad. Or a discharge summary could land in the clinic post office box and just sit there. And then you'd get the screams from the clinic, "We haven't got a discharge summary".*
>
> *The advantage with the [Aboriginal Mothers and Babies] service is that I know we've got the midwife, who's just her midwife. We are tightening up our communication with the clinic so we close the loop about how things are followed-up. Also, we've made arrangements with outreach to access the results on their system. The private cardiac service exists in a bubble. So, the amount of time we spend just faffing around, ringing up and saying, "Can you fax the echo report because I've got a patient in front of me?" Then the other problem is that we don't have the staff in Medical Records: there are piles and*

> *piles of paper that have never been filed. They're two years behind. So, we have to devise complicated methods of making sure that we've got follow-up. The women, they often don't come into town with that information. Which means again we have to request it from the communities. Their communities keep all of their handheld records in the clinic because they lose it. You spend an awful lot of time doing detective work.* [CL06 Obstetrician in tertiary centre with high risk population]

RHD program staff also referred to joining the dots in making sure clinical information across services was not missed. One service was asked to place all RHD-related information – bicillin secondary prophylaxis history, previous echoes, cardiac reports – at the front of women's antenatal chart to streamline information and circumvent an IT system where information was not readily accessible.

However, the RHD registers, as distinct systems, were often not automatically queried. On visiting one hospital maternity unit to review case records for the RHD-P study, this researcher found that the AMOSS data collector midwife knew about the existence of the Register, but seemed unsure of how to access it. Another AMS midwife described her clinic's priority was to ensure clients didn't miss their bicillin:

> *If we can see that they're due for their LA Bicillin we just give it. And then our [RHD] coordinator makes sure that information goes through to the Register* [CL07].

Another clinician commented that better information should not necessarily require more information in each system: rather, that information needed to be accessed more easily and transparently, emphasising that

> *…with RHD in pregnancy, the focus should be on identifying it.* [CR02]

While participants were aware of the importance of timely diagnosis and early risk assessment in pregnancy, there was less reference made to screening. Those who did [CL03, CL06, CL07, CR01, CR02, SP02, SP03] spoke in general terms, although one participant in a high prevalence region expressed reservations about feasibility and buy-in for a condition that was relatively uncommon [CR01]. They did, however, point out that there should be a much lower threshold for performing echocardiograms.

Detailed detective and forensic work was thus needed to track down history. Apart from the enormous resource and logistics demands, such models illustrate the consequent impact on women, their family and community.

Most of those interviewed also referred to system-blocks outside clinical spheres that directly impinged on women's experiences of care in pregnancy, such as transport and access to services:

> *There's always a bit of nervousness around whether someone's referral is received; whether they get an appointment and then, whether they attend the appointment. There are issues for the health service, for the private cardiac service and for the client as well. A challenge for some of the women of course is that they'll have children to care for. And it's really hard… they can't travel away to have investigations or appointments because there's no-one they trust to look after their children. And also, if they're still breastfeeding but the child is over a certain age, the child's not allowed to travel with the parent. And so, we have patient travel restrictions which impact on women feeling able to travel for appointments. …We need more flexible and compassionate patient travel systems.* [CL03]

One long-term midwife considered that care pathways for women with RHD in her AMB service generally worked well, noting that part of this framework required an early recognition and responsive referral system where required resources or abilities could not be provided for a woman at their centre. [CL04]

However, several other participants described systems with trajectories of care that nudged the bizarre. A woman who lives in one remote community near the border of Northern Territory and Queensland may visit up to five different hospitals straddling two (possibly three) jurisdictions in the course of her pregnancy depending on her state of health:

> *The obstetrics checks are here [regional Queensland] so that's where they come for ultrasounds and so on. But the midwife for her community antenatal care is based in Alice Springs [Northern Territory]. And then they fly here to give birth. So, midwifery care is in one jurisdiction, the tests and birth are in another – about 1500km apart. Then, they'll go to Townsville if they're not well which is the referral centre. Or Brisbane [2000km away], depending on the severity. Or,*

> *if they are in Alice at the time and get complications, they'd probably go to Adelaide [South Australia].* [CL011]

What does birthing on Country mean for these women where they may be transferred to multiple locations during pregnancy and even antepartum in emergencies?

Another example of a service delivery disconnect in a high-prevalence region (with a vast remote catchment area) was illustrated by one midwife, where the private provider echocardiogram contract for her regional hospital dictated tests would be done on the weekend.

> *I'd say,* "Why are we getting girls in on a Saturday?". "It's 'cause they're having an echo"*. On a Saturday. Yep. And then we'd wonder why they didn't turn up for antenatal checks unless the RHD team ran around to pick them up.*[CL11]

One participant talked about her experiences of disconnects between primary health services, particularly Aboriginal Medical Services and the Aboriginal Mothers and Babies service. While acknowledging these were tricky conversations, they called for more open discussion and improved accountability to community, including financial aspects [CL10].

Not all blocks were system-based. The impact of severe weather created another layer particularly in remote communities:

> *We're meant to have three cardiology visits - I'm not sure if we will this year 'cause we've had cyclones which disrupted things a bit.*[CL03]

Like much of what was discussed, there was not one cause or reason – or solution – in addressing logistics-based gaps in providing care. Most participants referred to the mix of system-driven, personnel-driven, resource-limitations, structural and personal factors that factored when it didn't 'work'.

Human resources and workforce. Aboriginal health practitioners.

Skills and roles were discussed in the context of work structures that supported (or not) woman-centred care for women with RHD, particularly in relation to the role of Aboriginal health practitioners. Frustration at an inappropriate use (and lack of recognition) of skills as well as missed opportunities to work collaboratively was expressed by both Aboriginal health practitioners and non-Indigenous clinicians. This was evident in both hospital and

primary health care settings. [CL01, CL03, CL05, CL06, CL13, SP02, SP03]. Appropriate recognition in terms of remuneration was also referred to as a barrier to a skilled workforce structure [CL01].

> *And I think a really, really big part of all the health stories in the NT is that we need more Aboriginal health staff, in the hospitals and in the community health services. When we don't have them, we don't work well. 'Cause they're our connection, that cultural connection, you know, worth their weight in gold and I don't think it's appreciated how valuable their role is.* [CL03 Clinician working northern Australia urban tertiary centre and remote]

> *If you've got a stable clinic, you're going to have stable health workers. For example, Junction Point [an independently-run clinic] is incredibly stable. Their AHWs [Aboriginal health workers] organise things. We come out, they say, "This is the Women's Doctor's day". We'll do education related stuff and other things in conjunction with that, which is basically driven by the Aboriginal health workers. But, in a lot of places they use the AHWs as drivers. And they're paid really badly. And the education… we did it better in the '80s and the '90s.* [CL06 Obstetrician working in northern Australia urban tertiary with remote outreach]

Regulations defining work that can be performed by Aboriginal health practitioner vary by jurisdiction, with inconsistent recognition of training, skills and abilities. A more restricted work environment to that of the 1980s was described, where in one location health workers had assisted in delivering babies *"…but since then there's been a conservative push to keep the status quo. And that's sad"*.[CR02]

Situations were described where, on a busy clinic day, clients waited for their secondary prophylaxis Bicillin injection but finally left without receiving it. Indeed, this occurred with a young pregnant Aboriginal woman while in a community this researcher was visiting. Meanwhile, a senior experienced Aboriginal health practitioner with Certificate IV training drove clients to and from clinic.

> *[Our state] has cut back a lot of our stuff that we can do. And we only can do basic stuff and driving and that's it. No injections. You know, to me, it's a waste of talent.* [CL13 Senior Aboriginal health practitioner with Certificate IV]

This health practitioner continued to describe her role as *'being expert in pulling everything together and doing that juggling'* (of meeting client and health service needs) [CL13].

> *The inability of [Aboriginal] health workers to give injections is a perfect example of that. And when it was in place, the legislation only permitted health workers to give injections if they work a certain distance away from pharmacies. What a pharmacy has to do with it, who knows? And so, in some locations with a large number of people on our Register, even if they were qualified, they couldn't actually give the woman an injection 'cause the island's not big enough to exceed the distance requirements from the pharmacy.* [SP03 RHD Program Coordinator]

One obstetrician in a regional hospital described where the Aboriginal health practitioner was part of the regular multidisciplinary (MDM) meeting [CL02] but this was otherwise not referred to.

The lack of recognition and limitations of professional scope associated with the Aboriginal health practitioner role had consequent impact on the overall workforce structure, with one clinician commenting that their health service now spent more effort encouraging entry to medical and nursing education. [CR02]

Breaking down silos

As well as the barriers and complex networks described above, collaborative care processes were also described. Some of these were system-based, while others were solutions and workarounds that had evolved in response to system blocks. Participants spoke of brokering care [CL12] and 'being the expert on pulling it all together' [CL13].

Integral to the structure of Aboriginal Mothers and Babies (AMB) services is the partnership between the Aboriginal health practitioner and (Indigenous or non-Indigenous) midwife working together to provide care during pregnancy and the early post-partum. The shape of this naturally varied according to location and service. One midwife who had extensive experience working in remote communities described her experience co-establishing an Aboriginal Mothers and Babies service in a regional setting and how the slow process shaped working relationships and the evolving integrity of the program:

> *There was no (Aboriginal) health worker employed. So, nothing was going to happen in the community until that happened. Maggie didn't come on board*

until maybe three months later. Which was fine 'cause it gave me a chance to get to know the community, the systems, working for the government again so… I had time to do my own research. Then Maggie and I hit the ground running. You know, I think we both knew how it would work and what had to be done. And so we were fortunate… we were left alone to do what we had to do and over time, very slowly, built it up. I think the reason we were so successful is that we were so compatible. There was mutual respect. Certainly my 16 years in Aboriginal health set me up for this. Yeah. And I think that's why we ended up with such a solid program here. [CL10 Midwife]

The role of midwives and midwifery group practices in supporting streamlined systems and continuity of care was referred to by several participants. [CR02, CL02, CL03, CL04, CL06]

There's an outreach midwife and antenatal coordinator now. Before that we didn't even have a midwife clinic. And the outreach midwives have made a tremendous difference. And we've got midwifery clinics and they do a lot of the routine visits. That allows you to see more complex cases in a controlled fashion rather than copping the complex cases as emergencies. [CL02 Obstetrician in regional hospital with high risk RHD population]

Making sure the woman has her echo; cardiology review; having the LA Bicillin regularly if she needs it, would be the role of the midwife, caring for her, to oversee that. And one of the challenges can be that we have alternating midwives. And in the past, that's lead to, I think, a breakdown in continuity of care. But that's improving. [CL03 Clinician Northern Australia]

The various AMB program models for Aboriginal women and the RHD Programs in particular were often seen as a system-based 'glue' to bring together services in a woman-centred model. This was particularly evident in all of the AMB services (and some RHD programs) which were based on a partnership model of Aboriginal health practitioners and (Indigenous or non-Indigenous) midwives or program coordinators working together. This 'glue' helped join the dots in the nexus between family, community and across health services, with a resultant improvement in earlier antenatal and cardiac care, adherence to secondary prophylaxis and better outcomes for mother and baby including less pre-term birth.

> *Often it will be the older females in the family - Mum, Aunty, whoever, who are very quick to let Deb and the other health workers know when someone's expecting and get them in for the tests. So, generally it is a family affair. And then the women themselves generally start volunteering and asking questions. We'll either link them in with the maternity services or they'll come up and present themselves. And then the obstetric team, nursing and health workers, will contact our program. Because we work very closely with cardiology and make sure that echoes are up to date, and so forth. We usually deliver the bicillin [during pregnancy] ourselves because the women are more comfortable continuing within the same clinic setting.* [SP04 RHD program coordinator]

> *We keep up pretty well with the antenatal clients because the midwives are a bit obsessed about them and if there's a midwife in the community, you know you've got a person that you can call and talk to.* [CL11]

In the systems described above, breaking down silos was built into the model of care from the outset (and extended beyond pregnancy).

A participant from a tertiary high-risk clinic described the brokering role they played in matching the woman's desired birth plan to the clinical requirements as determined by the multidisciplinary team:

> *So, the doctors might say, "This is what we need", and I'll say, "Well this is what she wants". And then we try and make a care plan that fits in with both.*[CL12]

They went on to describe their conversations with women, stepping through the physiological impact of labour at different stages on her heart, and why possible interventions or monitoring may be required. Part of that education may be for the midwives, where, for instance, different regimens of Syntocin may be required. [CL12]

Other models and services were described by participants that supported collaborative processes and communications. The Indigenous Cardiac Outreach Project (ICOP) is staffed by a cardiologist, echocardiographer and Aboriginal health practitioner/program coordinator, providing services to regional and remote Queensland communities. At each community, the team worked alongside all clinic staff, providing echocardiograms, reviewing status of patients and referring where necessary. Aboriginal health practitioners

perform electrocardiograms together with ICOP staff, and reviews automatically included all service providers, often together with the client.

> *It's good to have that communication with the doctors and the Indigenous workers that come with them. You know what's happening from one visit to another. After they finished their clinics we all sit down, have a cuppa and just a normal conversation. They'll have a list of people and they tick and flick and then they'll explain everything to us. "We still need these people to stay here on this". You know. Things like that, so…I reckon it benefits the whole team.* [CL13]

Workarounds such as that described by CL06 above addressed information and resource blocks. Other workarounds required political strategizing, navigating health services territories and domains. One participant explained how they worked 'under the radar' with a peer in a different community health service to bypass strictures that prevented the two services (that had political and personal differences) collaborating in order to get the care women needed, while another detailed protracted negotiations between services to prevent a woman being unnecessarily being transferred 2000k away to give birth. These instances described complex responses, demanding ingenuity and ill-afforded time to achieve solutions.

A midwife at one of the communities where delivery of care crossed over jurisdictions also worked in the regional hospital, so they picked up some of the antenatal care in conjunction with the outreach midwife. In another community, the mobile women's health midwife nurse opportunistically diagnosed pregnancies and would do antenatal checks with the women with whom they had built up a rapport. [CL11]

Such methods of supporting a coherent care model were based on a mix of individual relationships, professional roles, commitment and serendipity.

However, as for much of the approaches that 'worked', strategies that were often mediated by individual work relationships and connections were less likely to continue in a sustained fashion because they were not system-based. One midwife described follow-up where women requiring transfer to an interstate tertiary hospital some 2000km away for high-level care and to give birth:

We used to have a midwife down in Adelaide who had worked here who would often go and visit women on her own if we told her about them. … But unfortunately, that doesn't happen now. [CL04]

"Layers on layers"

As the analysis progressed, the complexities of addressing care requirements for a chronic disease during the point in a woman's life where she was preparing to give birth became more evident. Apart from the clinical care needs and priorities described earlier - often with co-morbidities including diabetes, other cardiovascular disease such as hypertension, the spectrum of renal disease - the service delivery across multiple services and sectors and geographical locations introduced another layer of complexity. These services were provided against a backdrop of competing priorities in family and community, including access to healthy housing, food security, and social barriers.[CL03, CL05, CL07, CL10, CL11, CL12, CL13, CR01, CR02]

'…when I think back, there was so much else that was also going on that we were also focussing on, so, yes, RHD was not on the radar. I s'pose insofar as it impacted on their health renal function was more of a focus. And yeah, you know, whether they'd been beaten up...' [CL10]

Participants saw these complexities as demanding multi-tiered responses that did not have a single focus and could not have a single response:

We need to understand that nothing's a single strategy. Everything has to be multi-strategic and it's got to all eventually lead into the same end. I think all the work that I've done over the years has led to that development. Rather than just come in with a single focus. [SP03]

Competing priorities with other chronic conditions was referenced by several clinicians. Addressing other conditions particularly diabetes, hypertension and renal disease meant that RHD was often not 'on the radar':

RHD is a small problem by comparison. The biggest is diabetes. Then, after that you have your preeclampsia. Because there are other factors which relate to it, you know, like smoking, alcohol, lifestyle and violence and all those kinds of things. But rheumatic heart is - I wouldn't say it's a large part. We've had a few

> *who have had the valve replacements but not many. But it's the impact. And other conditions… just a few days back I had one girl with ischemic heart disease who needed stents. She is hardly 20.* [CL05]

Such experiences equally impacted urban settings, with one participant in a high-risk antenatal clinic describing RHD as being drop in the ocean to other chronic diseases: *'We're just overwhelmed with obesity and diabetes'* [CL12].

Parallels of the burden and system-level responses to RHD were drawn with other overall rare conditions such as Hansen's disease (leprosy), syphilis and tuberculosis. Participants who had long-term experience working in communities described the wane of diseases that had been endemic - with subsequent recent resurgence. [SP02, CR02, CL10]

> *Syphilis management was an obsession of the Health Department here in the '80s and '90s in a population where it was endemic. Until a couple of years ago there was little syphilis left. Now we've had an outbreak of syphilis again 'cause the central management of infectious diseases has declined, and there was little capacity to respond from regional management. So outbreaks are left to fester away for a while when they're small: it's only when they big someone starts to do something about it.*[CR02]

This participant commented that the regional public health units were unable to address such conditions due to large amounts of staff turnover, workforce capacity and lack of local knowledge; rather, a centralised management approach at the state level was required. [CR02]

On the other hand, a centralised system model in the context of a tertiary institution focus without individualised care was challenged by another participant, who argued that providers and systems could learn from chronic diseases such renal disease which (ideally) provided more individualised care…

> *Assuming patients are going to take responsibility for their own health care, swallow their prescribed tablets without having consulted with their family and having adequately provided information about the reason it matters – it's very very flawed.* [CR02]

Another Aboriginal health practitioner spoke of competing priorities in their generalist role in a small community:

> *But when you're in the smaller communities like that you're in the field, you've got to do everything. You can't focus on one thing. You've got to focus on everything that's happening and, you know, all the sick people, diabetes, everything. Which makes it really hard.* [CL13 Aboriginal health practitioner]

There were flow-on effects for communities contesting an overall poor health status. Young women needing transfer to tertiary centres may wish or require accompanying support from family members who themselves have significant health issues such as renal disease:

> *We have that generation in the 40, 50, 60 age group, quite a few who are renal impaired, on dialysis.* [CL05].

Complexities flowed through into appropriate care defined by age as well as cultural background and the woman's individual circumstances:

> *If you've got a 13 year old, yes, you've got to deal with the fact that they're a 13 year old. You've also got to deal with the fact that they're pregnant. There's layers on layers of complexity*. [CL06]

Targeting RHD as health policy priority within these complex landscapes remained paramount for one participant who described how effectively addressing this chronic disease in turn required addressing the 'causes of the causes' that underpinned so many other conditions:

> *The thing about RHD is that it's a sentinel disease for poverty, for household crowding and all the rest of it. So, … by focusing on that one disease, if you're able to make effective changes, there should be wider ramifications than just that. If you're holding up RHD as being the poster child for poor living conditions, and you're making efforts to improve those living conditions, that would spill over into better ear health and other health too.*[CR01]

Shared understandings

Understandings: for women's, for health services. Fear and knowing 'the full story'. In the shadows of knowledge

Perspectives of women's understanding of RHD, its impact in pregnancy and the role of health providers in supporting improved awareness varied among health services. Some maternity services spoke of having little focus on education and health literacy for women: *'I can't ever remember discussing RHD with a woman. Isn't that terrible.*' [CL11]. An obstetrician in the same regional maternity service concurred, assuming that women knew 'in lay terms' what was involved in, for instance, valvular heart surgery, but saying that they 'did not tend to go into it in much detail' [CL02], implying that cardiac knowledge was not their expertise.

By contrast, a midwife in a regional AMB described the improved awareness among Aboriginal health practitioners and midwives from workshops followed by sustained engagement and collaboration with RHD-P project staff. They felt it brought RHD onto the radar for both the AMB and that this awareness flowed through to conversations with women, community and other primary health services such as child and family health [CL08].

A physician in remote northern Australia spoke in detail about the scaffolded approach they took when a heart murmur was noted. They made the singing noise of the murmur, and explained that the 'extra noise' had to be investigated. The visual reference point of antenatal ultrasonography - which many women were familiar with – also helped in describing the cardiac echocardiogram that women were then referred to[CL03], as did the sonographer or doctor explaining the details of the echo as it was being performed[CL07]. Drawn pictures were also used to describe what may be happening with the valve [CL03].

Although consistent with best practice, participants described women regularly not receiving medical information in a way that made sense for them. [CL13, CL06, CL08, CL13]

> *Don't use that medical "jargon", they call it these days. And I always say to the clients, "If you don't understand, ask them what does it mean". But then some are so shy they don't ask at all. It's the brave ones that ask, "I don't know what that means. Can you tell me?" So I ask and explain it to the client later on*[CL13]

Where women had seen RHD program coordinators and educators through their secondary prophylaxis regimen they tended to 'know the story' better [CL03].

> *I think they do have an awareness that their heart function can be compromised with a pregnancy, I think, a lot of them do understand that. And, they want the best outcome for the baby and themselves. So, they're very quick to let you know that they're pregnant. And they're often very concerned about having the bicillin and if that will impinge on the baby.'* [SP04]

Other factors influencing awareness and knowledge included the community that women came from and whether another family member had been diagnosed with RF/RHD, with both supporting an improved awareness in general [CL07].

Another midwife spoke of women talking about 'holes in their heart' *'You investigate a bit more about that. "OK. So what do these holes look like?" Ask more questions'*. [CL08] They continued to describe how fearful women were when they were diagnosed: *'"These doctors", you know, "they come in and tell me I'm sick. What does that mean for my baby?".* Having someone with the woman who understands and can help tell the story such as a grandmother can make a big difference*. "Oh, so, I caught a bug and it's affecting my heart". Yeah. "And that's why I've had sore joints and other things going on." Yeah. "So I need that medicine to make me better so I'll go and have my medicine." Yeah.'* [CL08]

Clinicians commented on the impact of RHD for Aboriginal health practitioners themselves:

> *We had a health worker in that area with RHD. But it wasn't until she attended the [AMOSS RHD-P] workshop that she could relate to what was being said.* [CL10 Midwife]

> *An Aboriginal health worker in Flat Creek [a remote community] had a murmur noted which I also heard but other people who had listened to her heart didn't think it was anything. She became pregnant - 5th or 6th pregnancy. Then out of the blue, late in second trimester she went into heart failure. In Flat Creek. And we did get her to the tertiary centre [2,500 km away] and she did do alright and she did have the baby, a big healthy bouncing boy. But it really highlighted for me the danger of us not being aware of RHD in pregnancy.* [CR02]

Situations were described where patients made allowances for clinicians who did not 'get it':

> *Sometimes I think that the patients make allowances for us not actually understanding things properly, they're quite patient with our attempts at explaining stuff when we just come from such different viewpoints. For others, they are just frustrated by what we say or how we do things.* [CL09]

Providing effective care that promoted informed decisions on the part of women paralleled other health conditions:

> *Of course it's not just RHD, it's across other diseases. What's the evidence-base, the best care for that person? Getting that woman to explain what they think is wrong with them and what they want to do. Understand what their options are. Being able to provide the education to the family, as well as that person. So that they can support her in that process. Finding out what the barriers are and helping them access the services they need.* [CL08]

Participants spoke of dilemmas in providing care that fostered agency and autonomy in women whilst acknowledging an often complex social landscape that required intervening. One described taking a 13 year old in a remote community who was in an unsafe situation with a high risk pregnancy from a community visit back to the tertiary centre. *'That's intervening',* they acknowledged, reflecting on the ethical dilemmas and clinical and cultural imperatives of care. [CL06]

Workforce and women

The need for a workforce to meet women's needs required ability and expertise which extended beyond educational clinical knowledge. Several participants commented on gaps in effective recruitment and a skilled workforce and the impact that had on quality of respectful care in remote communities that were so disproportionately impacted by RHD:

> *You know, if you want to work in Antarctica you have psychological screening, you have training for a good two years prior to being selected. But if you want work in a similarly remote highly challenging environment looking after our most vulnerable people in the country, you just go. And you usually don't get any training. And you don't go through a vigorous selection process 'cause they're desperate for staff.* [CR01].

Resources and education

Several participants expressed frustration at the availability and appropriateness of educational resource materials[4], calling for an overall strengthened investment in resources with 'easily available, appropriate educational tools in language' [CL03].

While the resource availability has developed over the last few years, sharing understandings of that information is more challenging.

> *There are assumptions made by health providers along the lines of…,* "I have explained this"*. Well, we may have said the words. But what those words mean, in relation to that other person are two things. We're travelling along parallel pathways* [CL06].

This clinician continued with a scathing appraisal of where health education was dumbed down, rather than having conversations in a way that met women in the contexts of their lives and culture: *'And in these complex Western medical constructs we talk about things that are difficult to understand for someone who's got your own cultural view of the world and who speaks your language when they have no medical knowledge – let alone when they don't share that.'* [CL06]

Resources with high visual content were preferred … *'Watching movies and stuff like that, that'll stick to their brain. But, giving them stuff to read, it ain't going to work'* [CL13] and regular face-to-face group sessions were described [CL07] or proposed [CL13]:

> *You need to have a get-together with the clients who have rheumatic heart and sit them down all together, on a monthly basis, all together: I reckon it'd work really good. "Well, this is education time for us". Do it 'round the time when the doctors here so they can ask them questions about their heart. And, you know, everyone's all together and some of the questions, like, one person will ask, somebody else want to ask the same one. And have food and a cuppa and normal conversation. You pick up a lot of stuff in normal conversation. [*CL13]

In response to this interviewer quoting a community Elder who called for *'the full story. Don't simplify it. Tell it to us right*', an RHD coordinator agreed, but reflected that this took

[4] As well as the references in this section, see Discussion section in this chapter for updates since these interviews were conducted.

time, and often required unravelling underlying social issues. They saw this as integral to the model of care for women. [SP04]

Notable changes in the resources available in the three years since the first interviews were conducted were described. Two short films in English, Kriol and Burarra languages explore fertility, pregnancy and family for Indigenous women with RHD [120]. Significantly, these were written and produced by Aboriginal women in partnership with researchers, with content driven by a reference group of young and old Indigenous women about what they thought their communities needed to know, and directed by a young Indigenous film director. '*And we can see a young person's eyes light up as soon as they see it's in Language.'*[SP01]

Process and trust

Descriptions of effective learning and education were premised on trust, shared knowledge and respect: between the health team as well as women. The model of education in Aboriginal Mothers and Babies (AMB) services and the mutuality of learning between midwife and Aboriginal health practitioner was emphasised:

> *We (Aboriginal health practitioner and midwife] spent a lot of time together in the car and what not. She would hear me, what I was saying to people and lots of talking in the car… She learned my stuff and I learned a lot of her stuff. And it was really good because sometimes I might throw in a word that's too medicalised she would clarify, or if I had missed something, she would just come in and explain. It just worked very well.* [CL10 Midwife]

Service delivery shaped by women and community was seen as imperative to getting better outcomes:

> *We educate the women so that they can make choices. We're not the experts in their life. It's recognising that they're the experts. Quite often, we go, as health professionals, into a mother's home, with an agenda about what we need to do. But the AMB service is about going into the home and actually working with them about defining what their priority needs are – which is more likely to empower them to do something about it.*
>
> *Our service model has an Aboriginal reference group as part of it. Each model is tailored to each community and we can run the data and see what the problems are in each community. For instance, if there's a lot of drug and*

alcohol in that community, then we can think about, "OK. Talk to the Elders in that community and ask them how we tackle that". [CL08]

Similar partnerships were described by some of the RHD programs that have a coordinator (Indigenous or non-Indigenous) and Aboriginal health practitioner working together and visiting homes. One coordinator described the slow building of trust to work together and within the community:

Now we both go together to do a risk assessment in the home. I got cheeky once and asked if I could bring a student. And Deb (RHD Aboriginal health practitioner] just looked at me and said, "Sara, it's taken me long enough to get them to let you come to their home". [SP03 RHD Program coordinator]

Underpinning these approaches was the importance of time. Taking the time required with each woman required 'being brave'[CL06] against the pressure of getting through the client list, but also required skilfully juggling and using judgement to ensure women with often complex clinical problems did not leave because they had to wait too long, particularly in community [CL06]. There was the time needed, too, in accepting the benefits of these changes in the culture of health provision were not a quick-fix process. *"Long term, it's [improved baby outcomes] much better because we won't get the chronic diseases in 20 years. But it is a slow journey under this primary health care approach."* [CL08]

Reproductive health, interpregnancy planning, preconception care

Participants spoke of the way they approached reproductive health in the context of the woman's clinical status and life circumstances.

One AMB midwife described how they worked with women to make a decision related to conception planning and reproductive health, going through available options to work out what was best for each woman, considering factors such as chronic disease.

If the woman is under our services and postnatally she says, "I know that I can't have another baby. And I'm scared 'cause I had all these problems" we'll ask who she spoke to and follow it up. Because we're not the expert in this field. [CL08]

They saw her role as advocating for the woman, helping her understand the impact of choices she made and getting her into a service where she felt comfortable.

The importance of simply giving the heart a rest was stressed, with Implanon advocated to help space out pregnancies. [CL05]

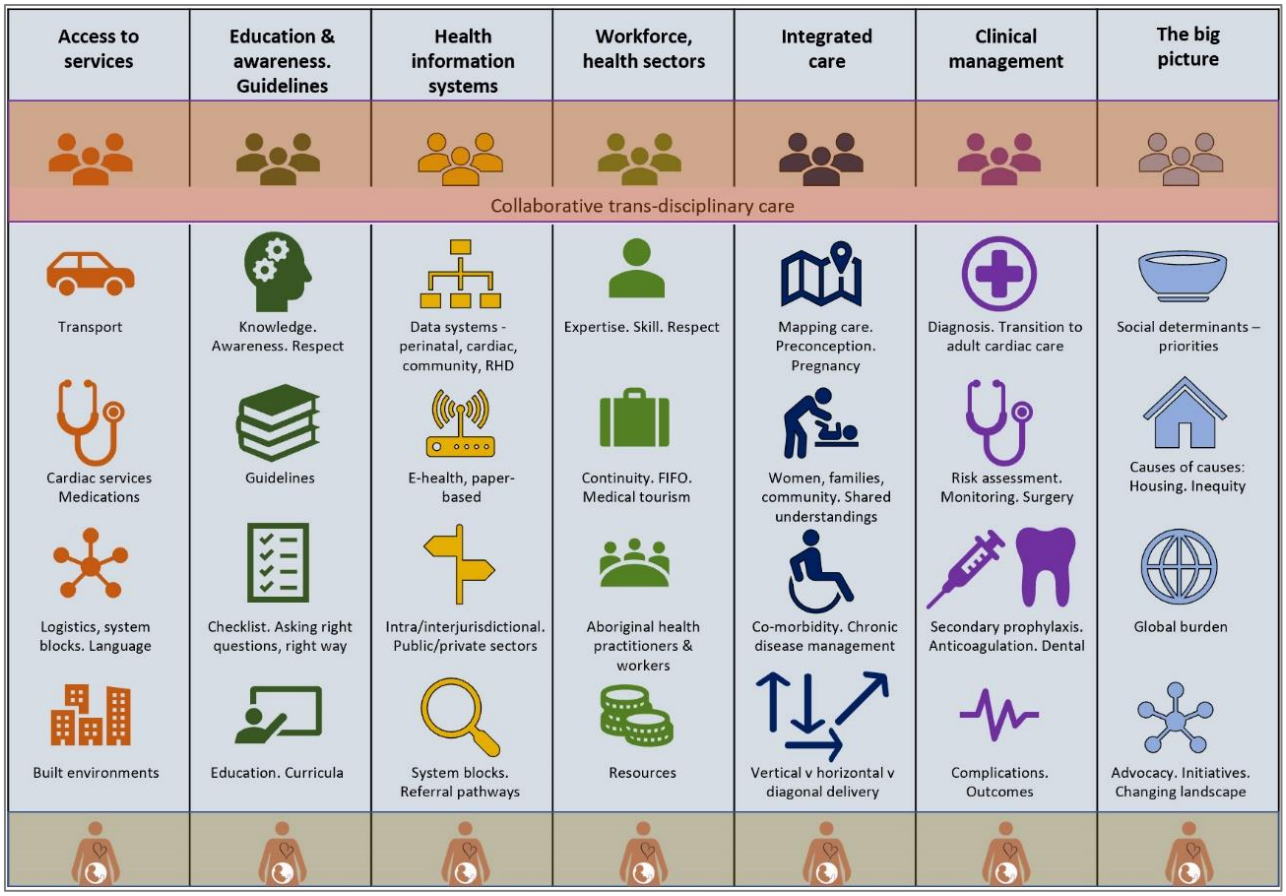
An RHD Program coordinator spoke of their service' evolving role in promoting conversations around contraception and planned pregnancy with women by health sites, which they saw as having 'room for improvement'. [SP02]

Frustration was expressed by one participant where women presented without having had required follow ups in between pregnancies. *'… and so you're making a decision if they need valvuloplasty during pregnancy, which would be better dealt with preconception.'* [CL09]

Overview

Drawing out the themes and subthemes described above, the concerns and perspectives of participants are summarised below in Figure 5-3.

Figure 5-3: Perspectives of health services: what matters for pregnant women with RHD



Discussion

During the thematic analysis of interview data, I found a broad range of factors which participants identified as potential barriers and facilitators to the care pathways for women with RHD. The research reflected the demand for a richer understanding of the contextual realities of providing care for pregnant women with RHD.

From the interviews, attributes of effective models of care included early diagnosis of RHD, good access to services, respectful pregnancy and RHD care delivered by a skilled workforce that worked collaboratively. These were associated with a mix of system-based and personnel-driven criteria.

The depth of knowledge, expertise and awareness of participants was overall high: not surprising given the purposeful sampling methods. However, those interviewed described an often-poor level of awareness and knowledge among their colleagues in maternity services particularly, although some also referred to a changing landscape of increasing advocacy, strategies, programs and research. These gaps have particular significance for RHD. Other research of clinician awareness related to women and all-heart disease highlights substantial deficits [347,348]. RHD carries added potential risk in the not-uncommon situation that previously undiagnosed disease will become symptomatic during pregnancy or postpartum: unlike congenital heart disease which is more likely to be known.

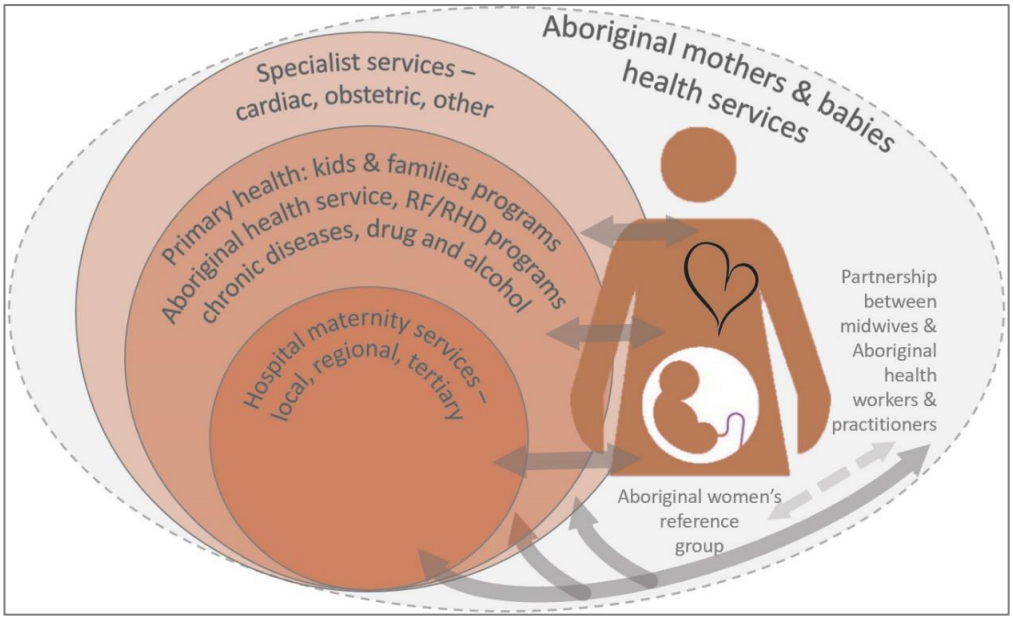
In identifying deficits in care, participants described a broad range of barriers to and some facilitators for the provision of optimal health care for Aboriginal and non-Aboriginal women with RHD-P that spanned health service access, workforce issues, clinical management, clinical information systems, and education.

Many of the issues highlighted in this study are endemic in the Australian (and other) health contexts. They address chronic disease, pregnancy and specialist care access, rural and remote health and Indigenous health issues and deficits. But there is a particular nexus for RHD-P that brings an imperative - and an opportunity - to improve care delivery for this preventable disease. While overall uncommon, the consequences of undiagnosed RHD-P can be catastrophic. Disjointed systems in maternity, cardiac, Aboriginal and primary health care in particular contributed to poor communications and challenged integrated care. Few participants were able to point to systems that supported shared understandings of the impact of RHD, particularly in pregnancy.

Illustrations of elements of care that promoted better outcomes often centred around the workforce. Particularly in remote high prevalence regions, staffing was often provided on a short-term locum basis which circumvented the continuity of care that was identified as critical. Much of the discussion about Aboriginal health practitioners and workers was associated with criticism and frustration related to recognition, capacity and scope of training. In particular, Aboriginal Mothers and Babies (AMB) health services [162,349-354] were described as providing conduits of care that promoted better outcomes for women with RHD-P (Figure 5-4). So, too, RHD coordinators and programs were referred to in some jurisdictions as providing these links.

Figure 5-4: Aboriginal mothers and babies health services: conduits of care

Linking community, primary health, tertiary, specialist services throughout pregnancy



Study findings are consistent with a qualitative study of secondary prophylaxis adherence, which pointed to high staff turnover rates particularly impacted on achieving improved adherence to medication regimens [133]. This review found a strong association between achieving implementation objectives at sites with characteristics of effective communication both within clinics and between other relevant bodies (such as the jurisdictional RHD control program); respectful, aware, knowledgeable engagement with

the community; and prioritising strategies to achieve objectives, particularly time resources for training and education [133].

Examples of breaking down these silos referred to a mix of system-level change, services and workarounds that were often personnel (and personally) driven: similar to this study of RHD-P.

Limitations

The exploration of health services' perspectives of models of care and issues related to pregnant women with RHD used single interviews (either in person or by phone). It could be argued that prolonged engagement through, for instance, 'walking the journey' with participants in clinic settings as they provide care, would minimise the risk of participant bias such as withholding information. On the other hand, a risk of prolonged engagement is researcher bias [282]. Recognising the resource burden on participants, single interviews were conducted. However, member checking was conducted to verify and clarify statements where required. Additionally, the other two studies undertaken (review of the AMOSS study and the narrative review of models of care) formed a data triangulation that served broaden perspective and provide a comprehensive picture. Analytic triangulation was provided through multiple coding. Peer debriefing support was conducted on an informal basis with members of the AMOSS RHD-P research team.

Participants differed greatly from each other in their professional, cultural and language characteristics, but the data obtained should not be considered to represent health service or professional views. There was considerable heterogeneity.

Interviews for this qualitative study were held over a four-year period. During this time the RHD political, policy and funding landscape evolved significantly. In response to the critical need for more (and more appropriate) resources and a change in scope, RHDAustralia strengthened its mandate to develop and disseminate 'evidenced-based resources to support health systems and health staff in their work, and advocate and provide culturally appropriate educational resources for people with ARF/RHD and their families' [114]. Central to the development of these co-designed resources has been their being informed and directed by the lived experience of those with RHD.

Conclusions

A complex interplay of systemic and cultural factors impacts the provision of optimal health care for pregnant women with RHD. Characteristics that underpin and support improved outcomes for mother and baby include early diagnosis, timely access to multidisciplinary care, effective health information systems and collaborative, respectful care. Providing this care requires a coordinated health system response that draws on and strengthens the capacity of the health workforce to address women's needs.

Chapter 6 Review of strategies and lessons learnt implementing a population-based study of rheumatic heart disease in pregnancy in Australia

List of presentations and publication from this chapter

This Chapter is the accepted version of the article which has been published in final form at DOI 10.1093/inthealth/ihy048 as detailed below:

Vaughan G, Tune K, Peek M, Jackson Pulver L, Remenyi B, Belton S, Sullivan, EA. ***Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia*****. Int Health. 2018;10(6):480-9.**

The chapter is written and formatted according to International Health Journal OUP guidelines. The article as published is copied in Appendix 6. Authors' contributions and signatures are listed in Appendix 7.

About this Chapter

Commencing in 2013, a two-year surveillance and descriptive study of the prevalence, management and outcomes of RHD-P across Australia and New Zealand was undertaken by the Australasian Maternity Outcomes Surveillance System (AMOSS RHD-P study).

This review that follows in this chapter describes the implementation of the AMOSS RHD-P study. It identifies challenges of surveillance in the Australian arm of the study, and describes the strategies developed to strengthen reporting by - and improve awareness among - health services.

The study extends the themes developed in chapters 4 and 5, and in turn highlights many of the issues of care that have been described in those chapters.

Abstract

Background: The global burden of rheumatic heart disease (RHD) is two-to-four times higher in women, with heightened risk in pregnancy. In Australia, RHD is found predominantly among Aboriginal and Torres Strait Islander peoples.

Methods: This paper reviews processes developed to identify pregnant Australian women with RHD during a two-year population-based study using the Australasian Maternity Outcomes Surveillance System (AMOSS). It evaluates strategies developed to enhance reporting and discusses implications for patient care and public health.

Results: AMOSS maternity coordinators across 262 Australian sites reported cases. An extended network across cardiac, Aboriginal and primary health care strengthened surveillance and awareness. The network notified 495 potential cases, of which 192 were confirmed. Seventy-eight percent were Aboriginal and/or Torres Strait Islander women, with a prevalence of 22 per 1,000 in the Northern Territory.

Discussion: Effective surveillance was challenged by a lack of diagnostic certainty; incompatible health information systems and varying clinical awareness among health professionals. Optimal outcomes for pregnant women with RHD demand timely diagnosis and access to collaborative care.

Conclusion: The strategies employed by our study highlight gaps in reporting processes and the opportunity pregnancy provides for diagnosis and re/engagement with health services to support better continuity of care and promote improved outcomes.

Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia

Introduction

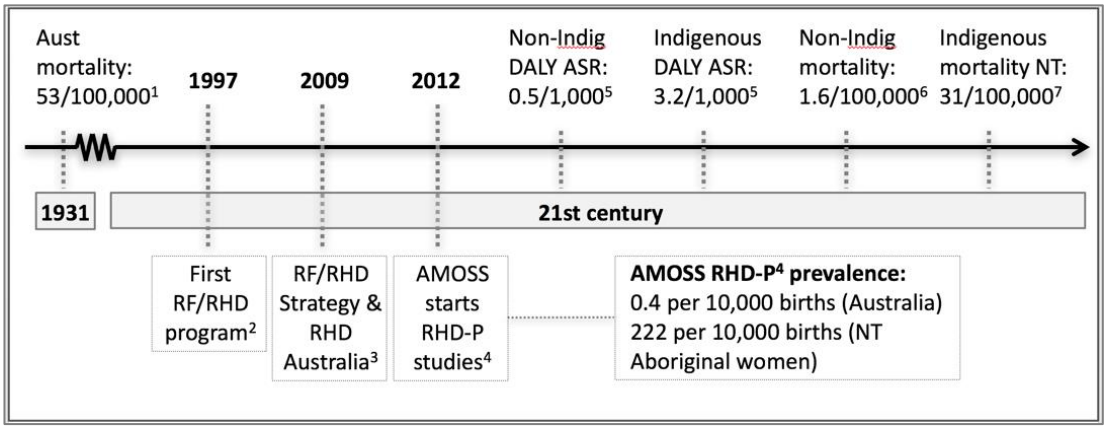
Rheumatic heart disease (RHD) is a condition of paradox in the high-income countries of Australia and New Zealand (ANZ): rare overall, but common in disadvantaged populations, with the burden of RHD among Indigenous and Pacifica peoples in ANZ among the highest documented rates in the world [101,110,116].

This non-communicable disease of inequity is a serious sequela of (usually repeated) episodes of the Group A streptococcus infection of rheumatic fever (RF), resulting in chronic damage to heart valves. Two-to-four times as many women as men are diagnosed with RHD [11,69,101]. Along with other high-income countries, the overall incidence of RHD in Australia dropped dramatically in the second half the 20th century (Figure 6-1). However, for Aboriginal and/or Torres Strait Islander women RHD is among the leading 20 causes of fatal burden (years of life lost) and among the 20 specific diseases contributing to the gap in total burden (disability-adjusted life years DALYs), with a rate ratio of 6.9 Indigenous compared to non-Indigenous Australian women [101].

An increased cardiac workload in pregnancy can unmask undiagnosed RHD and exacerbate clinical symptoms in women with known disease. The risk of poorer maternal and perinatal outcomes escalates, particularly for women requiring anticoagulant therapy and for women with mitral stenosis [202]. The burden of RHD in pregnancy is under-researched: the majority of studies examine severe disease in non-pregnant adults, all-cardiac disease in pregnancy or single-site studies, mostly in high-prevalence countries of sub-Saharan Africa, Asia and South America [239,320,355,356]. There are no known national population-based studies of RHD in pregnant women.

Commencing in 2013, a two-year surveillance and descriptive study of the prevalence, management and outcomes of RHD-P across ANZ was undertaken against a backdrop of increased advocacy and growing recognition of the burden – both global and country-specific - of RHD [19,126,357,358].

Figure 6-1: RHD prevalence and surveillance timeline in Australia



Notes: *1.'All-female Australia age-standardized RHD death rate' (Mortality and Morbidity: Cardiovascular disease: 20th century trends, 2002). Section 127 of the Australian Constitution excluded Aboriginals from official statistics until 1967. The first regular collection of Aboriginal health data began in 1957 (NT). Jurisdictional legislation did not allow for registration of vital statistics by race until after 1979[359]. 2. First Rheumatic Fever (RF)/RHD control programme established Northern Territory (NT) jurisdiction of Australia[113]. 3. Register and control programmes in three jurisdictions (five of eight jurisdictions by 2016); National coordination to develop resources and data collection system through RHDAustralia[360]. 4. 2012–2016: Rheumatic heart disease in pregnancy. National Health and Medical Research Council (NHMRC) project grant #1024206. An AMOSS study. 5. Australian Institute of Health and Welfare (AIHW). Australian Burden of Disease Study: Impact and causes of ill- ness and death in Aboriginal and Torres Strait Islander people 2011. Canberra, Australia: AIHW, 2016. BOD 7. 6. Table 1.1 'Underlying cause of death, All causes, Australia' (All-female age-standardized)[361]. 7. Table 12.5 'Underlying causes of death, leading causes by Aboriginal and Torres Strait Islander status, NSW, Qld, SA, WA and NT, 2010–2014' (All-female age-standardized). In: 3303.0 Causes of Death, Australia, 2014. ABS 2016.* **Notes:** *DALY, ASR, disability adjusted life years age-standardized rate; all rates except AMOSS RHD-P prevalence are for females with RHD.*

Objective

The AMOSS RHD in pregnancy study aimed to identify all pregnant women with RHD who presented at any of 284 participating AMOSS maternity sites across Australia (n=262) and New Zealand (n=22) during 2013 and 2014, for inclusion in a descriptive study of clinical backgrounds, models of care and cardiac, obstetric and perinatal outcomes. The objective of this paper is to identify the challenges of surveillance in the Australian arm of the study, and the strategies developed to strengthen reporting by - and improve awareness among - health services.

Materials and methods

Inclusion

The AMOSS study used World Heart Federation (WHF) criteria based on echocardiographic diagnosis of RHD[73] to identify women for inclusion. Pregnant women (20 or more weeks' gestation) were included if they had confirmed RHD based on their most recent echocardiogram report, or an historic echocardiogram where the most recent echocardiogram report was not available. Because a case was defined during pregnancy, women could potentially be included more than once during the 24-month study period. Women were excluded if they had a miscarriage or termination of pregnancy before 20 weeks' gestation, or if the baby's date of birth was outside the study period (1/1/2013-31/12/2014).

Reporting

The study was conducted under the umbrella of the Australasian Maternity Outcomes Surveillance System (AMOSS) [110]. This hospital-based surveillance and research system was established across ANZ in 2009 in response to the lack of national information on the incidence, risk factors, management and sequelae of rare and serious conditions in pregnancy. Volunteer site coordinators at participating AMOSS maternity sites (predominantly midwives and obstetricians - 'AMOSS data collectors') report and complete web-based surveys on nominated conditions over defined study periods. De-identified data are entered from case notes.

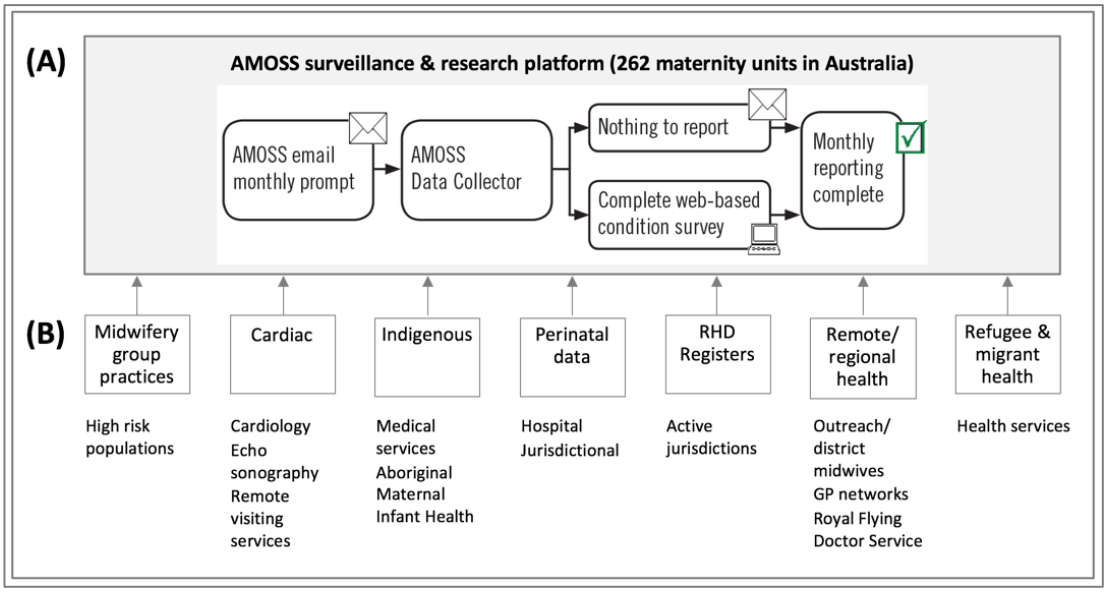
Negative reporting was employed: monthly emails were sent to AMOSS data collectors during 2013-2014 asking whether or not they had any women under their care with RHD in the preceding month. This established AMOSS reporting system (Figure 6-2) was supplemented by additional notification methods in the RHD-P study (Figure 6-3 and Table 6-1) although the AMOSS remained the central notification point. This extensive network of stakeholders helped optimise surveillance and strengthen awareness of the study.

Table 6-1: Confirmed cases of women giving birth with RHD reported according to source[5]

Confirmed cases: notification source	n	% of total 192 cases
AMOSS		
AMOSS participating maternity site data coordinator *(sole notification)*	106	55%
All Australian cases: AMOSS participating maternity site data coordinator or NT AMOSS coordinator *(multiple notification with other sources possible)*	181	94%
NT cases: AMOSS NT coordinator *(multiple notification with other sources possible)*	59	31%
Other sources *(non AMOSS coordinator)*		
Midwifery group practice / Antenatal care / Remote area or district midwife / Remote medical officer / General practitioner	22	11%
Obstetrician / Obstetric physician / Obstetric registrar	7	4%
Aboriginal Mothers and Babies (AMB) / Aboriginal Medical Service / Indigenous Cardiac Outreach Program	6	3%
Cardiologist / Cardiac nurse / Anaesthetic / Echocardiogram technician / Other specialist service	8	4%
RHD Registers (four active jurisdictions during study)	26	14%
TOTAL	**192**	

Figure 6-2: AMOSS surveillance and research platform

Usual AMOSS reporting cycle (a) supplemented by RHD-P enhanced network (b).



[5] (More than one source notification possible, percentages are of the total 192 cases)

Specific approaches to case ascertainment were developed according to jurisdiction and site. AMOSS maternity site data collectors primarily used perinatal data and health information systems. The AMOSS project coordinators from the study team queried additional systems in high prevalence regions including remote/primary health information systems and RHD control registers, using broader search terms of 'cardiac' and 'rheumatic' in the search criteria, and reviewing individual case notes. Similar enhanced case note reviews of perinatal data systems were conducted by the study team at three major tertiary sites outside NT. In the NT, which has the highest reported rates of RHD in Australia among Aboriginal women, a dedicated project coordinator conducted a validation study. Where the echocardiogram report was inconclusive in this jurisdiction, the actual echocardiogram was reviewed by a cardiologist.

In New South Wales (NSW), through consultation with Western NSW Aboriginal Maternal Infant Health Service and the 'ObstetriX/eMaternity' perinatal data working group, questions for women at the antenatal booking visit were revised to include detailed prompts on RF/RHD history, including whether the woman remembered having regular intramuscular injections (prompt for secondary prophylaxis) as a child as well as her heart history.

Following case confirmation, AMOSS site data collectors or the project coordinators completed web-based surveys, covering demographic, general medical/obstetric history, pregnancy pathway and maternal obstetric/cardiac and perinatal outcomes. We revised the study protocol during a two-month pilot phase to send echocardiogram reports directly to the study team for entry, in order to confirm case inclusion, reduce resource burden and achieve optimal consistency. In addition to the NT, AMOSS project coordinators supported case note review and completion of surveys in other high prevalence regions, particularly the Kimberley region of (north) Western Australia and far north Queensland, and/or where the resource burden meant that onsite staff required assistance.

Specific data items identified whether the woman had been reported in a previous pregnancy during the study period, as well as number of months since the last pregnancy.

Duplication was checked through a tiered process, including continual monitoring of the reporting database, probabilistic methods (using a series of concatenated data fields) and

checking with the AMOSS site data collector. The survey was completed at the site where the woman gave birth.

Ethics and consultative processes

Ethics approval requests under the aegis of the AMOSS were submitted to 32 Australian ethics committees and over 200 affiliated governance sites [110]. Subsequent amendments repeated this process as the study protocol was revised during the pilot phase. Access to sites for case note review and/or data entry directly by the research team was also approved where requested by the AMOSS site coordinator and for all NT. De-identified data were collected and reported at an aggregate level only. No consent was required for this quantitative arm of the RHD-P study. Aboriginal health services and Aboriginal Maternal Infant Health Services endorsed the study through letters of support.

Formal and informal consultative processes were established and continued throughout the research project in order to build awareness of the study (and more generally about the impact of RHD-P), optimise notification processes and provide avenues for dissemination of findings (Figure 6-5).

An Advisory Group comprised Australia and New Zealand investigators, the study team, and invited representatives from Aboriginal, cardiac and maternity services, and public health, jurisdictional, RHD organisations, and related policy bodies.

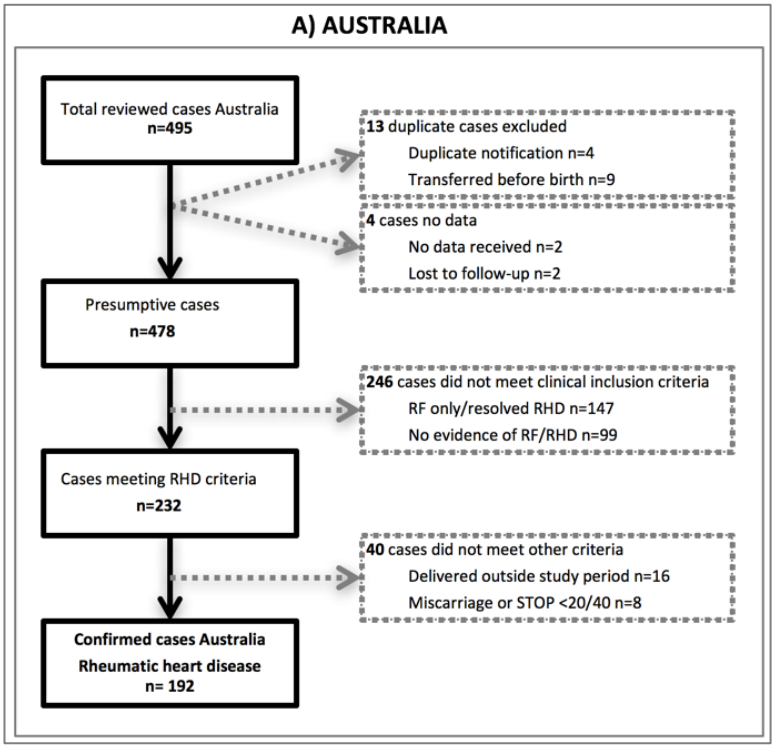
Results

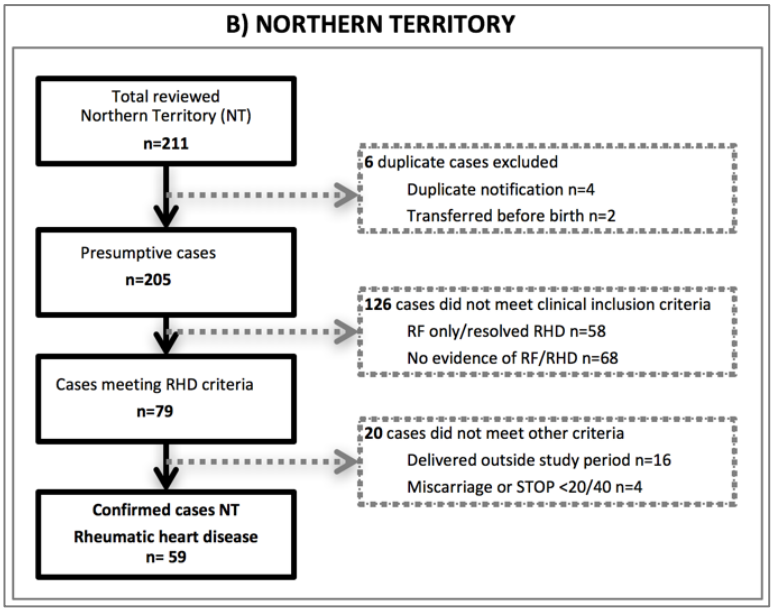
Surveillance

The AMOSS network of 262 Australian sites notified 495 potential cases (Figure 6-3). Of these, 246 did not meet the inclusion criteria There was no evidence of RF or RHD in 99 cases (identified predominantly through the perinatal data system search) and in 147 cases there was RF only or a valvular RHD lesion that had resolved. An additional 32 women gave birth outside the study period and eight women who miscarried or who had a surgical termination of pregnancy before 20 weeks' gestation were excluded. No data were received for four cases and 13 cases were duplicate notifications. A total of 192 pregnancies of Australian women with confirmed RHD (according to WHF criteria for echocardiographic diagnosis of RHD) who gave birth (≥20 weeks' gestation) were included in the Australian arm of the study.

Figure 6-3: Surveillance of pregnant women with RHD

(a) Australia and (b) Northern Territory 2013–2014





In the NT, where the validation study was performed, 126 of 211 cases reviewed did not meet inclusion criteria. Sixty-eight cases reviewed were excluded due to no evidence of RF or RHD, and a further 58 had either RF only or resolved RHD (Figure 6-4).

Data collection processes

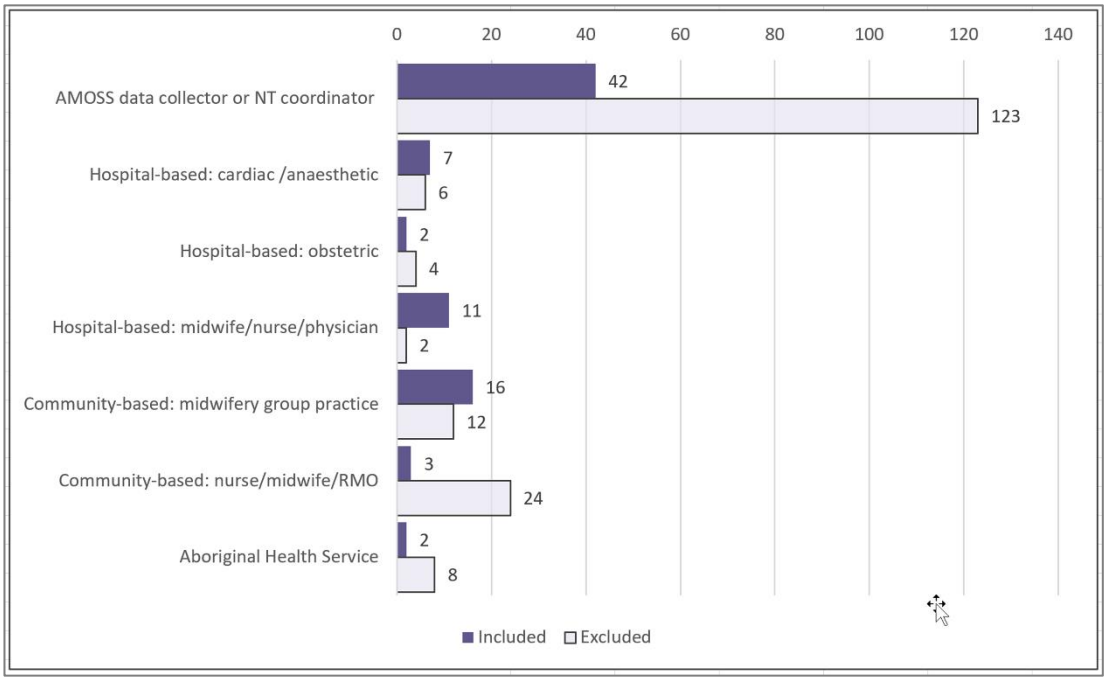
Notification and data collection processes varied according to jurisdiction, geography and site.

Notification

Cases were principally notified by data collectors at participating AMOSS maternity units or the NT-based AMOSS coordinator (181/192 94% eligible women), supported by other notifications such as Midwifery Group Practices, antenatal clinics and cardiac, community, remote, primary health and Aboriginal health services including the Indigenous Cardiac Outreach Project (Table 6-1) , with multiple notifications possible). In the NT, notification sources were documented by health group and location (Figure 6-4).

Figure 6-4: NT breakdown of reported cases by health group/location and inclusion

262 maternity units in Australia. NT breakdown of reported cases by health group/location and inclusion.



The RHD-P study partnered with jurisdictional RHD Control Programs to promote Register functions and purpose, with Registers assisting in notification. Additionally, RHD Program

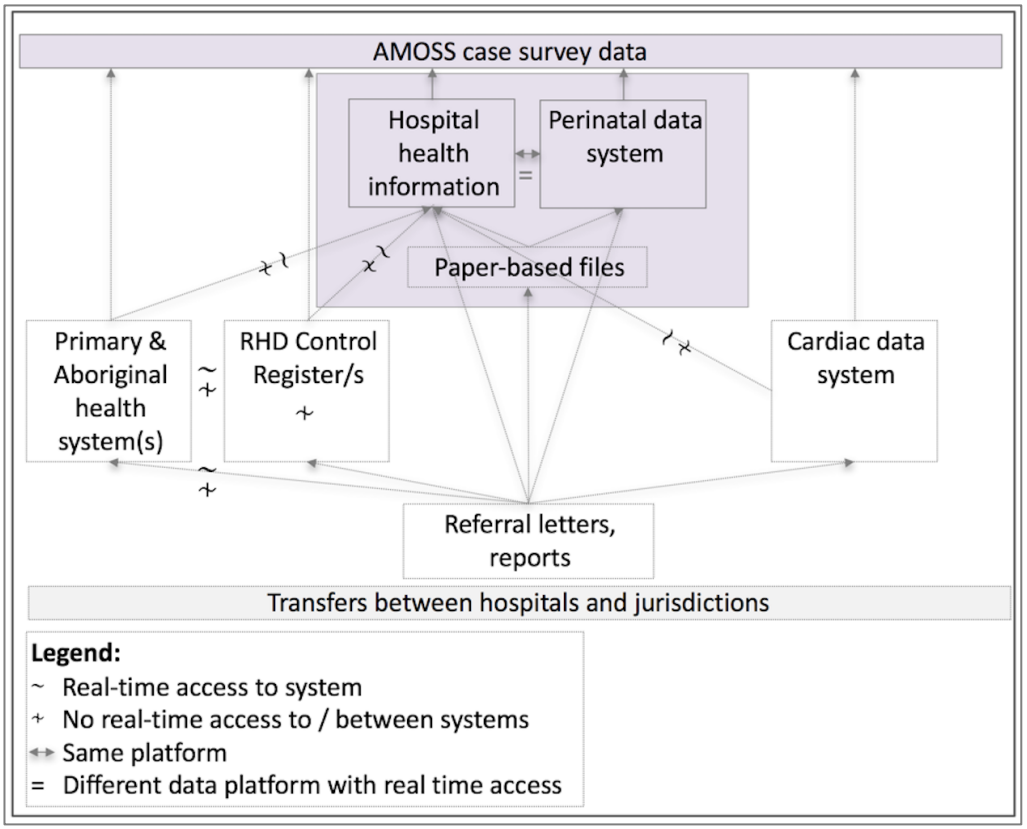
staff in some jurisdictions had developed care plans and notification prompts that aided reporting. One hundred and six cases (55%) were notified solely by the AMOSS maternity site data collector.

Where transfer of care occurred, a post-hoc review of data to ascertain any duplication of cases was conducted. Two women were lost to follow-up when they moved away in early pregnancy from the maternity site where they attended the antenatal 'booking' visit.

Data collection

Case note reviews entailed querying up to seven distinct information systems, in addition to reviewing paper-based files in sites where hybrid electronic-paper systems were active. In the high prevalence NT, the seven information domains included two discrete primary health care information systems that did not integrate with each other, and a privately run cardiac practice that provides services to the predominantly government-funded NT health system (Figure 6-4). Jurisdictional RHD registers are separate entities from each other, that may or may not articulate with the hospital and primary health information systems.

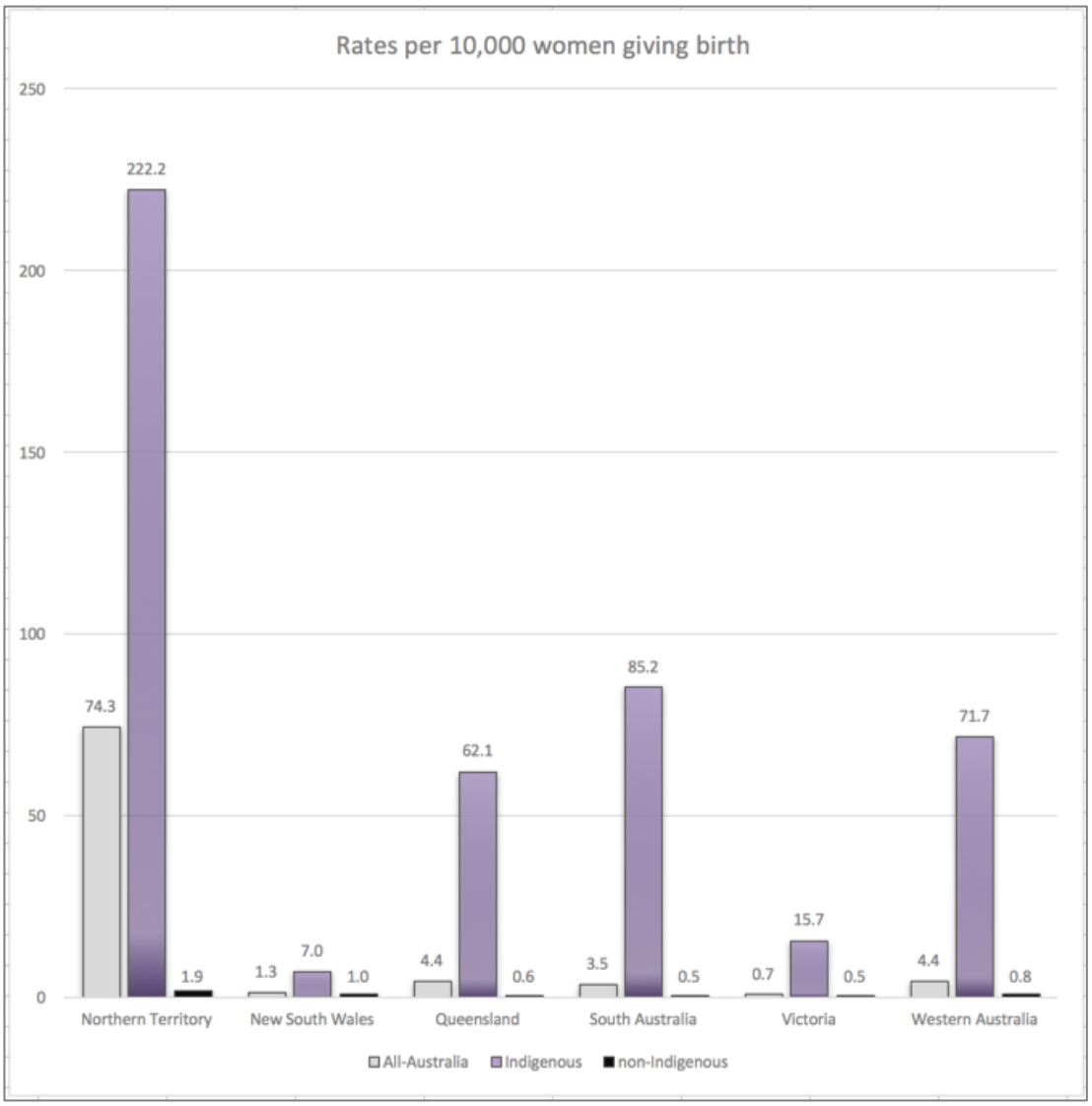
Figure 6-5: Data sources: surveillance and data collection



Prevalence

The Australian rate of RHD in pregnancy was 4.3 per 10,000 women giving birth. Of the confirmed 192 cases from six jurisdictions included in the study, overall rates ranged from 0.7 per 10,000 women giving birth in Victoria, to 74.3 per 10,000 women giving birth in the NT (Figure 6). There were no confirmed cases in two jurisdictions. Rates among Aboriginal and/or Torres Strait Islander women ranged from 7.0 per 10,000 women giving birth in NSW, to 222.2 per 10,000 in the NT (Figure 6). Overall, 78% (n=150) of Australian women included in the study were Aboriginal and/or Torres Strait Islander. The NT had 59 (31%) of cases, 99% of whom were Aboriginal women.

Figure 6-6: Rates per 10,000 Australian women giving birth with RHD by jurisdiction



Discussion

As well as providing multidisciplinary clinical care for women with known RHD, pregnancy provides the opportunity to identify newly diagnosed cases of RHD, re-engage with women who may have 'dropped out' of care as they transition from paediatric to adult specialist cardiac services, and provide language-appropriate health education that promotes a shared understanding of the impact of RHD and its implications in pregnancy [20]. However, our study highlights challenges to effective provision of care across several tiers, relating to case ascertainment, burden of reporting and health information systems. These factors impacted on a group of at-risk women with complex health needs and high rates of co-morbidities. Strategies developed to help support case notification underscored the value of an extended reporting network across health sectors, and reinforce the value of collaborative care.

Case ascertainment

High false positive rates (Figure 6-3, Figure 6-4) are consistent with preliminary active case finding work being undertaken by jurisdictional RHD Registers (personal communication) and the 'END RHD' Centre for Research Excellence [362]) which suggests significant over-ascertainment of hospital cases of RHD through ICD 10 coding, including valvular heart disease of unspecific origin. In the NT, where the validation study was conducted, a shortened case survey data collection was performed of the 58 women excluded from the study with either RF only or resolved RHD, with the aim of examining the burden on NT health services of being incorrectly diagnosed with RHD according to WHF criteria.

As well as over-reporting, under-reporting is assumed to have occurred of women with mild asymptomatic RHD without a case history or resultant complications who did not have an echocardiogram and were not reported. A number of women with a history of RF were reported but definitive diagnosis or exclusion of RHD could not occur due to lack of supporting evidence (by echocardiogram/cardiac review), including five women born overseas.

The AMOSS system is based on reported cases of women with RHD admitted to maternity units at 20 weeks' gestation or more: yet RHD is associated with higher rates of miscarriage as well as perinatal death[6]. In the NT review, an estimated 7% of otherwise eligible women

with RHD miscarried, or were recommended termination of pregnancy due to their cardiac condition. Thus, the true prevalence of RHD-P is higher than documented in our study.

Burden of reporting

The AMOSS system researches morbidities in pregnancy with an (overall) estimated prevalence of less than 1:1000 women giving birth – completed studies include, for example, H1N1 influenza, amniotic fluid embolism, vasa previa. Smaller maternity sites typically report an AMOSS condition once every several years, if at all. However, in this study, there was a significant burden on data collectors in high prevalence regions, where sites with 200-300 births per year had eight or more cases of women with RHD. This site burden was compounded by the work involved in information retrieval (Figure 6-4).

Health information systems

Our study found a lack of cohesion in standard data collection systems, including multiple systems, gaps in accurate case ascertainment and effective transfer of information between and across health services and jurisdictions. There were multiple sources of data, with some sites employing a mix of electronic and paper-based record systems, various electronic data systems in remote and primary health care networks that did not articulate, and gaps in sharing that information across networks including Aboriginal health services.

There was a significant variation in the amount and quality of information provided in echocardiogram reporting used to determine inclusion. Reports ranged from detailed records to handwritten excerpts. They were often not included in patient notes where women were transferred or had the echocardiogram performed by a private provider, or were unavailable for women born outside of Australia. Additionally, the lack of standardised reference values in echocardiogram reporting impacts on data integrity and clinical decision-making [363], particularly for pregnant women.

The World Health Organisation Roadmap for Action [364] notes that *"Only by disaggregating and analysing data can populations in need of health services be identified and included in informed policies and programmes"*. Our study highlights the need for improved RHD annual reporting and analysis at a national level with disaggregation by gender together with Indigenous identification and age.

The National Data Collection System (NDCS) (RHDAustralia-managed during the study period) is the central repository for the collection and reporting of RF and RHD data, yet it only reports on data from the jurisdictional Register-based control programs for RF/RHD (four out of a total of eight Australian jurisdictions during the study period) [114]. Pregnancy status was a recommended data item in the NDCS recommended dataset in 2011, however no jurisdictions currently monitor pregnant status within RHD Registers. This sharing of health information would be further strengthened by including pregnant status as a data item in RHD Registers.

Strategies to improve surveillance and build awareness

Streamlined, multi-tiered surveillance processes were established in our study to identify pregnant women with RHD across ANZ. While the AMOSS system remains a timely and reliable primary source of notification of (overall) rare conditions in pregnancy, these multi-tiered processes demonstrated that employing additional reporting sources can provide an effective surveillance adjunct [365], and a useful augmentation strategy to better inform the research of health risk, diagnosis, management and pregnancy outcomes of women with RHD. This is particularly significant for RHD, where a missed diagnosis or lack of awareness can have such a detrimental impact in pregnancy.

The call for multi-disciplinary care that straddles specialist cardiac and obstetric disciplines for pregnant women with cardiac disease is not new [202,203]. However, our study highlighted the benefit - and imperative - of building integrative, diagonal approaches to care [358,366] across all maternity, Aboriginal, and primary and public health care services (including RHD programs), in order to support an optimal pathway of care, particularly early diagnosis and assessment.

This more integrative approach to care is consistent in study findings and reports ranging from chronic disease in Aboriginal peoples, to international studies of maternal mortality [219,367], to global RHD initiatives. Many chronic conditions face similar hurdles in achieving effective health information and communication sharing, particularly at the interface of primary and hospital-based care and especially with remote Aboriginal and Torres Strait Islander health communities [368]. In their critique of Aboriginal health cardiac rehabilitation, Digiacomo et al similarly argue that inadequate referral systems, fragmented health information systems, and gaps in coordinated health services create significant barriers to

access for Aboriginal patients. They also point to inadequate resourcing of the Aboriginal Health Worker workforce: instrumental in supporting continuity of culturally competent care and making connections with various health agencies, also emphasised by Kelly[369]. Integration of RHD interventions with essential packages of health services including maternal and child health is consistent with strategies called for by RHD advocacy groups and initiatives at the global public health level [199,370].

Study limitations

Miscarriage or surgical termination of pregnancy often occurred outside participating AMOSS maternity units. Thus, accurate case ascertainment of all women with pregnancy outcomes <20 weeks' gestation was not possible across Australia in this cohort of women with a higher risk of pregnancy complications due to their cardiac status.

The study reports on one high-resource country and findings may not be as applicable in low-income countries. These findings do, however, parallel the overall health status of Aboriginal and Torres Strait Islander peoples in Australia. Indigenous identification was likely under-reported and thus the burden of RHD under-estimated in this population, highlighting the need for health professionals to ask all women whether they identify as being of Aboriginal and/or Torres Strait Islander descent.

Moreover, themes raised by our study related to RHD awareness among maternal and primary health services are consistent with conclusions and recommendations in studies of RHD in pregnancy in endemic regions, and in global RHD strategies.

Conclusions

Optimal outcomes for pregnant women with RHD demand timely diagnosis, access to integrative, appropriate models of care and health systems. The potential consequences of undiagnosed RHD in a pregnant woman demonstrate the value of enhanced reporting. Our study of the implementation of a population-based study of RHD in pregnancy identifies gaps in reporting and health information at a system level, as well as strategies developed to address these.

Our study findings call for implementation of pregnant status as a data item in jurisdictional RHD Registers, specific questions related to RF/RHD history built into perinatal data systems, improved real-time access to RHD Register information by hospital sites (and to

other Registers), improved education to support a more informed health workforce, and a more granular reporting of RHD in national data sets of prevalence and mortality, broken down by gender. The study highlights the need to promote greater awareness among the maternity sector in order to detect RHD, provide early monitoring in pregnancy and strengthen collaboration between disciplines in the care of pregnant women with this preventable condition.

Acknowledgements

The RHD in pregnancy study (NHMRC #1024206) could not have occurred without the ongoing support of data coordinators at all AMOSS maternity sites, as well as all those working in Aboriginal and remote/primary health care; Midwifery Group Practices; Aboriginal Maternal Infant Health Services, particularly Western NSW LHD; cardiac services including NT Cardiac and the Indigenous Cardiac Outreach Project; jurisdictional RHD Control Registers and RHDAustralia – and many other individuals and groups. Thank you.

Funding and other support

Gratefully acknowledged from the Australian National Health and Medical Research Council (NHMRC) project grant #1024206 and NHMRC Postgraduate Scholarship #11332944; University of Technology Sydney Chancellor's Research Scholarship; and END RHD Centre of Research Excellence, Telethon Kids Institute, University of Western Australia. Sincere thanks to the facilitators and participants of the Twelve Weeks to Publication Program funded by Faculty of Health, University of Technology Sydney.

Chapter 7 Beyond pregnancy:

Discussion, recommendations and conclusions

Introduction

In this chapter, I summarise the underlying rationale for the thesis and present a synthesis of the gaps and facilitators related to models of care for women with RHD-P. The chapter commences with an overview of study findings. I highlight how the data from the different studies have been integrated to address the research questions.

The imperative to improve care pathways for pregnant women with RHD is contextualised within the sustainable development goals (SDGs). This mapping process of findings against the SDGs reinforces an underlying theme that emerged from the studies: that effective health services for women with RHD commences in adolescence and takes a life-course approach, in which pregnancy represents a crucial point in time for re-engagement with health services, monitoring and provision of cardiac care.

The implications for clinical and community practice are then discussed in the context of health system strengthening and the quality improvement of care provision for women with RHD through a chronic care lens.

Recommendations are made based on the study findings, along with implications for practice, calls for further research and final reflections on my research journey.

Study framework, recap of aim and research questions

The overarching aim of this doctoral research was to investigate the factors that impact on optimal health care for women with RHD-P, with a focus on health services. Three studies were conducted to support this aim and answer the research questions below:

1. What guidelines and reviews inform or have relevance for models of health care for pregnant women with RHD? (Study 1)
2. How are attributes of care for women with RHD-P reported in the literature and how do these align with outcomes described in evidence-based guidelines? (Study 1)
3. What levels of knowledge, expertise and awareness exist amongst health professionals regarding care pathways for pregnant women with RHD and its burden? (Studies 2, 3)

4. What do health professionals perceive to be the barriers and facilitators to the provision of optimal health care for women with RHD in pregnancy? (Study 2)
5. What policies and strategies do health professionals suggest are required to more effectively meet the needs of these women? (Study 2)

Summary of findings: - drawing out themes with reference to studies

The thesis studies all examined models of care delivery for pregnant women with RHD from various perspectives. A systematic review of the literature was preceded by a review of guidelines which guided formation of a reporting framework with reference to RHD-P. This first study found key gaps in what was reported related to diagnosis, clinical measures, treatment and outcomes for women with RHD-P, weakening any ability to measure burden, assess trends and produce evidence-based recommendations. It called for a Delphi testing of the detailed reporting framework and adoption of a core outcome set to support data consistency, comparability of studies, strengthen knowledge and awareness of burden (clinical and social) and to improve benchmarking of care for women with RHD.

The second study was an examination of health services' perspectives of models of health care delivery for women with RHD-P. The qualitive research employed a thematic analysis of semi-structured interviews with health services to identify and explore themes related to what worked and what didn't for women with RHD-P. Underpinning the three key themes of system blocks and conduits; layered complexities; and understandings of disease and care management were a complex interplay of systemic and cultural factors that impacted the provision of optimal health care for women with RHD-P. Characteristics that support improved outcomes for mother and baby include early diagnosis, timely access to multidisciplinary care, effective health information systems and collaborative, respectful care. Providing this care requires a coordinated health system response that draws on and strengthens the capacity of the health workforce to address women's needs.

The third study was a review of the processes developed to identify pregnant Australian women with RHD during a two-year population-based study using the Australasian Maternity Outcomes Surveillance System (AMOSS). In probing many of the issues described in the previous two studies, it both exemplified system-level barriers to provision of optimal care and evaluated strategies developed to enhance reporting. The study highlighted the need to promote greater awareness among the maternity sector in order to detect RHD,

provide early monitoring in pregnancy and strengthen collaboration between disciplines in the care of pregnant women with RHD.

Discussion: developing a framework for coordinated responses to improve care trajectories for women with RHD

According to the World Health Organisation features of optimal health services should assure …: *"(iii) integrated, high-quality, patient-centred services at all levels from primary to tertiary care; (iv) a combination of priority programmes for health promotion and disease control, including methods for prevention and treatment, which are integrated into health systems; (v) information systems that produce timely and accurate data for decision-making"* [371].

Findings from my research suggest that aspects of care for pregnant women with RHD do not assure these features, and that they are often not delivered in ways that are culturally safe.

In the process of drawing together findings, two further overarching considerations became apparent. Firstly, that improving models of health care for women with RHD during pregnancy required improving care through the life-course, before and beyond pregnancy. Secondly, that the complex care pathways and determinants required an approach that could make sense of the multiple health sectors and policy arenas that influenced models of care.

Two frameworks were determined to help contextualise and present these responses in a structured fashion, as well as provide a lens which to understand and view the emerging themes from this study. The Sustainable Development Goals (SDGs), a blueprint to 'achieve a better and more sustainable future for all' [372] is an interconnected framework that addresses global challenges related to health, poverty, inequality, environment, prosperity, and justice with specific goals and targets to meet by 2030.

The second framework of a chronic care model also helped provide a grasp of the complexity of contributing determinants that impact on care and outcomes. Actioning change and quality improvement for clinical and community practice through a chronic care lens gave a useful 'so what' next step in health system strengthening and improved provision of care for women with RHD.

Viewing findings through the lens of these two frameworks provided direction to establish recommendations arising from my research. Each of the aspects reviewed below was considered from a global perspective with individual country references, as well as from an Australian perspective.

The imperative for investment: mapping models of care for women with RHD to the Sustainable Development Goals

Mapping against the SDGs and their targets provides a helpful framework of reference in considering gaps and facilitators. The SDGs are all ultimately interconnected and thus all have some significance for women with RHD [373,374]. However, the eight targets circled below (Figure 7-1) have particular relevance for women with RHD, with Targets 3, 5 and 10 related to health, gender equity and reduced inequalities of especial significance. The final SDG 17 related to partnerships is a critical enabler to support achievement of the other goals.

The investment required to meet SDGs, improve health service responses and make change for women with RHD demands more than fiscal investment. Whether it be addressing the health workforce, reproductive health, preconception care, antenatal care, access to services, or the 'causes of the causes' [122] of living conditions and housing [375], injecting funds must be done carefully and strategically, but in itself will not be sufficient to achieve optimal health benefit.

Successful initiatives to improve health service responses for women with RHD are based on building a shared understanding and partnership with women, community engagement, an integrative approach across sectors and disciplines and appropriate access to services. They require sustained political will, appropriate infrastructure and funding.

Figure 7-1: Sustainable Development Goals: a reference framework for women with RHD

(Adapted from the UN Global indicator framework for Sustainable Development Goals [24])

Poverty (**SDG 1**) underpins many of the primordial contributors to RHD: simply, RHD is a disease of poverty and inequity [370]. Socioeconomic conditions contribute to both its causative pathology [250] as well as poorer outcomes, which are further compounded in pregnancy. Its burden is profound in low-income settings , which not only include LMICs but also vulnerable populations living in high-income economies. Socioeconomic position, a proxy measure for poverty, is associated with increased pregnancy risk factors such as hypertensive disorders, diabetes and (according to country and location) kidney disease and obesity [248,249,251,290]. In pregnancy, these morbidities overlay and interact with the impact of RHD, contributing to poorer perinatal outcomes.

Access to nutritious food (**SDG 2**) impacts in several ways, but particularly in addressing the increased nutritional needs for pregnant and lactating women. This applied to at-risk populations in high-income countries as well as low-resource settings: the AMOSS study of Aboriginal women with RHD [20] showed the immediacy of providing food security can become a competing priority with less apparently urgent or immediate health priorities [20]. The **SDG 2.1** and **2.2 Targets** call to end hunger (measuring undernourishment as well as moderate or severe food insecurity in the population) has particular reference to the nutritional needs of adolescent girls, pregnant and lactating women [24].

Achieving the **Health SDG 3** to … *"ensure healthy lives and promote well-being for all at all ages"* requires substantial investment on several levels for women with RHD. **SDG 3 Targets 3.1 and 3.2** to reduce maternal and neonatal mortality is directly compromised by the impact of RHD. Three key markers are maternal mortality, the timing of the first antenatal visit and the frequency of antenatal visits.

Maternal mortality figures are strong measures of how well or poorly a country's public health system is performing. Target 3.1 aims to reduce the global maternal mortality ratio to less than 70 per 100 000 live births by 2030 [24]. Maternal deaths are usually avoidable and occur disproportionately in low-income and middle-income countries [376]. While there has been overall progress in reducing maternal mortality [377], maternal mortality ratios (MMR)s vary substantially, with a regional MMR of 546 per 100,000 live births for sub-Saharan Africa [377] which has one of the highest burdens of RHD. In South Africa, which - unlike the majority of its low-income neighbours - has national facility-based mortality audits [378], close to half of non-obstetric maternal deaths in South Africa are due to cardiac disease[262]. In its 2011-13 maternal deaths report, RHD accounted for 25% of cardiac-related maternal mortality, five times higher than congenital heart disease [367]. The impressive decline in maternal death in South Africa since 2009, from 189 to 135 per 100,000 live births in 2016[379], is associated mainly with the success of antiretroviral treatments for HIV-positive women [379,380].

Incomplete data sets, infrastructure and resource challenges all contribute to under-reporting of maternal mortality [243,381,382]. Misclassification of maternal deaths to other causes and a lack of granularity result in poor case ascertainment of maternal mortality due to RHD, where it is often simply not documented, particularly in low-middle income countries [243,343]. In higher-income settings where RHD is overall rare, it is often categorised under an all-cardiac category.

As well as strategies to improve care and reduce MMR, including late maternal death in low-resource settings [343], improved documentation is required with disaggregation of causative pathology to more accurately assess the burden of RHD and trends over time.

The 2030 SDG Target to reduce global MMR to less than 70 per 100 000 live births is thus unlikely to be achieved in the absence of rapid and sustained investment in maternal survival [383]. As with many of the strategies and initiatives that are associated with improved outcomes, reduced maternal mortality requires a sustained political will to implement

programs [376,377]. While there are country-specific contexts and complexities, enough evidence of proven interventions is available to guide key strategic choices and prioritising[18,384], with one review suggesting the lack of required decision-making to 'get on with what works' reflects a lack of commitment beyond rhetoric" [385].

As for RHD, while the MMR is substantially less overall in high-income settings (12 per 100,000 live births in developed regions) [377], there is strong variation according to demographics. The USA has large (and increasing) racial and ethnic disparities in its MMR[386]: 56.3 per 100,000 live births for non-Hispanic black women compared to 20.3 for non-Hispanic white women[6] [387].

Similar discrepancies are highlighted in the UK, where the risk of maternal death in 2012–14 was found to be significantly higher among women from black ethnic backgrounds compared with white. This report also identified a quarter of women who died in 2012–14 were born outside the UK; 46% of whom were not UK citizens [219]. Improvements to care were identified which may have made a difference to outcome in 64% of the women who died as a result of valvular heart disease in this report [219].

In Australia which has an overall MMR of 7.1 per 100,000 live births, it is 4.6 times higher for Aboriginal and Torres Strait Islander women [109]. These data likely under represent the impact for Aboriginal women: for example, the 2016 maternal deaths report did not include the NT, where 30% of Australian cases of RHD occurred in AMOSS RHD study [15], including

[6] Included in this study by Macdorman et al were the 27 US states and the District of Columbia that had adopted the United States standard pregnancy question by 2008. The overall MMR in this study was 25.4 per 100,000 live births for 2013-14, compared to that reported by Alkema et al for all-USA of 14 per 100,000 live births in 2015 (itself an upward trend from 1990).

377. Alkema L, Chou D, Hogan D, et al. Global, regional, and national levels and trends in maternal mortality between 1990 and 2015, with scenario-based projections to 2030: a systematic analysis by the UN Maternal Mortality Estimation Inter-Agency Group. *The Lancet.* 2016;387(10017):462-474, 387. MacDorman MF, Declercq E, Thoma ME. Trends in maternal mortality by socio-demographic characteristics and cause of death in 27 states and the District of Columbia. *Obstetrics and gynecology.* 2017;129(5):811.

one maternal death[332], and where a third of women giving birth are Indigenous [388]. Indigenous under-reporting persists [106]; and cause of Indigenous maternal death reporting is not disaggregated by state.

SDG 3 Target 3.4 to reduce premature mortality from non-communicable diseases cannot be met without reducing the global burden of RHD, which demands availability of essential medicines and improved access to cardiac intervention and surgery. Central to this Target is early diagnosis of symptomatic RHD, optimally prior to pregnancy but otherwise as early as possible antenatally to avoid a potentially catastrophic deterioration due to heart failure in severe RHD.

There is growing recognition of the importance of early echocardiographic screening of all pregnant women in high risk populations (Wyber et al., 2018, Watkins et al., 2018). A large Ugandan longitudinal study is in progress using existing infrastructure to screen pregnant women and follow outcomes (Beaton et al., 2016). This study found pre-existing cardiovascular disease (nearly 90% due to RHD which was unknown prior to pregnancy in all but 3% of women) was responsible for a substantial risk of adverse maternal outcomes in low-resource settings (Beaton et al., 2018).

More research on the methods, efficacy and benefits of screening in pregnant women in high risk populations is needed in the Australian setting. Concern at the persistence of late diagnosis of RHD-P in the Northern Territory of Australia has led to increasing calls for screening of all Aboriginal pregnant women in the Top End of that jurisdiction and the proposed development of a pilot strategy is in progress [389,390].

Regardless, there should be a low threshold for echocardiogram and cardiac referral in at-risk populations.

Access to appropriate sexual and reproductive health services as outlined in **SDG 3 Target 3.7** is a basic human right. Is also a physiological imperative for women with RHD to support informed – if often vexed - choices about child-bearing and planning. Choosing a contraception method must consider risk and efficacy from a cardiac perspective as well as the usual considerations of reproductive health provision in a safe, respectful environment [391,392].

Access to this care should begin in adolescence with transition to adult cardiovascular care, regular review in the context of current cardiac status [217]and continue throughout a woman's reproductive years.

There were notable deficits in how this was reported in studies referred to in the Chapter 4 systematic review, consistent with concerns expressed in Chapter 5 interviews with health services, as well as those in other studies [132,207,393] and reports [394].

The overall aim of preconception care (PCC) is to improve health status and optimise pregnancy outcome through identifying and reducing risk before conception occurs [394]. It addresses the maternal and infant mortality and morbidity that exists disproportionately in marginalised communities: highly relevant for women with RHD [207]. Preconception assessment includes a full history and examination, with functional assessment, a detailed echocardiographic study and possible exercise testing [196], with discussion about the optimal timing of pregnancy, to improve the chances of an uncomplicated pregnancy [395]. It assumes the required health infrastructure and skills are available for any recommended cardiac intervention or surgery pre-pregnancy – which are minimal or absent in many low-income settings [13], and suboptimal for vulnerable populations in high-income countries [192].

Universal health coverage (UHC) as outlined in **SDG 3 Target 3.8** includes the three main principles of equity of access; ensuring health services are of sufficient quality to improve the health of recipients; and protection of patients from financial risk resulting from healthcare access. It thus calls to address the deficiencies of service access that are so common for women with RHD based on financial, geographic and social constraints. They have particular relevance for women who require access to surgery or valvuloplasty [192,336,337,396], as well as pharmaceutics such as benzathine penicillin (BPG) and other cardiac medications that are safe during pregnancy and lactation [192,337] **(SDG 3 subpoint 3.B)**.

The 2019 announcement of $35 million under the Australian government Medical Research Future Fund to develop a vaccine for RHD [397] provides a welcome and overdue injection of funding into this neglected area, which will have global impact. However, it does not obviate the need to address the underlying causative factors that persist, nor the need for strengthened health systems and services to address the various levels of preventive care.

The significance of primary health care in care of women with RHD is also recognised in UHC, and is positioned by the World Health Organization (WHO) as one of its key pillars [398].

SDG 3 Subpoint 3.C - health worker density and distribution - relates to a strengthened health workforce and is pertinent for women with RHD on several levels, and includes investment in education, onsite training and funding. The importance of primary health and midwives in often resource-challenged environments is increasingly recognised – along with the need to fortify health systems and education to support its workforce.

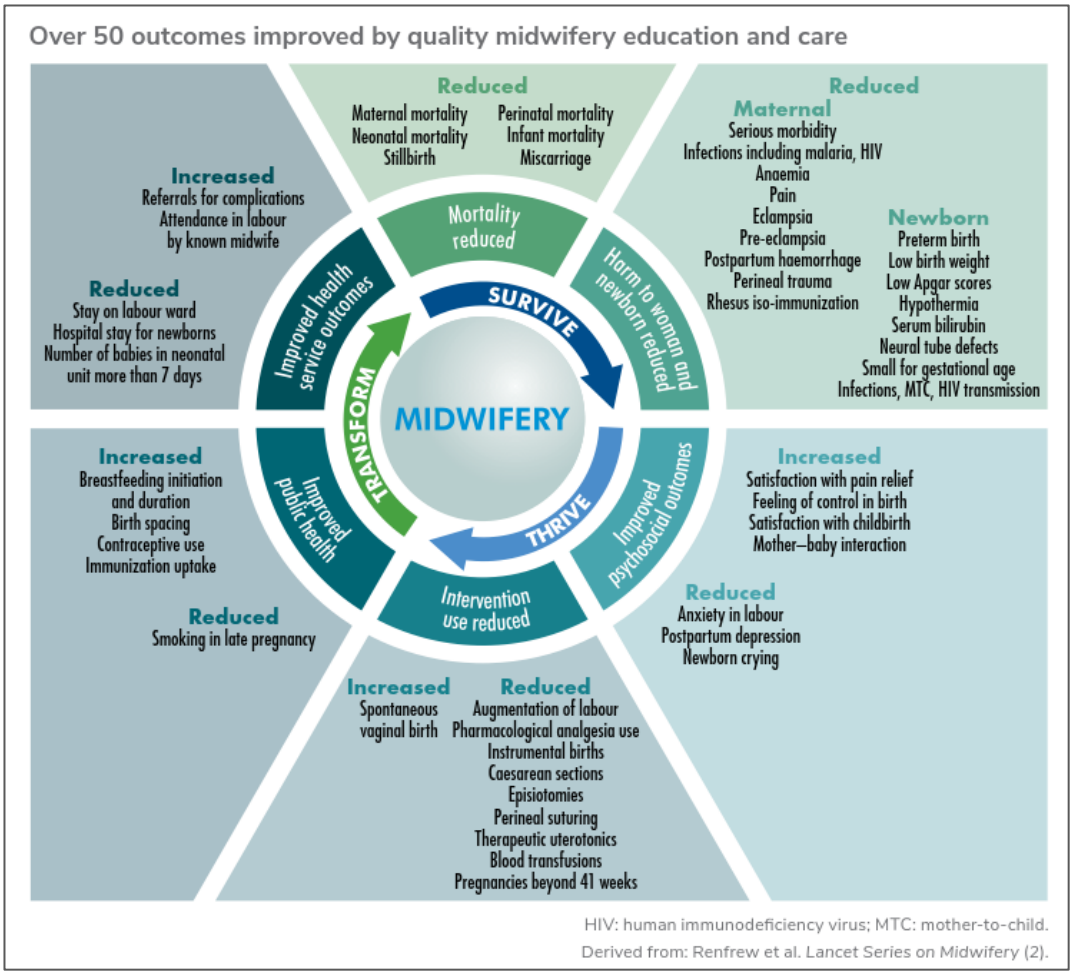
The primary health setting is usually the first point of contact women have during pregnancy, particularly in low and middle income settings [399]. Midwives, skilled birth attendants Aboriginal health workers, nurses and medical practitioners are uniquely able to provide essential services for women, providing a significant contribution to delivering on commitments made in the Astana Declaration on Primary Health Care [400] as well as SDG Targets. The benefits of a strengthened primary health and midwifery workforce are not isolated to RHD of course, but have particular consequence for this disease which can present so dramatically in pregnancy [343,401].

The scope of effective midwifery is broad. It extends beyond assisting childbirth to include family planning and reproductive health services and is delivered in communities as well as hospitals. Midwifery services are a core part of universal health coverage[402]. In settings where education is promoted to international standards and midwives provide family planning, midwifery can avert a significant percentage of maternal deaths, stillbirths and neonatal deaths [400,403]. With poor quality of care now argued to be a more formidable barrier to reducing mortality than insufficient access to care (Lancet Global Health Commission [404]), future gains towards achieving SDG goals demands improving this quality of care through more complex interventions, including midwifery, with an attendant scaling up required [400]. Figure 7-2 depicts maternal and perinatal outcomes improved by quality midwifery education and care.

The 'conduits of [integrated collaborative] care' that were described by participants in Chapter 5 as underpinning effective care trajectories for women with RHD is similarly emphasised [18,384,399,400,405,406], alongside quality training, regular supervision and with strong community engagement [399]. Reducing fragmented services for women and newborn infants to interdisciplinary and integrated skilled care and teamwork. Yet, under-investment in primary health and midwifery education, training and capacity-building persists. Consequently, the quality of education is variable. In many predominantly LMIC countries, midwife educators lack skills, supplies and equipment, with constrained access to clinical

sites for practical teaching [400,407-409]. Further, a lack of consistency in the use of the term 'midwife' means it is often not clear which health workers are trained to international midwifery standards [7] [400]. A similar lack of standardisation in names, training, and functions performed among skilled birth attendants (SBAs) exists, with one sub-Saharan Africa study finding a total of 21 different cadres of health care providers reported as SBAs [411].

Figure 7-2: Quality midwifery education and care: impact on outcomes
(from WHO Strengthening quality midwifery education for Universal Health Coverage [400])



[7] The 2014 Lancet series on Midwifery, in defining midwifery, notes the scope of care provided by qualified midwives may be limited by health system and cultural barriers, with overlap in roles and responsibilities between different health professionals. According to country and setting, aspects of midwifery care may be provided by *"obstetricians, family doctors, nurses, auxilliary midwives, community health workers or traditional birth attendants, or by inadequately-trained midwives, as well as by competent midwives educated to international standards, and by nurse-midwives who are trained both as nurses and midwives"*. It calls for a definition of midwifery which encompasses a package of care - regardless of provider. 410. Renfrew MJ, Homer CSE, Downe S, et al. Midwifery: an executive summary for The Lancet's series. *Lancet.* 2014;384(1):8.

Supporting this increased health workforce density and distribution requires a substantially increased health funding base in clinical education relevant to settings [24]. The recruitment, development, training and retention of the health workforce in low and middle income settings spans several levels and disciplines: from primary health through to tertiary care with specialist cardiac and maternal health disciplines. This was highlighted at both ends of the care spectrum in my research. Several studies reviewed in Chapter 4 discussed limited or no access to basic services, let alone cardiac interventions and surgery. However, models of care reviewed in this thesis together with the qualitative findings of Chapter 5 emphasised the centrality of a strengthened (and integrated) primary, Indigenous health and midwifery workforce in improving health services delivery for women with RHD.

In low-resource settings, access to care may be improved by training and enabling mid-level and lay health workers to perform specific interventions that might otherwise be provided only by cadres with longer (and sometimes more specialised) training. While this has obvious relevance in maternal health (for instance, with skilled birth attendants, primary health workers and midwives) [412], it can include other skills and training to support, for instance, echocardiographic screening [134,333]. Such task shifting strategies have achieved variable success. An evaluation of echocardiographic screening performed by briefly trained health workers in two high prevalence regions (Maningrida, Northern Territory and Timor Leste) found a high detection rate of moderate and severe RHD, but a lack of sensitivity for detection of mild RHD [413].

Such strategies are also dependent on varying local health, political, cultural and (according to workforce regulatory environments) legal contexts [341,412].

Critically, effective health workforce education initiatives in resource-challenged environments are community-driven and developed under an umbrella of national policy-driven changes incorporating poverty reduction strategies, gender empowerment, education and rural development initiatives [399].

While much of the above refers specifically to low-income countries, there were parallels in the Australian setting in my studies, with several participants calling for improved investment in appropriate education and training for Aboriginal health practitioners to work in partnership with clinical health providers. An emerging theme indicated informed and well-resourced antenatal care as provided by Aboriginal Mothers and Babies (AMB)

services in Australia aligns with optimal care for women with RHD, supporting early diagnosis, monitoring and minimising risk of complications.

Similarly, while midwifery programs in high income countries such as Australia are not impacted by the critical resource constraints of other regions, the imperative remains for a strengthened curriculum as well as a stronger focus on in-service education in high-prevalence regions for all health professionals. RHDAustralia is currently responding to this with the development of a midwifery curricula component in progress [121].

All such initiatives align with the call to appropriately resource and build capacity through the recruitment, development, training and retention of the health workforce. Investment is cost-effective, reducing the need for costly interventions and positively enhancing women's experience of care [400].

Linked to **SDG 3 Subpoint 3.C** health worker density and distribution is the imperative for improved access to appropriate education for primary and Indigenous health workers (and appropriate recognition of that education and training) as expressed in my Study 2 and referred to by **SDG Target 4.5**, which calls to eliminate gender disparities in education and ensure equal access to all levels of education and vocational training for the vulnerable, including Indigenous peoples.

Achieving Targets related to gender equality in **SDG 5** will have tangible effects in improving the lives of women with RHD - a disease that impacts twice as many women as men - as well as potentially reducing its occurrence. **Target 5.6** of ensuring universal access to sexual and reproductive health and reproductive rights requires appropriate information and education through a woman's reproductive years in order to make her own informed decisions regarding sexual relations, contraceptive use and reproductive health care in the context of her cardiac clinical status as well as personal preferences.

An implicit premise of **SDG 11** to make cities and human settlements inclusive, safe, resilient and sustainable is directly related to the **SDG 10** of reducing inequality within and among countries. Poor access to healthy housing is a primary determinant underlying rheumatic fever recurrence and RHD. Addressing over-crowded housing, lack of access to running water to wash people (and clothes/bedding) and impact of dust are key components of healthy habitat initiatives in Australia and New Zealand and have global relevance, with added impact in women [68,375,414-422].

SDG Targets 17.6 and **17.16** enjoin the global community to strengthen knowledge-sharing and cooperation related to science, technology and innovation across global socio-economic and political divides, as well as strengthen multi-stakeholder partnerships in support of the SDGs, in particular developing countries [24]. These underlying principles are exemplified in the World Health Assembly EB141/4, which secured a global commitment to eliminate RHD [17]. Similarly, **Target 17.9** calls for improved support for implementing effective and targeted capacity-building to support the implementation of the SDGs [24], a critical element of effective strategies and programs to reduce the impact of RHD.

Measuring any progress in meeting the SDG targets requires improvements in disaggregating data by gender, age and ethnicity) [24,423]. Deficits in data monitoring and accountability were highlighted in all three of my studies; achieving **SDG Target 17.18** directly addresses these gaps. The call for enhanced capacity-building support to developing countries to significantly increase the *availability of high-quality, timely and reliable data disaggregated by income, gender, age, race, ethnicity, migratory status, disability, geographic location and other characteristics relevant in national contexts* [24] (such as cause of death due to RHD, RHD diagnosed in pregnancy, and other recommended reporting measures specific to pregnancy, as outlined particularly in Study 1) will support better monitoring of burden and trends for women particularly as well as progress made.

Viewing models of care for women with RHD in the context of the SDGs both illustrates the complex multiplicity of factors that are part of its landscape, and gives a useful reference point from which to map against the findings of this research. This mapping process reinforces the imperative for a life-course approach, of which pregnancy represents a crucial point in time.

Implications for clinical and community practice through a chronic care lens

Examining models of care for women with RHD through the SDG framework shows the complex array of contributing determinants that impact on care and outcomes. Actioning change and quality improvement for clinical and community practice through a chronic care lens provides a useful 'so what' next step in health system strengthening and improved provision of care for women with RHD. It also provides an additional framework with which to understand and view the emerging themes from this study.

The mismatch between the needs of those with chronic care requirements and care delivery systems largely designed for acute illness is increasingly recognised. Health services

often do not address the needs for effective clinical management, psychological support, and information [424].

This second framework to help contextualise my research in a structured fashion is summarised below. Table 7-1 below outlines models of care for women with RHD through a chronic care lens. The model describes system changes associated with improvements in chronic illness care which are organised into an adapted framework to guide quality improvement. It adapts for RHD-P the extended model as defined by Davy et al, identifying patient driven health care case management and facilitated family support as key additional elements [425] to those in the Wagner chronic care model (CCM), which include mobilising community resources, health system improvements, enabling patient self-management, implementing care consistent with evidence and patient preferences, effectively using patient/ population data, cultural competence, care coordination, and health promotion [424,426,427]. Crucially, the Davy review suggests additional factors to the implementation of CCM elements may play a role, such as a work environment that supports reflective practice, with a sustained leadership commitment to the implementation of interventions and recognition of the importance of chronic disease care [425].

Table 7-1: Models of care for women with RHD through a chronic care lens

Adapted from Davy et al, extending the Wagner Chronic Care Model [424,425,428]

Health system improvement	**Delivery system design**	**Decision support**	**Information systems**	**Self-management support**	**Strengthened community support & linkages**	**Facilitated family support**	**Create supportive environments**
Key factors: • *Buy-in & strong senior leadership to advocate for improved systems* • *Quality improvement approach with goal-setting, rapid change cycles, & goal-attainment measures* [424]	*Key factors:* • *Clinical case manager functions delivered by experienced chronic disease clinicians* • *Support for self-management; follow-up to assessment & treatment adjustment* [424] • *Integrated approach* [425]	*Key factors:* • *Guidelines incorporated into registry, flow sheets & patient assessment tools* • *Related practice tools (eg disease severity assessments) link with treatment recommendations* [424].	*Key factors:* • *Information systems ability to produce treatment planning reports that serve as the visit record* [424] • *Broad-based integrated information systems include community data* [428]	*Key factors:* • *Shift focus from didactic patient education to support effective self-management* • *Individual/group interventions emphasise acquisition of self-management skills & patient empowerment* [424]	*Key factors:* • *Cost-effective way to tap into key services & links with relevant agencies* • *Enhance continuity of care & expand services or gather data useful to patient history* [424] • *Community-driven priorities and goals* [428]	*Key factors:* • *Holistic care approach* [425,429]	*Key factors:* • *System changes that acknowledge demonstrated connections between health and broader environmental conditions* • *Increase options available for people to exercise more control over their health and environments* [428]
Applicability for women with RHD							
System-level support for integrated care pre-pregnancy, antenatal and post-partum	Appropriately trained health providers underpinned by cultural competence, working with women & families	Easy access to guidelines and RHD program registers, with appropriate training to support awareness.	Align health information technologies with effective service delivery models [430]	Dedicated Aboriginal health practitioners & RHD educators to provide support & ongoing education	Aboriginal reference group informing priorities and health service delivery	Whole of life approach commences at adolescence with transition to adult cardiac and reproductive health care	Safe, healthy, housing
Address system & logistic blocks	Appropriately trained,	Consistent messages across	Improved sharing of clinical information	Preconception care: interventions,	Community driven priorities	Working with woman's partner &	Model of care supports Birthing on

Health system improvement	Delivery system design	Decision support	Information systems	Self-management support	Strengthened community support & linkages	Facilitated family support	Create supportive environments
	remunerated and recognised practitioners: Aboriginal health workers, skilled birth attendants, midwives, primary health workers	guidelines and practice tools (eg RHD Guidelines, CARPA manual)	between health services, sectors, jurisdictions, including primary, perinatal and cardiac care systems	surgery, secondary prophylaxis, anticoagulation considerations. Reproductive health: contraception, cardiac implications, planning pregnancy		family according to woman's wishes and as appropriate to age	Country or other culturally appropriate and respectful models according to country and population context
System-driven support for community-based programs such as Aboriginal Mothers and Babies with strong links to local as well as tertiary hospitals	Integrated multi-disciplinary care: cardiac, maternity and other chronic disease specialists together with primary care	Preconception care: interventions, surgery, anticoagulation considerations	Adequate medical records resourcing to migrate paper-based histories to electronic system	Antenatal planning and decisions re birth plan & clinical care – early, ongoing, review as needed.	Mutual accountability	Support to accompany woman – transport, accommodation	Recognise heterogeneity of communities and populations
Centralised system management structure and programs underpinned by individualised care	Identify RHD: consider antenatal screening in high risk populations. History, diagnosis, assessment, impact on pregnancy pathways	Integration with other chronic conditions. Holistic approach: dental check, secondary prophylaxis	Prompt post-discharge clinical communication and handover to all providers including primary care	Educational resources appropriate to population. Co-designed & produced with community	Prompt post-discharge clinical communication and handover to all providers including primary care	Support for accompanying children (eg if breastfeeding) to services	Built environments include birthing centres & clinical centres appropriate to population and community
Effective recruitment and training infrastructure to promote a skilled workforce	Antenatal cardiac risk assessment: identify risk and tailor birth plan with women accordingly	Discharge summary & plans		Scaffolded age-appropriate approach to learning and self-management, with review through the life-course	Trained interpreter services where required	Food security & supplement initiatives	

Health system improvement	**Delivery system design**	**Decision support**	**Information systems**	**Self-management support**	**Strengthened community support & linkages**	**Facilitated family support**	**Create supportive environments**
Adopt and implement core reporting measures for women with RHD	Individualised birth plan taking account of cardiovascular & obstetric issues			Improved systems to support pre-discharge review and conception planning with women and partner, family as wished		Kids and families: consider impact of rheumatic fever/RHD in children	
				Integrate with other services – RHD Program, other primary health and chronic care			
^^ Underpinning principles of woman-centred continuity of care ^^							

Recommendations

Drawing from my research and from the two frameworks of the SDGs and the chronic care model, a set of recommendations is proposed to address the care for women with RHD. They focus on the Australian context specifically. The recommendations are context-driven within Australia, according to sector, location and community. Effecting change on a large scale requires the scaffolding of sustained, respectful collaborations at a small scale[431]; the recommendations are premised on local initiatives being driven by local stakeholders – whether community, workforce, or women with RHD themselves.

The recommendations recognise that RHD jostles with many other competing health priorities at funding, policy and service delivery levels, underpinning the importance of aligning with other relevant initiatives where appropriate and feasible, to provide cohesion and encourage the collaborative approach that is so essential to effective care for women with RHD.

Table 7-2: Recommendations

Domains	Actions and strategies	Rationale / Comments
Working with women with RHD		
1) Ongoing advocacy and education: working with adolescent and young women to support shared understandings, promote agency and decision-making	Align with advocacy and action-based organisations, promoting translation of research findings and build on the evidence from other research: *RHDAustralia* Champions4change; *RHDAction* partnership; *World Heart Federation* initiatives; *Reach* technical support and policy translation; END RHD CRE; local initiatives.	Supported by Study 2 findings as well as those described in literature review: • That effective education and awareness is context-specific for a community and shaped by the women who have RHD.
Workforce		
2) Build capacity and workforce investment – Aboriginal health worker, midwife, skilled birth attendant and primary health as well as specialist care	Strategic engagement by peak bodies such as RHDAustralia, and research groups such as END RHD CRE with professional colleges, government bodies to impact	Supported by Studies 1 and 2 findings as well as those described in literature review: • That early diagnosis and optimal management of RHD

	capacity building in the health workforce. Particular emphasis in the primary health and community sectors, particularly Aboriginal health worker and midwives. Build partnerships and align with other relevant research and clinical practice groups to promote a cohesive influence on policy and strategies.	requires a better-resourced and skilled workforce • That improved links between community level care with district level services and appropriate referral pathways are critical to support continuity of care • That optimal care for vulnerable populations, particularly First Nation peoples, is promoted by a strengthened workforce. • That workforce development strategies and planning are developed in collaboration with relevant Colleges (eg in Australia Congress of Aboriginal and Torres Strait Islander Nurses and Midwives, Indigenous Doctors Association Australian College of Midwives) and other stakeholders.
3) Build on primary health partnership models that support improved collaborative maternal care for women at higher risk of RHD, such as the Aboriginal Mothers and Babies (AMB) services	Align ongoing partnerships and educational work with maternal health initiatives such as Aboriginal Mothers and Babies Services, leveraging collaborations with RHDAustralia. Other equivalent services globally.	Supported by Studies 1 and 2 findings as well as those described in literature review: • That principles of maternity care that promote best outcomes align well with those that promote optimal pregnancy care for women with RHD.

Education		
4) Curricula development: Aboriginal health workers and practitioners; midwives and nurses; obstetricians, physicians and undergraduate medicine	RHDAustralia-led midwifery curricula project (review in progress). Review current curricula in Aboriginal health practitioner; general and primary health nursing; and obstetric, physician and undergraduate medicine. Collaborate with global partners to share findings and resources.	Supported by Studies 1 and 2 findings as well as those described in literature review: • That effective and early care for women with RHD (particularly in pregnancy) requires a better informed and skilled workforce across all tiers and sectors. • In consultation with / under advice from professional groups and Colleges, and tertiary education providers, jurisdictional departments of health and points of health service delivery.
5) Inservice	Adapt and apply curricula components relevant to in-service. Support improved awareness and knowledge for all workforce, with focus on newly employed and short-term locum staff.	As above.
6) Guidelines	Improve specificity, granularity and scope of Guidelines that refer to women with RHD. Life-course approach that starts in adolescence and continues beyond (but focuses on) pregnancy. Share resources with global partners as relevant.	Supported by Studies 1 and 2 findings as well as those described in literature review, and the AMOSS RHD-P study: • The 3rd Edition Australian Guidelines incorporate substantially enhanced chapter on Women with RHD

		(under development, for release 2019).
Health information systems and communication		
7) Improved identification of pregnant women with previously diagnosed RHD	According to location and women's wishes, consider medi-tag alert such as bracelet. Include alert in medical record with direct link to protocols, guidelines and RHD Control Register record (where relevant).	Supported by Studies 1-3 findings as well as those described in literature review, and the AMOSS RHD in pregnancy study: • That childhood diagnosis of RHD may be missed in transition to adulthood. • That previous diagnosis of RHD may be missed in a first (or subsequent) pregnancy.
8) Delphi review of reporting measures included in research studies that include women with RHD-P	Conduct Delphi review of reporting measures with women and other global stakeholders, clinical providers across high prevalence regions. Data disaggregated by cardiac lesion, timing of diagnosis, and other recommended reporting measures for RHD-P.	Supported by Studies 1-3 findings as well as those described in literature review, and the AMOSS RHD-P study: • That there are gaps in what is reported in three framework categories related to RHD-P including of clinical reporting, risk in pregnancy and RHD through the life-course. • Core dataset proposed to more accurately benchmark care pathways, outcomes and burden of RHD-P.
9) Align health information technologies with effective service delivery models	Improved timeliness and sharing of clinical information between health services, sectors, jurisdictions, including primary, perinatal	Supported by Studies 1-3 findings as well as those described in literature review,

	and cardiac care systems (including RHD Control Registers). Improved articulation of legacy paper-based health records into electronic health systems. Strategic engagement in relevant data system reviews and committees, ensuring improved clinical reporting for women with RHD is considered in the overall context of health information technologies.	guidelines and the AMOSS RHD-P study: • That gaps in health information and communication systems create considerable logistic and workforce burdens, compromising women's care. • That historical diagnosis of RHD may be missed.
Models of care		
10) Transdisciplinary and cross-sectoral care	Improved collaboration, particularly working with community Ongoing work and engagement across all health sectors and settings, particularly maternal and child health in remote settings by groups such as RHDAustralia	Supported by Studies 1-3 findings as well as those described in literature review, guidelines and the AMOSS RHD-P study: • That optimal outcomes for women with RHD, particularly in pregnancy requires an integrative approach to care with early and regular review throughout pregnancy.
11) Underpinned by respect and women's choices	Country of Birth models of care or equivalent birthing principles according to population	Supported by Studies 1-3 findings as well as those described in literature review, guidelines and the AMOSS RHD-P study: • That care for women with RHD-P often did not meet their needs • That, in the Australian setting, the principles of Birthing on Country provide an

		appropriate transition to motherhood and parenting, and an integrated, holistic and culturally appropriate model of care for all [169]. • These approaches underpin care pathways that promote optimal outcomes and are relevant for all women with RHD.
12) Care navigator support	RHD care coordinator, with role defined according to location and RHD burden. For instance, a dedicated RHD role in very high prevalence regions such as northern Australia. Other navigators may be school-based, primary health or RHD program-based. In alignment with RHDAustralia and RHD jurisdictional program strategies and existing health programs.	Supported by Studies 1-3 findings as well as those described in literature review, guidelines and the AMOSS RHD-P study: • That a defined role is required to walk with women as they navigate through the life-course: adolescent, adult, through pregnancy and interpregnancy, supporting access to all health services, transport.
13) Cardiovascular risk assessment in all pregnant women, with or without symptoms particularly in high-risk populations for RHD	Align with initiatives such as RHDAustralia as well as Colleges and other stakeholders to continue advocating for best practice principles of antenatal cardiac care and risk assessment. At a minimum, low threshold for echo in high-risk populations. Consider echocardiographic screening of women in high-prevalence regions.	Supported by Studies 1-3 findings as well as those described in literature review, guidelines and the AMOSS RHD-P study: • That for women previously undiagnosed with RHD (or those where previous diagnosis was not picked up in the index pregnancy), the development of clinical symptoms in

		pregnancy was often associated with complications and poorer outcomes. • That for women previously diagnosed with RHD, early and regular review in accordance with risk assessment was associated with better outcomes.

Other reflections: No quick fixes. Cautionary tales

While research findings support specific recommendations to improve service delivery for women with RHD, there are cautionary lessons from other research and initiatives that should guide strategies.

Simply, there are no successful programs in the long-term that are not driven from the outset by local communities, supported by appropriate human and capital resources, with political will and buy-in at a policy level. It is unlikely that improved health care pathways for women with RHD - particularly during pregnancy – would be any different. Successful programs require sustained, ongoing effort, investment and dialogue at community, clinical practice and policy levels.

Lessons can be learnt from programs and strategies outside the direct sphere of women with RHD. One review of an evidence-based program to improve housing functionality in Indigenous Australia (the Housing for Health program [375,421]) describes lessons of reform from the perspectives of its program leaders, who wielded effective strategic-administrative interventions and were able to translate evidence to action. The success of this housing amenity reform, which was initiated under the directive of a community Elder[8] in the 1980s and premised on a simple set of healthy housing principles, was grounded in the ability of its protagonists to accompany their field research *"with other actions—good story telling, forbearance, repetition, and astute political advocacy—that created the conditions of possibility for the uptake and sustainability of their work"* [421]. Lea continues

[8] Kumanara Lester *(dec)*, a Yankunytjatjara Elder and activist from Anangu Pitjantjatjara Yankunytjatjara Lands in northern South Australia.

with a compelling description of their[9] approach which she argued was central to success in an Australian Indigenous service delivery setting: *"they were smart enough to understand the language of both paddock and boardroom, and irreverent enough to cut through sly bureaucratese and the community development platitudes of public health researchers with well-aimed, mutinous analyses. They were, as Fred Hollows might have put it, unburdened by the odour of sanctity* [432] in [421]. Again, the community-driven approach was critical.

So, research findings must be acted on with a sustained commitment to the long term. Guidelines become effective provider behaviour change agents only when they are woven into the fabric of patient care (Wagner et al., 2001). Similarly, the recommendations from this study and others require an integrated approach across multiple sectors and domains, always walking with the women who are affected by this disease.

Political will at all levels is critical to make change – from policy and agenda-setting to service delivery and of course all those that provide the services. This in turn requires a fundamental change in mindset: that RHD is not acceptable. Such efforts require sustained and determined attention: they cannot 'wax and wane' according to which political party is in power.

Indeed, the global efforts to control and end RHD are at a pivotal juncture. In tandem with the passing of the WHA resolution to end RHD globally [17], there has been an upsurge of renewed political will and engagement in several countries over the last decade. In Australia, there is commitment to develop a roadmap towards RHD elimination, with bipartisan commitment to end the disease. This has been paralleled by an upsurged focus to address the hitherto neglect of RHD-P – and also recognise the opportunities this critical time provides to support better care pathways and outcomes throughout the life-course. My research shows that Aboriginal health workers, midwives, nurses and RHD program staff in particular are critical partners in that process in Australia.

[9] Paul Pholeros *(dec)*, an architect, Dr Paul Torzillo, a local medical officer and Stephan Rainow, a local anthropologist with experience in community development projects.

Conclusion

This is the first known study to explore gaps and facilitators of models of care for women with RHD-P with a focus on health services. It examines the impact of health care systems and care trajectories and provides important recommendations for improving care for women with RHD, with a particular focus on the Australian setting.

Emerging from the study findings was a recognition that optimal care for women with RHD requires a woman-centred life-course approach that supports transition to adult care, considers surgery and other interventions in the context of reproductive health and preconception care, as well as pregnancy and postpartum care.

While the three studies focused on different aspects and issues of health care delivery, common themes were identified that span maternal, Indigenous, cardiac, perinatal and primary health systems. They all call for sustained policy-driven approaches that promote improved health information systems and access to health services delivered by a better informed and skilled workforce.

Underpinning these findings is the need for partnership models that support improved integrated care for women with RHD throughout the life-course.

For the 25% reduction in premature deaths from RF and RHD among individuals aged <25 years by 2025 and the 2030 Sustainable Development Goals targets to be achieved, maternal health must be better integrated into RHD strategies and RHD must be better addressed in maternal health.

Appendices

Appendix 1: Glossary and abbreviations

Glossary

Acquired heart disease (AHD)	Valvular (including RHD) and ischaemic heart disease
Aboriginal Mothers and Babies health service (AMB)	The Aboriginal Mothers and Babies health services are Australian jurisdiction-based programs where midwives and Aboriginal health practitioners/workers partner to provide culturally-appropriate holistic support and care throughout pregnancy. They are known by various names across Australia: NSW Aboriginal Maternal Infant Health Services (AMIHS); SA Aboriginal Maternal and Infant Care (AMIC) and Aboriginal Family Birthing Program; NT Midwifery Group Practice; QLD Mums and Bubs.
Australasian Maternity Outcomes Surveillance System (AMOSS)	A five-year NHMRC-funded (#1024206) research project (2009-2014) that conducted surveillance and research studies of specified maternal morbidities across nearly 300 participating maternity sites in Australia and New Zealand.
AMOSS RHD-P study	A four year (2012-2016) National Health and Medical Research Council (NHMRC)-funded (#1024206) [Australia] and [New Zealand]: population-based descriptive (Australia and New Zealand) and qualitative (Northern Territory) studies of the impact of RHD in pregnancy
Cardiac disease (CD)	All cardiovascular disease including congenital, genetic and acquired
END RHD Centre for Research Excellence (CRE)	The END RHD CRE is an Australian National Health and Medical Research (NHMRC) funded centre that focuses priority research projects to achieve the singular target of producing the Endgame Strategy. The projects run across several disciplines of research including epidemiology, economics, biomedical sciences, clinical practice, health services research and social sciences, with a special focus on engaging the RHD community

	and documenting the experiences of those living with the disease. The CRE's investigators are working closely with individuals and communities living with RHD to fill knowledge gaps, working towards producing a costed, step-wise strategy to end RHD as a public health priority in Australia. The strategy - to be delivered to the Commonwealth as the RHD Endgame Report in 2020 - will include an 11-year plan to achieve disease control by 2031, reducing the incidence of ARF and bringing the prevalence of RHD for Aboriginal and Torres Strait Islander Australians down to the same level as non-Indigenous Australians. http://endrhd.telethonkids.org
Gini	A measure of statistical dispersion that represents the income or wealth distribution of a nation - most commonly used measurement of inequality.
Infective endocarditis (IE)	Infection in the heart valves or endocardium, usually caused by bacteria entering the bloodstream and infecting the heart. Although rare, people who have heart conditions such as RHD or who have artificial heart valves are at higher risk
Mechanical heart valve (MHV)	A manufactured artificial heart valve. It is the longest-lasting type of replacement valve, but carries the highest risk of thrombosis.
National Rheumatic Heart Disease Data Collection (NRHDC)	The NRHDC is an Australian Institute of Health and Welfare (AIHW)-housed data collection. It comprises information from jurisdictional registers of notified cases of Acute Rheumatic Fever/Rheumatic Heart Disease (ARF/RHD). This de-identified population-based collection, was initiated by RHDAustralia as part of the Australian Rheumatic Fever Strategy 2009, funded by the Australian Government Department of Health. The data collection enables reporting on the Key Performance Indicators (KPIs) of the Strategy, reporting local level data to jurisdictional registers and supporting relevant data development activities [433].

Percutaneous balloon mitral valvuloplasty (PBMV)	Interventional procedure to dilate the mitral valve in the setting of rheumatic mitral valve stenosis.
Preconception care (PCC)	Comprises counselling and the provision of biomedical, behavioural and social health interventions to optimise the health of women and their partners prior to pregnancy and improve health related outcomes for themselves and their children. Incorporates a package of promotive, preventive and curative health interventions shown to be effective in improving maternal and child health
Prosthetic heart valve (PHV)	May be mechanical, bioprosthetic or homograft
Reach	*Reach* is a technical support and policy translation initiative to amplify rheumatic heart disease control efforts locally, regionally and globally. Aims to identify, describe and disseminate solutions for RHD and to reduce burden on vulnerable populations around the world. Reach partners with a broad range of stakeholders - including clinicians, other disease communities, academics, funders, governments, industry and people living with RHD - to achieve our common goals. [68]. Founding partner of RHD Action. http://www.reach.org
RHD Action	A coalition of global organisations leading the movement to reduce premature mortality from RHD and contributing to both the World Health Organization 25X25 goal and the World Heart Federation. 25x25<25 goal. The founding partners of RHD Action include Medtronic Foundation, the World Heart Federation and Reach. Working to establish a scientific and technical support community available to all countries, draw on this technical knowledge to advocate for policy change for better heart health, support and empower all people living with RHD and to foster multi-sectoral partnerships to support and sustain the global movement. www.rhdaction.org
RHD Register	The RHD register allows the surveillance and monitoring of ARF/RHD including administration of secondary prophylaxis, thus providing accurate data to assist in the monitoring of patient outcomes

	The first RHD register in Australia was established in the NT (1997) and, under the auspices of RHDAustralia is now being established throughout other participating states (currently QLD, WA; SA, NSW).
RHDAustralia	An Australian Federal government funded centre established in January 2010 under the Menzies School of Health Research, in partnership with James Cook University and Baker IDI. The scope of RHDAustralia has shifted since 2017, when responsibility for the national RF/RHD data collection that it had been coordinating moved to the (national government) Australian Institute of Health and Welfare. The centre now has three key mandates: updating, disseminating and integrating the Australian Guideline for the prevention, diagnosis and management of ARF/RHD; developing and implementing evidence based education and training resources for health professionals, people with ARF/ RHD, their families and communities; and supporting health systems to achieve evidence-based best practice for focused prevention activities in high-risk communitieswww.rhdaustralia.org.au
(Acute) Rheumatic fever ((A)RF)	An autoimmune response to the bacterial infection caused by Streptococcus A
Rheumatic heart disease (RHD)	Damage to the heart valves and muscles as a result of (usually multiple episodes) of rheumatic fever.
Secondary prophylaxis (SP)	An antibiotic regimen of (usually bicillin injections) given every 21-28 days over a 10 year (through to lifetime according to severity of disease) period to prevent repeated attacks of rheumatic fever and worsening RHD. Considered safe in pregnancy and lactation.
Valvular heart disease (VHD)	Any valvular heart disease (whether congenital or acquired).

Abbreviations

ACC	American College of Cardiology
ACM	Australian College of Midwives
AF	Atrial fibrillation
AHA	American Heart Association
AHP (also AHW)	Aboriginal health practitioner (also Aboriginal Health Worker)
AIDA	Australian Indigenous Doctors' Association
AMOSS	Australasian Maternity Outcomes Surveillance System
(AMOSS) RHD-P project	AMOSS RHD in pregnancy study (NHMRC project #1024206)
AMB	Aboriginal Mothers and Babies health service (generic term)
AMIC	Aboriginal Maternal Infant Care (South Australia)
AMHIS	Aboriginal Maternal Infant Health Service (NSW)
A&NZ	Australia and New Zealand
ARF or RF	Acute rheumatic fever or rheumatic fever
CATSINaM	Congress of Aboriginal and Torres Strait Islander Nurses and Midwives
CARPREG	Cardiac risk score
CC	Cardiac clinic or department
CDiP	Cardiac disease in pregnancy
CDM	Dedicated cardiac disease and maternity clinic
CHD	Congenital heart disease
CHW	Community health worker
CoC	Continuum of Care model for RHD: Medtronic CoC framework and interventional targets
COS	Core Outcome Set
CRS	Cardiac risk score
CVD	Cardiovascular disease
END RHD CRE	End RHD Centre for Research Excellence
ECG	Electrocardiogram
Echo	Echocardiogram (transthoracic TTE unless otherwise indicated)
ESC	European Society of Cardiology
GP	General practitioner
HRPC	High risk pregnancy clinic, with multidisciplinary care (not specifically cardiac)
ICOP	Indigenous Cardiac Outreach Project (Queensland, Australia)
LMWH	Low molecular weight heparin
MHV	Mechanical heart valve
MGP	Midwifery Group Practice
MM	Maternal mortality
MMR	Maternal mortality ratio
MOC	Models of care
MS	Mitral stenosis
NHMRC	(Australian) National Health and Medical Research Council
NMW	Nurse Midwife

NYHA	New York Heart Association functional class (I-IV)
PLwRHD, PLWRHD	People living with RHD
PBMV, PMV	Percutaneous balloon mitral valvotomy
PM	Perinatal mortality
PHV	Prosthetic heart valve
RHD	Rheumatic heart disease
RHDA	RHDAustralia
Reach	RHD: Evidence, Advocacy, Communication, Hope
RHS	Reproductive health services
ROPAC	Registry Of Pregnancy And Cardiac disease (within the European Society of Cardiology ESC)
SBA	Skilled birth attendant
SP	Secondary prophylaxis
TIPs	Tools for Implementing RHD Control Programmes (TIPS) Handbook, 2nd edition
VHD	Valvular heart disease
WHF	World Heart Federation
VKA	Vitamin K antagonist. Oral anticoagulation (eg warfarin)
ZAHARA	Cardiac risk score used in congenital heart disease

Appendix 2: Guidelines, Standards and Reviews, chapter 2

Framework/Domain
RHD or all-cardiac disease
Pregnancy and maternal health
Indigenous health and vulnerable populations
Health systems

Notes
'RHD* or all-cardiac disease' with asterisk indicates specific section and/or chapter content referring to RHD
Greyed rows = included in Part B systematic review and content analysis
Type [G] = Guideline or Standard; [R] = Review or report
Abbreviations: VHD = Valvular heart disease; CVD = Cardiovascular disease; PHV = Prosthetic heart valve; LMIC = low-, low-middle and middle-income countries; HIC = high-income countries

	Guideline/Review	**Comments**	**Framework/domain**		**Type**	**Country/Region**
			Primary	Addresses …		
1	Anthony et al (2016) *Valvular heart disease in pregnancy* [205]	Outlines management of specific lesions in pregnancy, with particular reference to post-rheumatic valvular heart disease. Discusses epidemiology and burden	RHD* or all-cardiac disease Pregnancy and maternal health		[R]	Africa
2	Brennand et al (2016) *Addressing the heart of the issue: Standards of good clinical practice in the shared obstetric and cardiology care of women of childbearing age* [187]	Response to MBBRACE-UK (Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries in the UK) 2014 report that called for coordinated action across a wide range of health services to address the problem of indirect maternal death, with focus on cardiac disease. Develops standards that outline best practice in the inter-professional care of women of child-bearing age who have cardiac disease.	Pregnancy and maternal health RHD or all-cardiac disease	Health systems	[R]	Scotland

	Guideline/Review	Comments	Framework/domain		Type	Country/Region
3	Brown et al (2015) *Essential Service Standards for Equitable National Cardiovascular Care for Aboriginal and Torres Strait Islander People ('ESSENCE')* [192]	Outlines elements of care necessary to reduce disparity between Aboriginal and Torres Strait Islander and non-Indigenous Australians in access and outcomes for five critical cardiovascular conditions (including RHD). Gives levels of evidence.	RHD* or all-cardiac disease Indigenous health and vulnerable populations Pregnancy and maternal health	Health systems	[G]	Australia
4	Elkayam et al (2005) *Valvular Heart Disease and Pregnancy Part I: Native Valves* [12]	Risk management, clinical assessment, surgery and intervention for VHD in pregnancy. Clinical focus, no reference to epidemiology. *'Mitral stenosis is the most common lesion, almost always due to RHD'.*	Pregnancy and maternal health RHD* or all-cardiac disease		[R]	North American-centric ('for CVD around the world')
5	Elkayam et al (2005) *Valvular Heart Disease and Pregnancy Part II: Prosthetic Valves* [188]	Clinical focus. *There are three main issues related to PHV and pregnancy that will be discussed in this review: 1) selection of PHV in women of childbearing age who desire to become pregnant; 2) maternal and fetal risks associated with pregnancy in patients with PHV; and 3) the management of patients with PHV during pregnancy.* Discusses selection of PHV for women of childbearing age 'difficult and needs to be individualized' and anticoagulation.	Pregnancy and maternal health RHD or all-cardiac disease		[R]	North American-centric ('for CVD around the world')
6	Elkayam et al (2016) *High-Risk Cardiac Disease in Pregnancy Part I* [202]	Reviews available published reports on high-risk cardiac disease in pregnancy and provides recommendations on risk assessment and management.	Pregnancy and maternal health RHD* or all-cardiac disease		[R]	North American-centric ('for CVD around the world')

	Guideline/Review	Comments	Framework/domain		Type	Country/Region
7	Essop et al (2005) *Rheumatic and non-rheumatic valvular heart disease: epidemiology, management, and prevention in Africa* [204]	Systematic review of valvular heart disease with some reference to pregnancy especially anticoagulation therapy. Calls for guidelines developed appropriate to African continent in context of 'socioeconomic issues and the HIV pandemic'.	RHD* or all-cardiac disease Pregnancy and maternal health	Health systems Indigenous health and vulnerable populations	[R]	Global with focus on Africa
8	Hameed et al (2017) *Improving Health Care Response to Cardiovascular Disease in Pregnancy and Postpartum. Developed under contract #11-10006 with the California Department of Public Health, Maternal, Child and Adolescent Health Division* [217]	Review in response to increasing maternal mortality, with *'One-fourth of the deaths …judged preventable if heart disease had been included in the differential diagnosis and timely diagnosis and treatment occurred'*. Brief specific reference to RHD under valvular disease. Includes: algorithm to guide stratification and initial evaluation; contraception counselling, CV medications during pregnancy/lactation; long-term health issues, contraceptive options, pregnancy planning; Discussion on racial and ethnic disparities in all-CVD prevention and diagnosis	Pregnancy and maternal health RHD or all-cardiac disease	Health systems Indigenous health and vulnerable populations	[R]	California USA
9	Heart Foundation of NZ (2014) *New Zealand Guidelines for Rheumatic Fever: Diagnosis, Management and Secondary Prevention of Acute Rheumatic Fever and Rheumatic Heart Disease: 2014 Update* [116]	Pregnancy section adapted from Australian RHDA CSANZ guidelines. Limited reference/ discussion related to models of care. Detailed reference to anticoagulation.	RHD* or all-cardiac disease Pregnancy and maternal health	Indigenous health and vulnerable populations Health systems	[G]	New Zealand
10	Knight et al (2016) MBRRACE-UK report *Saving Lives, Improving Mothers' Care* [219]	Response to 2012-14 maternal deaths report with focus on cardiac disease. Also refers to the Confidential Enquiry into Maternal Morbidity with focus on women with prosthetic heart valves in outlining best practice in the inter-professional care of women of child-bearing age who have cardiac disease. Specific reference to RHD.	Pregnancy and maternal health RHD or all-cardiac disease	Health systems	[R]	United Kingdom

	Guideline/Review	Comments	Framework/domain		Type	Country/Region
11	Mocumbi et al (2012) *Women's cardiovascular health in Africa* [203]	Africa. Reviews all-cardiac disease epidemiology/management in pregnancy with reference to RHD. Focus on access to services, reproductive health. This review focuses on the current knowledge of cardiovascular healthcare of women in sub-Saharan Africa, particularly their propensity for various forms of heart disease, access to healthcare, treatment received within the respective healthcare system, response to therapy and mortality. It highlights the gaps in knowledge and the paucity of data in most of these aspects.	Pregnancy and maternal health RHD* or all-cardiac disease	Indigenous health and vulnerable populations Health systems	[R]	Africa
12	Nishimura et al (2014) *2014 AHA/ACC Guideline for the management of patients with valvular heart disease: a report of the American College of Cardiology/ American Heart Association Task Force on Clinical Practice Guideline* [193]	Guidelines. Ch 13 Pregnancy and VHD Refers to Associated Guidelines and Statements - eg Table 2 Guidelines on the Management of Cardiovascular Diseases During Pregnancy ESC 2011 Risk management, clinical assessment, surgery and intervention for VHD in pregnancy. Clinical focus, no reference to epidemiology.	RHD* or all-cardiac disease	Pregnancy and maternal health	[G]	America. USA focus
13	Palafox et al (2017) *The World Heart Federation (WHF) Roadmap for Reducing CV Morbidity and Mortality Through Prevention and Control of RHD* [199]	Report. Gaps that limit access to and uptake of proven interventions for prevention and control of RHD with some reference to maternal health. *The devastating consequences of RHD during pregnancy and labour also require RHD control to be integrated with maternal care. Ensuring adequate counselling for women of childbearing age living with RHD, adequate antenatal case detection services and appropriate care during the perinatal period are essential components of a comprehensive RHD program. There are also recent initiatives that seek to integrate RHD care into other existing chronic disease management programmes. One innovative approach being trialled in Uganda aims to*	RHD* or all-cardiac disease Pregnancy and maternal health	Health systems Indigenous health and vulnerable populations	[R]	Global

	Guideline/Review	Comments	Framework/domain		Type	Country/Region
		integrate RHD surveillance and treatment within the existing HIV/AIDS infrastructure that has been successfully scaled-up after many years of domestic and foreign investment.				
14	Royal College of Obstetricians and Gynaecologists (RCOG) (2011) *Cardiac Disease and Pregnancy* [197]	UK. Good Practice guidance, with some reference to RHD.	Pregnancy and maternal health RHD or all-cardiac disease		[R]	United Kingdom
15	Remote Primary Health Care Manuals (2017). *Minymaku Kutju Tjukurpa Women's Business Manual.* 6th edition [391]	Standard treatment manual for Women's Business in remote and Indigenous health services in central and northern Australia. Section on RHD.	Pregnancy and maternal health Indigenous health and vulnerable populations RHD* or all-cardiac disease	RHD or all-cardiac disease	[G]	Australia
16	Regitz et al (2018) *ESC Guidelines on the management of cardiovascular diseases during pregnancy (European Society of Cardiology (ESC))* [196]	Detailed review with levels of evidence. *'Rheumatic valvular disease dominates in non-western countries, comprising 56–89% of all cardiovascular diseases in pregnancy'*.	Pregnancy and maternal health RHD* or all-cardiac disease		[G]	Europe with global relevance
17	RHDAustralia, Heart Foundation Australia et al (2012) *Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease (2nd ed)* [6]	Gives levels of evidence. Limited reference/discussion related to models of care. The 3rd Edition under development (release 2019) is substantially revised, with a new chapter Women with RHD that incorporates a whole-of-life approach as well as risk management, clinical assessment, anticoagulation, surgery and intervention for	RHD* or all-cardiac disease Pregnancy and maternal health	Indigenous health and vulnerable populations Health systems	[G]	Australia

	Guideline/Review	Comments	Framework/domain		Type	Country/Region
		RHD in pregnancy. Discusses epidemiology, context of disease burden.				
18	Roberts (2012) *Clinical assessment of women with cardiovascular abnormalities* [434]	Midwifery focus. Assessment, management and care for women who have or develop mitral valve stenosis during pregnancy	Pregnancy and maternal health RHD* or all-cardiac disease		[R]	United Kingdom
19	Siu et al (2004) *Cardiovascular problems and pregnancy: an approach to management* [435]	Global. HIC centric. Areas to be considered in clinical approach to the woman with heart disease who is pregnant or considering pregnancy: 1) risk stratification, 2) antepartum management, 3) peripartum management, 4) recurrence of congenital lesion in the neonate, and 5) site of antepartum and peripartum care. Proposed risk score (subsequently evolved into CARPREG score)	Pregnancy and maternal health RHD* or all-cardiac disease		[R]	Global, focus on high-income country relevance
20	Sliwa et al (2015) *Management of valvular disease in pregnancy: A global perspective* [218]	Review of all-VHD in pregnancy epidemiology/ management including surgery. Principles of care. Compares LMIC to HIC. Guide to risk assessment and overview of optimal cardiac and obstetric management, including surgical intervention, taking into consideration the resources available in higher and lower-to-middle income countries. Provides a practical approach and is not replacing comprehensive guidelines on the management of VHD or cardiovascular disease in pregnancy.	Pregnancy and maternal health RHD* or all-cardiac disease	Health systems Indigenous health and vulnerable populations	[R]	Global, compares LMIC to HIC
21	Zuhlke et al (2016) *Pre-conception counselling (PPC) for key cardiovascular conditions in Africa: optimising pregnancy outcomes* [207]	Call for shared responsibility and mandated PCC in cardiac care. Maps against WHO consensus. Discusses relevance of and need for PPC in resource-challenged settings. Also reference to HIV/AIDS and hypertension.	Pregnancy and maternal health RHD* or all-cardiac disease	Health systems Indigenous health and vulnerable populations	[R]	Africa

Appendix 3: Participant information and consent form

Australian Centre for Public and Population Health Research, Faculty of Health

Approval No UTS HREC REF NO. ETH17-1349

THE UNIVERSITY OF TECHNOLOGY SYDNEY

PARTICIPANT INFORMATION STATEMENT AND CONSENT FORM

[Title of project]

Rheumatic heart disease (RHD) in pregnancy: challenges of health service provision

[Participant selection and purpose of study]

You are invited to participate in a study that is investigating the challenges of provision of care for pregnant women with rheumatic heart disease (RHD).

We (Doctoral candidate Geraldine Vaughan, UTS Sydney, with supervisors Professors Elizabeth Sullivan [University of Technology and Conjoint Professor UNSW Medicine]; Michael Peek [Australian National University]; and Associate Professor Angela Dawson [UTS Sydney] are conducting two studies which aim to identify the principles of current best practice care of women with rheumatic heart disease (RHD) in Australia and the structural and cultural barriers that limit timely access to this care.

STUDY 1: quantitative study of surveillance and health information systems related to RHD in pregnancy that will review existing data sets, and validate surveillance data against the AMOSS RHD in pregnancy study; and

STUDY 2: a qualitative examination of models of care for pregnant women with RHD from the perspectives of the health workforce and policy makers, exploring specific factors that impact on – and contribute to – perceptions, health literacy and awareness of RHD in pregnancy by health services in Australia, and the people that provide those services.

You were selected as a possible participant in **STUDY 2** because you:

- Provide care for pregnant women with RHD;
- Work with health information systems that collect data related to pregnant women with RHD; and/or
- Work in areas that shape policy related to the care of pregnant women with RHD

[Description of study and risks]

The interviews will explore knowledge and awareness of RHD, particularly in pregnancy and perspectives on health care provision. Interviews will explore models of pregnancy care provided for women with RHD (antenatal, transfer from community, working with family and community, health services and interventions, strategies for coordination). Other themes will relate to education/information for women with RHD and working with related health services (eg maternity care with RHD Control registers).

If you decide to participate, Geraldine Vaughan or another investigator on the study will interview you, either in person or over the phone. It will be in a semi-structured interview format. Interviews will be audio-recorded.

Each interview will take an estimated hour to an hour and a half. There will be one or two interviews. The interviews would be held at a time and location that is convenient to you.

You may also be asked for the researcher to observe you in the clinical setting.

Access to notes of pregnant women with RHD may be requested. Where this was proposed, it would be conducted after any interviews/observation in clinical settings.

Participation in this research is unlikely to give you any direct benefit personally. You may find it useful and/or satisfying to discuss issues related to RHD in pregnancy. We cannot and do not guarantee or promise that you will receive any benefits from this study.

It's intended that this research will be used to inform guidelines, improve care and support better outcomes for mother and baby.

There is the potential – particularly if you work in communities with high rates of RHD - that you may be impacted by the disease either directly (through having RHD yourself) or that your family and community may be affected by the disease. If you became distressed by any part of the interview (or wished to stop the interview for any other reason) it would be stopped and not commenced again until if and when you were comfortable to do so.

You will be provided with a resource kit of currently available information that may be useful to yourself as a care provider for women with RHD in pregnancy, and to the women themselves.

[Confidentiality and disclosure of information]

Any information that is obtained in connection with this study and that can be identified with you will remain confidential and will be disclosed only with your permission, except as required by law. If you give us your permission by signing this document, we plan to discuss and publish the results in peer-reviewed journals and present findings at conferences and meetings. In any publication or presentation, information will be provided in such a way that you cannot be identified.

[Recompense to participants]

If you agree to participate, any costs involved in travel or other expenses would be incurred by us, the investigators.

[Complaints]

Complaints may be directed to the Research Ethics Manager, UTS Sydney, 2007 AUSTRALIA (phone (02) 9514 1279, email Brie.Turner@uts.edu.au). Any complaint you make will be investigated promptly and you will be informed out the outcome.

[Feedback to participants]

You will be asked your preference to receive outputs from the study. This will include whether to receive material (including copies of any peer-reviewed publications) and abstracts from conference presentations, as well as a summary of research findings from the study in printed or electronic format.

[Your consent]

Your decision whether or not to participate will not prejudice your future relations with the University of Technology Sydney or any researcher involved in the study. If you decide to participate, you are free to withdraw your consent and to discontinue participation at any time without prejudice.

If you have any questions, please feel free to ask us. If you have any additional questions later, Geraldine Vaughan, (Doctoral candidate); Geraldine.vaughan@uts.edu.au tel:) will be happy to discuss with you.

You will be given a copy of this form to keep.

THE UNIVERSITY OF NEW SOUTH WALES

PARTICIPANT INFORMATION STATEMENT AND CONSENT FORM (continued)

[Title of project]

Rheumatic heart disease (RHD) in pregnancy: challenges of health service provision

You are making a decision whether or not to participate. Your signature indicates that, having read the information provided above, you have decided to participate.

……	……
Signature of Research Participant	Signature of Witness
……	……
(Please PRINT name)	(Please PRINT name)
……	……
Date	Nature of Witness

REVOCATION OF CONSENT

(*Title of project*)

I hereby wish to **WITHDRAW** my consent to participate in the research proposal described above and understand that such withdrawal **WILL NOT** jeopardise any treatment or my relationship with The University of New South Wales.

…… ……

Signature Date

……

Please PRINT Name

The section for Revocation of Consent should be forwarded to:

Geraldine Vaughan, Doctoral candidate, Faculty of Health, University of Technology, Sydney. geraldine.vaughan@uts.edu.au

Appendix 4: Interview guide

Rheumatic heart disease (RHD) in pregnancy
Qualitative study: challenges of health services

Health Professionals

Research questions

1. What degree of knowledge, expertise and awareness exists amongst health professionals providing care for women with RHD in pregnancy?
2. What needs do Aboriginal and non-Aboriginal women have that are not currently addressed in health service access, counselling and clinical management of RHD in pregnancy, and how does that vary between services?
3. What barriers exist to the provision of optimal health care from the perspectives of health professionals who provide that care?
4. How can health services more effectively meet the needs of these women, including access, education, counselling and clinical management of RHD in pregnancy?
5. What factors enhance/hinder the capacity of health information systems (medical record systems, RHD Control Program registers) to support health professionals in providing timely well-informed care for pregnant women with RHD?

List of topics to be covered in the interview:

- Professional role and location (maternity, cardiac, Aboriginal health, remote health, RHD Control program staff).
- Knowledge and awareness of ARF/RHD: disease, treatment, management (general and in pregnancy). Knowledge of access to resources.
- Experience of provision of care for pregnant women with RHD: when diagnosed, what the woman was told, her knowledge and understanding of the disease and treatment. Health professionals' experience and understanding of secondary prophylaxis, echocardiography, issues about anticoagulation. Experience and understanding of surgical or other interventions.
- Health care service provision for pregnant women with RHD: logistics, access, strategies, policies, resources. Challenges and gaps. Identifying what works and what doesn't.
- Urban and regional/remote differences in health (general, cardiac and maternity) care access and services.
- Health information systems and surveillance: what degree of coordination exists between systems to support optimal and timely sharing of information? What gaps exist? How integral is the RHD Control register perceived as being in the provision of

care for women with RHD? What gaps exist, and what strategies have been successful in promoting timely effective sharing of health information?

Interview guides

- Clinical
- Non-clinical

Clinical

Conversational style in your own words at appropriate times	**Topic**
1. **Professional and experience:** ***Tell me about your experience working with women with RHD.*** *Role; type of health unit (maternity – antenatal, labour ward, postnatal, MGP, remote health, cardiac); length of time and experience in this or other clinical setting.*	T1 T5
2. **Knowledge/awareness. Models of care** ***Can you describe the process of identification and clinical care for pregnant women with RHD in your centre/unit?*** *Notification – health information systems, surveillance strategies* *Knowledge/awareness of management strategies - Secondary Prophylaxis (SP), safety in pregnancy* *Knowledge/awareness of ARF/RHD Register, how to access, which jurisdictions* *Use of resources - online course, phone app, guidelines, quick reference.* *Clinical policies/guidelines in this clinical setting associated with the care of pregnant women with RHD? Continuous Quality Improvement (CQI) or similar program?* *Resource impact - where does ARF/RHD sit in relation to other (chronic) diseases/conditions in this service/centre/unit (prevalence, resource impact, awareness)*	T2 T3 T4 T5 T6

Changes in the approach to care for pregnant women with RHD over the last five/ten years?

3. **Delivery of care:** T3
What are the key issues in delivery of care for women with RHD in pregnancy? T4

Collaborative care with cardiac, maternity, Aboriginal health, AMIC, services, RHD Control Register (Other?) T5

Continuity of care. Logistics of coordination – antenatal and cardiac care – SP treatment, echo.

Health information systems, information transfer

Transfer, transport, access to services

Support for women - interpreter, Aunty, Aboriginal Health practitioner.

Disconnect between model of care policy and practice.

4. Education and health literacy for women: T3
What do you ask/talk about with pregnant women who have RHD? How would you describe women's understanding of the pathology of the disease? Examples? Resources you use? What would support Improved understanding/adherence to treatment? T4

Heart pathophysiology, SP and other preventive strategies, management, anticoagulation if relevant, contraceptive counselling, RHD in pregnancy, adherence to treatment.

Who attends – partner, interpreter, Aunty, maternity, cardiac.

5. Service structures T6
What changes could be made to better meet the needs of pregnant women with RHD? T5

Provision and structure of services – cardiac, Aboriginal health, remote.

Access - transport, transfer

Health information systems – data collected and how this is managed/shared. Efficiencies and accuracy.

6. **Experiences** Thinking about your experiences of care for women with RHD in pregnancy. Can you remember a time where you thought things went well for her care. Why? What happened? Can you remember a time where you thought there were challenges in her care. Why? What happened?	T3 T4
7. Other comments/thoughts?	

Non-Clinical

Conversational style in your own words at appropriate times	**Topic**
1. **Professional and experience:** ***Tell me about your experience working with women with RHD.*** *Role; type of service/facility/function; any experience in clinical settings.*	T1 T5
1. **Knowledge/awareness. Models of care** ***Can you describe what you know about the process of identification of RHD and how that's managed in pregnancy. Resources available.*** *Notification – health information systems, surveillance strategies* *Knowledge/awareness of management strategies - Secondary Prophylaxis (SP), safety in pregnancy* *Knowledge/awareness of ARF/RHD Register, how to access, which jurisdictions* *Use of resources - online course, phone app, guidelines, quick reference.* *Clinical policies/guidelines associated with the care of pregnant women with RHD? Continuous Quality Improvement (CQI) or similar program?* *Resource impact - where does ARF/RHD sit in relation to other (chronic) diseases/conditions in this setting (prevalence, resource impact, awareness, policy priority)* *Changes in the approach to care for pregnant women with RHD over the last five/ten years?*	T2 T3 T4 T5 T6

2. **Delivery of care:** T3

What are the key issues in delivery of care for women with RHD in pregnancy? T4

Collaborative care with cardiac, maternity, Aboriginal health, AMIC, services, RHD Control Register (Other?) T5

Continuity of care. Logistics of coordination – antenatal and cardiac care – SP treatment, echo.

Health information systems, information transfer

Transfer, transport, access to services

Support for women - interpreter, Aunty, Aboriginal Health practitioner.

Disconnect between model of care policy and practice.

3. Education and health literacy for women: T3

What resources are available for women with RHD? What could be done to improve understanding/awareness/knowledge of the disease (particularly in pregnancy) for women? For those working in clinical settings? T4

Health literacy: for women; for those working in clinical settings.

Resources and availability

8. **Service structures** T6

What changes could be made to better meet the needs of pregnant women with RHD? What are the key issues in delivery of care? T5

Provision and structure of services – cardiac, Aboriginal health, remote.

Access - transport, transfer

Health information systems – data collected and how this is managed/shared.

Efficiencies and accuracy.

9. Other comments/thoughts?

Appendix 5: Coding categories & codes: study two chapter 5

Care pathways for pregnant women with RHD: perspectives from health professionals

Coding categories

The thematic coding process involved segmenting and categorising the qualitative data for subsequent thematic analysis[280]. Coding followed the recursive approach outlined in methods section (Figure 3-2). Work outlined in previous chapters and the implementation of the AMOSS RHD-P study informed the development of an initial set of codes which were revisited, expanded and refined over several iterations. The initial categories with associated codes formed in NVivo included:

- Health professional education & awareness of RHD in pregnancy
- Reference to Guidelines
- Access to services
- Health information systems and referral pathways
- Health workforce, sectors, resources, funding
- Models of care and clinical management
- RHD programs and RHDA
- Women with RHD
- The big picture. Politics of RHD. Advocacy. Other RHD research

As analysis progressed, these categories were further refined.

The data were de-contextualised from their original interview. Coding categories were reconceptualised, renamed, reorganised, merged and separated as the analysis progressed: *'categories are seldom static and never inviolate, as they are subject throughout the analysis to the search for alternative interpretations or disconfirming evidence'*[280].

Topics were typically associated with multiple codes and were allocated and re-allocated in an iterative process under the following categories:

Education & awareness of health providers

Education. Knowledge & awareness. Health provider literacy

Deficits in awareness and knowledge extending beyond specific clinical expertise were commented on:

> *…the new guys [clinicians] up here they just don't understand the situation that these patients are in…They have zero insight into logistics and cultural nuance* [CL01 Clinician in urban tertiary centre with high risk population, referring to specialist services]

Others with an evident expertise spoke of their proactive role in promoting RHD awareness and knowledge, particularly for those new staff who may not have worked in high prevalence regions previously:

> *For midwives, when opportunities come up such as the [RHD] seminar the other night, I had two new midwives who don't know very much about rheumatic heart in terms of its effect on pregnancy who came. So there are those opportunistic things that happen. But also, when I have new midwives I either get someone to come here and talk and it would usually be the rheumatic heart nurse educator or whatever. I'm not sure of her title but I know her*. [CL04 Midwife in Aboriginal Mothers and Babies (AMB) service]

Education reaching out to maternity

Formal and informal education for maternity and other health services, particularly for staff who had little or no exposure or awareness of RHD was a theme expressed by many participants.

> *I try to [provide education to maternity] at least once or twice a year, to try and catch the new staff ... and that care plan, for ladies with RHD. The long-term staff understand the process. But for any new staff, yeah, no, they're not always— They're not aware there's any such thing as RHD - haven't had any exposure to it so it's a whole learning curve.* [SP04 RHD coordinator]

RHD Program participants spoke of the growing awareness of impact in pregnancy and possible strategies to respond:

> *I became more aware of rheumatic heart disease in pregnancy following the excellent work of the AMOSS study. Those pieces of research and then the follow-up forums that were held started to shape thinking about how RHDAustralia could respond.*[SP01 RHD Program]

Achieving improved awareness among health services required a similar fluid approach as in other RHD strategies that often crossed boundaries to 'make things happen'. While the formal education (encompassing educational resources and systems) was seen as

fundamental, the delivery of these often happened outside formal structures. An agile approach that harnessed multiple resources and stakeholders was referred to:

> *… and I think having services pick up [available educational resources], building that into systems so … there is a suite of resources that can be picked up by clinical educators, in hospital services and the like, that are topical, information specific for whatever audience then I think we start to build an energy, in terms of increasing health literacy amongst care providers.* [SP03 RHD coordinator]

RHD coordinators described their role in providing this, working in clinical settings to promote awareness and knowledge, whilst acknowledging resource and other limitations:

> *…it's about having structures and processes in place to increase the awareness and capability of existing staff …We're closing the loop over time. And I think having detailed information, having services pick that up, building that into systems so that the reliance isn't on an RHD Register program to deliver that education [by]… having a suite of resources that can be picked up by clinical educators, in hospital services and the like… we start to build an energy, in terms of increasing health literacy amongst care providers.* [SP03 RHD coordinator]

> *As well as providing education and orientation for health staff, looking at ways that data from the Register can help inform practice is an important part of the [RHD] Register and Control Program. And we've started looking at pregnant women who are on the Register. We recently started sending out reminders to the health sites, not only around secondary prophylaxis for these women, but also around echocardiograms. So, has the woman had an echocardiogram? Prior to her third trimester, has a delivery plan been put in place? Has the woman seen either a cardiologist or an obstetrician? And I guess one of the main triggers for us initiating this, was a woman where the RHD wasn't picked up during pregnancy and she deteriorated after birth. The fallout effect of all the emergency measures was really traumatic. So, she lived about seven hours' drive from [remote/regional centre] and from there had to be transferred another 2,500 km to the tertiary centre. And the poor woman was trying to take in information about her new sick baby as well as information about this RHD diagnosis - all on the run. And whilst we don't have screening during pregnancy, the lesson that we learned from that was that was how important*

to not miss diagnosis and support health sites so that we minimise those traumatic emergency responses.[SP02]

RHD and other chronic diseases

The thing that rheumatic heart disease keeps coming up against is getting accepted in the (regional/remote) region as a priority when we've got lots of other research projects going on. That's the tricky bit. There's not that many of them so how will this project which will involve a lot of people's time and effort help anybody? [CR02 Clinician/researcher in regional high prevalence area]

RHD is a small problem by comparison. The biggest is diabetes. Then, after that you have your preeclampsia. Because there are other factors which relate to it, you know, like smoking, alcohol, lifestyle and violence and all those kinds of things. But rheumatic heart is— I wouldn't say it's a large part. We've had a few who have had the valve replacements but not many. But it's the impact. And other conditions… just a few days back I had one girl with ischemic heart disease and had had stents and all that. She is hardly 20.[CL05 Obstetrician]

Richer, more intensive information and training. Tiers of education. Having systems and structures in place to deliver

Health information systems. Referral pathways. Logistics

Health information & perinatal data & primary health care info & RHD Registers systems, talking to each other; data linkage. Logistics. System blocks, delays, gaps

There was near universal frustration across jurisdictions and health services expressed by participants who referred to health systems and managing information [CL01, CL03, CL04, CL06, SP01, SP02, SP03]. RHD Program staff were particularly cognizant of the barriers created by poor articulation of clunky systems that did not talk with each other, often with no real-time access, multiple tiers of silo'ed information, multiple players across public and private sectors and multiple technologies including legacy paper-based… and the direct impact that had on quality of care and outcomes. This was particularly evident in primary health care systems.

In remote areas we're now we're seeing multiple facilities being established and so, it's not just about the data sharing arrangement with one facility, per community, it's two or three. [SP03 RHD]

Like much of what was discussed, there was not one cause or reason – or solution – in addressing logistics-based gaps in providing care. Most participants referred to the mix of system-driven, personnel-driven, resource-limitations, structural and personal factors that factored when it didn't 'work'.

> *And what has been a real challenge in that process is there's many points of the journey that can go wrong. It can be that the referral was never received. Or it was never sent adequately and you didn't know because you've left the machine and it's… The piece of paper saying it didn't send came out and the person clearing the fax, the machine, doesn't know what to do with the pieces of paper and it gets lost for ever so you don't know that it's never sent. That there's no receipt.* [CL03]

> *I think they think we're stupid when they have to tell the same story for the twentieth time. Which could be solved by better IT systems. That's very frustrating particularly when you're doing it through an interpreter. So, I think the IT systems, for me, are the weakest link but the easiest to fix. Logistics.* [CL01]

The theme of RHD and not missing its diagnosis being a mix of system and personnel criteria is nicely described over time by a clinician who had worked in remote communities in one region for over 30 years. The 1980s, while more challenging in terms of diagnosis (with no electronic data and lack of direct access to cardiac services and echocardiogram) were seen to have better continuity with paper-based lists, which meant people with RHD were followed up and bicillin injections tracked. There was seen as associated with a 'fair degree' of staff stability and cohesion. The scope of the public health perspective of medical care was less complex during this time:

> *We did worry about diabetes and we did worry about rheumatic heart disease. But we didn't worry about a lot of other chronic diseases.*[CR02]

However, this continuity of care and monitoring of RF/RHD was seen as community and staff-dependent.

> *It was probably very erratic across the region, and over time. Towards the end of the '80s a lot of that declined, through changes of circumstance; change of staff and it becoming less a priority. You know, if the paper list didn't get handed on to the next person, well, it got lost and so, we started again. Until*

there was the Register, I don't think it was actually better and it might have even been worse than it was in the '80s. [CR02]

An obstetrician in an Aboriginal Mothers and Babies service described the blocks to knowing about a woman's history, which were mostly system and resource-based, as well as the workaround she had implemented to get a better picture. She describes the impact of the Aboriginal Mothers and Babies service acting as a conduit between community and tertiary centre.

I've instituted a teleconference [between the Aboriginal Mothers and Babies service and communities]. And I go through every single friggin' antenate with two systems open on my computer in front of me. Using the hospital record system and my system. So using my system I can say, "Hey, hang on a second, let's go back" and hopefully I've got their notes. That means I can look at what actually happened in a previous pregnancy. Because your discharge summary is only as good as the doctor who wrote it and some of those discharge summaries are great but some are really, really, really bad. Or a discharge summary could land in the clinic PO box and just sit there. And then you'd get the screams from the clinic, "We haven't got a discharge summary". The advantage with Aboriginal Mothers and Babies service is that I know we've got the midwife, who's her midwife, we are tightening up our communication with the clinic, and so we're just trying to make sure that we close the loop about how things are going to be followed-up. Also we've made arrangements with outreach to access (another system) so that we can get hold of results. NT Cardiac exists in a bubble. So, the amount of time we spend just faffing around, ringing up and saying, "Can you fax the echo report because I've got a patient in front of me?" Then the other problem is that we don't have the staff in Medical Records: there are piles and piles of paper that have never been filed. They're two years behind. So, we have to devise complicated methods of making sure that we've got follow-up. The women, they often don't come into town with that information. Which means again we have to request it from the communities. Their communities keep all of their handheld records in the clinic because they lose it. You spend an awful lot of time doing detective work. [CL06 Obstetrician in tertiary centre with high risk population]

RHD program staff were also referred to joining the dots in making sure clinical information across services was not missed:

The rheumatic nurse has been fairly pivotal– they hound down the outreach project to get reports, for example.[CL02]

However, the RHD registers, as distinct systems, were not automatically queried. On visiting one hospital maternity unit to review case records for the RHD-P study, the AMOSS data collector midwife knew about the existence of the Register, but seemed unsure of how to access it.

I would have to say that we probably are not as active in seeking out the information from the registers as we might be. [CL04]

Another clinician commented that better information did not necessarily require more information in each system:

We overcomplicate things. So, for example, the point of a Register, is predominantly that people get their prophylactic [BPG bicillin injection for secondary prevention]*. That's where the big saving is. So we should focus on that. Yet the more extra things we start to do, the less the focus is on the secondary prophylaxis. With RHD, in pregnancy, the focus should be on identifying it.* [CR02]

The call for better-integrated systems was directly linked to improved outcomes for women with RHD:

We need information systems that actually talk to each other and are centralised. One patient identifier across the board. We used to have five, I think we've only got three now. But I don't know what's happened to all the history that was on those ones that aren't used. Probably disappeared into the ether. So, I think improving information systems is Number 1 to help improve outcomes in a tangible way. [CL06 Obstetrician in tertiary centre with high risk population]

Patient information entered in multiple media (paper-based and electronic) and across multiple platforms was confusing and potentially missed. Interviewees referred to up to seven systems that they had to query.

How do we find out about women, their pregnancy and RHD? Well, you've got your hospital system; which includes health data plus demographics. You've got the RHD Program; you've got primary health. And Royal Flying Doctor

Service. And they don't talk to each other. Generally they either print out notes and scan or fax or email them to us. And we put them in the paper-based record. We have perinatal data - that tends to get entered retrospectively. That's generally how it goes. And we try and remember to send the information back. That sometimes works and sometimes doesn't. [CL11 Midwife in regional hospital]

These descriptions matched with my own when entering data for the AMOSS RHD-P research, which included five urban tertiary centres, three regional and one remote hospital, all of which had disparate paper-based and electronic systems that often did not articulate. This impacted at two levels: confirming diagnostic criteria for inclusion in the study, and the ability to tap into the detail required to populate the data collection – both historical and during the index pregnancy, particularly if a woman was transferred. Data collectors at participating AMOSS tertiary-level sites found supporting notes and reports were not forwarded from referral centres, which impacted on the ability to confirm inclusion.

We haven't been able to send referrals electronically. And we keep telling the IT people that and it's not sorted out yet. They receive them either by fax or scanned/emailed. But, even then, sometimes people fall off the list so the people who need to go and have the echo done from community may not get an appointment or they get an appointment but then when our patient travel system, might make mistakes and the wrong date is entered because of literacy levels of the people doing the patient travel. And… so patients may miss appointments. That wasn't their fault. They might not have been notified. Or where people can have an echo and we don't know how it was organised and we're not notified of the result and there doesn't seem to currently be a connection between… If you perform an echo and you diagnose rheumatic heart disease on that, there doesn't seem to be a requirement to notify the RHD register. In one woman's case we didn't even know she'd had the echo, which said she had moderate to severe, I think, mitral stenosis or it might have been regurg actually. And she should have been on LA Bicillin. But she left and didn't wait to see the cardiologist. And no-one notified us that they're concerned that she's got RHD. So, a number of months passed before it was picked up - very close to when she delivered. [CL03 Clinician]

Other clinicians had better experiences. One reflected on what she saw as improved clinical processes and a shared electronic record, particularly in relation to RHD management and the program coordination …

> *All the electronic systems they have, these recalls are so much easier to capture, you know. [CL05 Obstetrician]*

One midwife working found that information systems worked better in their regional/remote primary health centre:

> *We'd know about them straight away when they came in because, well hopefully we've already got the diagnosis on our computer system. And we strongly encourage our antenatal clients to be signed up to [My e-Health]that so we can link into other communities. As well, if they come from somewhere else. I try to go through the list at least weekly or fortnightly, to see if they've been seen elsewhere. Like if I can see on that they're getting regular check-ups at [another community], I just don't worry about them. It's only when I can't see that they are getting anything done anywhere that I might start looking around to ring up somewhere. Making sure antenates are on my-Health is a bit of a priority for us. 'Cause the hospitals can also see that information as well. You know, my routine is to wait for the 28 weeks GTT and then send the antenatal notes to the hospital. But if they turn up there at 20 weeks, then at least they've got some information on them if they're on a myHealth record. And also making sure we document things like when they had their last LA Bicillin and that if they're on it. Communicating electronically has made it just a little bit easier but other than that, I think we've always pretty much been on the same page with this one. Yeah.* [CL07 Midwife]

Incorporating systems to actively capture information at the antenatal booking visit has been slowly implemented in high prevalence regions. A partnership program between RHDAustralia and NT outreach midwives saw specific reference to RHD on the midwife antenatal intake form:

> *There was a bit of pushback saying, "Well, you know, every clinician wants their own disease to be included in there" but when we highlighted, "Actually, if you miss it, they could potentially die, during pregnancy or postpartum, 'cause you missed it. So, actually it's extremely important".* [SP01 RHD Program]

(other reference – AMHIS NSW pull it in if it's here already or enter]

The jurisdictional RHD program data systems had their own challenges:

> *Any duplication of information, or information that's locked in discrete systems that don't talk to each other creates a drain on resources. So there's impact on workload but more importantly, it's the missed information in the patient records, that then impacts on outcomes and quality of care. Also awareness that the Register exists in the first instance. And then, if it* is *noted, what does it actually mean for practice especially during pregnancy? [SP01 RHD Program]*

All RHD program staff interviewed called for improved articulation of information systems to support their work.

> *Without significant improvement to data systems, we won't be capable of knowing how many of our clients are being cared for through antenatal processes.* [SP03 RHD Program]

They emphasised that information should not be (re)entered by program staff into the RHD data system, but rather tap into existing data systems:

> *If there was an intent for us to collect additional information about pregnancies, about complications … then we would need to add those data fields. It becomes a data collection nightmare.* [SP03 RHD Program]

Privacy concerns

Concerns about privacy were referenced by one participant, who continued to describe that information-sourcing was personnel rather than system-based:

> *First of all we ensure that Registers are secure databases, so we can guarantee peoples' privacy. And then I think we need to look at what other systems can securely feed into our system. And again, upholding all of those principles of privacy so people don't feel violated but the Registers can obtain information that's relevant to their RHD - for instance, about pregnancy. At the moment RHD programs are heavily reliant on staff knowledge and on them informing us. The issues in relation to data linkage are that there are multiple patient information systems particularly among primary care providers. We would like to look at how they can support some of those patient information linkages into one system.* [SP02 RHD Program]

Like so much of RHD, the reporting of this disease to Registers had social and historical context and consequent potential barriers. The experience described below was described by one midwife in NSW (where rheumatic fever and RHD if under 35 had recently become notifiable diseases).:

> *Then the public health unit places the woman on the Register, with consent. It was interesting… when we first talked about the Register, Aboriginal health workers thought that it was a trigger for a report to DOCS [Community Services NSW] if they didn't turn up for their Bicillin.* [CL08]

Referral pathways

> *Essentially, it's a paper-based referral system so there's no electronic referral system. And, not even the core services have a decent referral system. Which is a bit sad. Just in the last couple of weeks we've actually instituted an email account referral system which we're hoping is going to allow us to track patients better. And we're going to be able to place their expected delivery dates into a calendar so that we can actually keep track of all of our peri-operative referrals for high-risk women. [CL01 Clinician]*

> *And what has been a real challenge in that process is there's many points of the journey that can go wrong. It can be that the referral was never received. Or it was never sent adequately and you didn't know because you've left the machine and it's… The piece of paper saying it didn't send came out and the person clearing the fax, the machine, doesn't know what to do with the pieces of paper and it gets lost for ever so you don't know that it's never sent. That there's no receipt. [CL03]*

Health workforce, sectors, resources, funding

Health workforce factors in promoting effective, respectful models of care for women with RHD in pregnancy centred particularly on Aboriginal health practitioners, midwives and RHD program staff. These practitioners were all referred to the context of making links and establishing continuity of care for women. Many of the themes (such as resourcing, education and training, remuneration, workplace structure, gaps in communication) suggested global system issues but with heightened relevance for RHD and pregnancy.

There were also several references to what was seen as inappropriate use of Aboriginal health practitioner skills and poor investment in training and education, which in turn had specific relevance to the care and outcomes for women with RHD.

Aboriginal Health practitioners. Aboriginal liaison Aboriginal Medical Service

Models of care provided by Aboriginal and/or Torres Strait Islander practitioners reflected the nature of service and where this was located and varied accordingly.

Frustration at inappropriate use (and lack of recognition) of skills and missed opportunities to work collaboratively was expressed by both Aboriginal health practitioners and non-Indigenous clinicians. This was particularly evident in hospital and primary health care settings. [CL01, CL03, CL05, CL06, CL13, SP02, SP03]

> *There was a lot of money spent training Aboriginal health practitioners [in the 80s and 90s]. … And no one knew what they were there for. People just thought they were social workers... So apparently, they weren't used [sic], so the government unfunded them all and… There are a few left. I think that was a hugely wasted and missed opportunity.* [CL01 Clinician in urban tertiary centre high prevalence region]

Appropriate recognition in terms of remuneration was also referred to as a barrier to a skilled workforce structure:

> *One of the big issues in that space is that they're [Aboriginal health practitioners] … paid less than if they hadn't done their training, which blows my mind. So they need a career progression pathway. So they can see a future in what they're doing and they can progress to roles which remunerate fairly. And justly. I think, a big issue is purely just getting them on an award structure. And having a career progression structure which would support good people.* [CL01 Clinician in Northern Australia urban tertiary centre]

> *And I think a really, really big part of all the health stories in the NT is that we need more Aboriginal health staff, in the hospitals and in the community health services. When we don't have them, we don't work well. 'Cause they're our connection, that cultural connection, you know, worth their weight in gold and I don't think that's appreciated how valuable their role is.* [CL03 Clinician working northern Australia urban tertiary centre and remote]

> *I don't think we've given enough thought to how we need to support people to learn our culture - particularly the culture of health services. It's hard enough for non-Aboriginals to navigate through the system. There are some positive things happening. And certainly in the [specified community] they have had a*

very good Aboriginal health practitioner program where you have mentors who are life mentors as well as clinical mentors. [CL03 Clinician working in urban tertiary centre and remotely, northern Australia]

I think it's also got to do with stability within clinic staff. If you've got a stable clinic you're going to have stable health workers. For example, [specified independently-run clinic] is incredibly stable. Their AHWs [Aboriginal health workers] organise things. We come out, they say, "This is the women's doctor's day". They do all sorts of stuff. We'll do education related stuff and all sorts in conjunction with it, which is basically run by the Aboriginal health workers. But, in a lot of places they use the AHWs as drivers. "Go and pick up so and so." And they're paid really badly. And the education… we did it better in the '80s and the '90s. [CL06 Obstetrician working in northern Australia urban tertiary with remote outreach]

Regulations defining work that can be performed by Aboriginal health practitioner vary by jurisdiction, with inconsistent recognition of training, skills and abilities. Situations were described where, on a busy clinic day, clients waited for their secondary prophylaxis Bicillin injection for a while and then left without receiving it. Indeed, this occurred with a young pregnant Aboriginal woman in a community I was visiting. Meanwhile, a senior experienced Aboriginal health practitioner with Certificate IV training drove clients to and from clinic.

Several participants referred to Aboriginal health practitioners' predominant role as a driver. [CR01, CR02, CL01, CL12, CL13]

Queensland has cut back a lot of our stuff that we can do. And we only can do basic stuff and driving and that's it. No injections. You know, to me, it's a waste of talent. [CL13 Senior Aboriginal health practitioner with Certificate IV]

The inability of [Aboriginal] health workers to give injections is a perfect example of that. And when it was in place, the legislation only permitted health workers to give injections if they work a certain distance away from pharmacies. What a pharmacy has to do with it, who knows? And so, in some locations with a large number of people on our Register, even if they were qualified, they couldn't actually give the woman an injection 'cause the island's not big enough to exceed the distance requirements from the pharmacy. [SP03 RHD Program Coordinator]

One clinician commented that they now spent more effort encouraging entry to medical and nursing education rather than Aboriginal health worker programs [CR02]. Others talked about the embedded structure that supported Aboriginal midwife or nurse training and the women who worked part-time at the health service whilst completing their education [CL04, CL07]

Other models and services were described by participants. The Indigenous Cardiac Outreach Project (ICOP) is staffed by a cardiologist, echocardiographer and Aboriginal health practitioner/program coordinator, providing services regional and remote Queensland communities. At each community, the team worked alongside all clinic staff, providing echocardiograms, reviewing status of patients and referring where necessary. Aboriginal health practitioners perform electrocardiograms together with ICOP staff, and reviews automatically included all service providers, often together with the client.

> *It's good to have that communication with the doctors and the Indigenous workers that come with them. You know what's happening from one visit to another. After they finished their clinics we all sit down, have a cuppa and just a normal conversation. They'll have a list of people who… You know, they tick and flick and then they'll explain everything to us. "We still need these people to stay here on this". You know. Things like that, so…I reckon it benefits the whole team.* [CL13 Senior Aboriginal health practitioner with Certificate IV]

One obstetrician in a regional hospital described where the Aboriginal health practitioner was part of the regular multidisciplinary (MDM) meeting [CL02] but this was otherwise not referred to.

RHD programs have a coordinator (Indigenous or non-Indigenous) and Aboriginal health practitioner working together and visiting homes. One coordinator describes the slow building of trust to work together and within the community:

> *Now we both go together to do a risk assessment in the home. I got cheeky once and asked if I could bring a student. And Joan (RHD Aboriginal health practitioner] just looked at me and said, "Sara, it's taken me long enough to get them to let* you *come to the house".* [SP03 RHD Program coordinator]

Integral to the structure of Aboriginal Mothers and Babies services are the Aboriginal health practitioner and (Indigenous or non-Indigenous) midwife working together to provide care

during pregnancy and the early post-partum. The shape of this naturally varied according to location and service.

Two of the midwives interviewed described their experience establishing an Aboriginal Mothers and Babies service:

> *There was no (Aboriginal) health worker employed. So nothing was going to happen in the community until there was a health worker. Maggie didn't come on board until maybe three months later. Which was fine 'cause it gave me a chance to get to know the community, the systems, working for the government again so… I had time to do my own research. Then Maggie and I hit the ground running. You know, I think we both knew how it would work and what had to be done. And so we were fortunate… we were left alone to do what we had to do and over time, very slowly, built it up. I think the reason we were so successful is that we were so compatible. There was mutual respect. Certainly my 16 years in Aboriginal health set me up for this. Yeah. And I think that's why we ended up with such a solid program here.* [CL10 Midwife]

Another described how the service evolved in response to need and as the service grew:

> *We had Aboriginal health practitioners to start [in an Aboriginal Mothers and Babies service]. And then, we moved to have a strong woman worker. Because the need wasn't there to have, you know, as much direct clinical input. So now there is one of each which is a nice balance.* [CL04 Midwife in Aboriginal Mothers and Babies service]

Clinicians commented on the impact of RHD for Aboriginal health practitioners themselves.

> *We had a health worker in that area with RHD. But it wasn't until she attended the [AMOSS RHD-P] workshop that she could relate to what was being said.* [CL10 Midwife]

> *An Aboriginal health worker in Flat Creek [a remote community] had a murmur noted which I also heard but other people who had listened to her heart didn't think it was anything. Then she became pregnant - 5th or 6th pregnancy. And… Then out of the blue, later in second trimester she went into heart failure. In Flat Creek. And we did get her to the tertiary centre [2,500 km away] and she did do alright and she did have the baby, a big healthy bouncing boy. But it*

was… Yeah. It really highlighted for me, that, that particular danger of, you know, us not being aware of RHD in pregnancy is a big thing. [CR02]

The model of education in Aboriginal Mothers and Babies services and the mutuality of learning between midwife and Aboriginal health practitioner was described by a midwife:

I love the education side of things. It's something that's easy for me and it's so important. So we (Aboriginal health practitioner and midwife] spent a lot of time together in the car and what not. And… You know, she would hear me, what I was saying to people and lots of talking in the car… Yeah. She learned my stuff and I learned a lot of her stuff. And it was really good because sometimes I might throw in a word that's too medicalised she would clarify, or if I had missed something, she would just come in and explain. It just worked very well. [CL10 Midwife]

Midwives and nurses

The role of midwives and midwifery group practices in supporting streamlined systems and continuity of care was referred to by several participants [CR02, CL02, CL03, CL06:

There's an outreach midwife and antenatal coordinator now. Before that we didn't even have a midwife clinic. And the outreach midwives have made a tremendous difference. And we've got midwifery clinics and they do a lot of the routine visits. That allows you to see more complex cases in a controlled fashion rather than copping the complex cases as emergencies. [CL02 Obstetrician in regional hospital with high risk RHD population]

Making sure the woman has her echo; cardiology review; having the LA Bicillin regularly if she is someone on LA Bicillin, would be the role of the midwife, caring for her, to oversee that. And one of the challenges can be that we have alternating midwives. And in the past, that's lead to, I think, a breakdown in continuity of care. But that's improving. [CL03 Clinician Northern Australia]

An increased awareness among midwives was referred to:

Some of the wins over the last five years? That there's buy-in from the midwives and the Australian College of Midwives and Australian Council of Midwifery. There's a genuine interest in bringing it to the front. And wanting to know more about it. Initially there wasn't much known. And unpacking those issues related to RHD that are not just about medical management. …. So, I

think has been a growing awareness within midwifery circles and that there is a willingness to actually address it. That it's preventable. And it's not just another condition that needs to be suffered and tolerated. [SP01 RHD Program]

The key areas where we have high prevalence of RHD, there have been midwives who've been around for a long time … these people are under enormous pressure but they have a lot of knowledge and… and so I think their approach to women with RHD in pregnancy is consistent. [SP02 RHD Program]

Quality of staffing. Expertise. Turnover. FIFO & locums. Medical tourism. Structural gaps and barriers. Continuity.

A constant theme of the need for an appropriately skilled, respectful and well-resourced workforce was threaded through most interviews. Clinics and services described in a positive fashion universally referred to continuity of staff (quotes below also CL05 and check others).

There are some health centres where there is stability of staff and they work fantastically. And there are other places that the turnover is terrible and it shows [CL06 Obstetrician]

I get a little bit wary of – it's almost medical tourism - up here. Those who come up and sort of want to be involved in it for a bit. And I just think, you know, they just want to come up and manage [eg RHD] pathology. You know, it's actually not managing the pathology, it's managing the circumstance. Obviously you manage pathology but that's, you know… You can train a monkey to do that. But it's actually the bigger picture that I think gets glossed over. [CL01 Clinician in tertiary centre Northern Australia]

We're a teaching hospital which means we've got Registrars so we've got turnover. But they don't know then how to deal with what they're seeing. You get your cultural awareness course, which is this bullshit term for sessions that are a couple of hours, and that's better than a poke in the eye with a sharp stick but not much. There's no course that's actually going to give people any insight into what's going on unless they're actually confronted with it. [CL06 Obstetrician in northern Australia tertiary centre]

Clinicians commented on gaps in effective recruitment and a skilled workforce and the impact that had on quality of respectful care: [CL01, CL03, CL06, CL10, CR01, CR02]

You know, if you want to work in Antarctica you have psychological screening, you have training for a good two years prior to being selected. But if you want work in a similarly remote highly challenging environment looking after our most vulnerable people in the country, you just go. And you usually don't get any training. And you don't go through a vigorous selection process 'cause they're desperate for staff. [CR01 Physician researcher in northern Australia]

Juggling multiple roles and priorities was referred to by participants [CL07, CL13, SP03], particularly in the primary health care arena:

But my [midwifery] role here then expanded quite dramatically to include the family and children. And after all, mother is only well when their children are well. But we try to make sure we have one day a week that we concentrate on antenates. We've done that for a very long time, even before I came here and we have the same doctor every Tuesday that works with us. [CL07 Midwife in primary health regional with outreach]

One of the… I mean, and it's an issue for many areas and many regions, is that you have great people working as hard as they possibly can but they're terribly under-resourced. So… I… Not that I question their practice but I just question the amount of time they have to be able to provide women with the information as appropriately as the woman needs. So, again, I think there's a paucity of resources. So it's reliant on practitioners doing that one-on-one conversation around some of the issues. And, as we know, sometimes when someone talks to you, you hear about a third of what they say. [SP02 RHD Program]

The slow build-up of trust and how that impacted on care provision is described in one midwife's experience of her first six months in a community:

When I arrived in the community I was a single, young, white woman. So I had no say. I had to earn my respect and I found that really challenging. In the clinic, it was OK because I was seen as the midwife/nurse but outside of that, you know, I was a nothing really. By the end of six months, I had earned respect and was given a skin name. It was strictly women's and men's business. The day I arrived, I was sitting on the doctor's balcony and this very pregnant woman went by and he said, "She has had no care, you'll need to see her tomorrow." And of course she knocked on my door at 4 o'clock the next

morning. That was my first birth, yep. And I remember thinking, "Maggie Myles [exemplary midwife practitioner], eat your heart out". [CL10]

It was a strong community. We worked alongside the Nunkurri [traditional healer] who was a paid member of staff, as well as the traditional midwife. Women didn't want to go into town to give birth: they would hide. If you were going to birth on a community these were generally trouble-free births so I actually played quite a minor role. There was often a lot of people. I tried to stay on the fringe. And then we'd, you know, watch the sun come up together and later in the day, bury the placenta and have the smoking… Yeah. Very special. [CL10]

Models of care and clinical management

Existing literature on preconception and reproductive health care is predominantly focused on congenital heart disease. RHD-specific research to test the evidence is required to strengthen the rigour of recommendations, better understand the effects of pregnancy and choose the best individualised plan for ongoing care.

Collaborative cross-disciplinary care with health services, family working together.

One midwife described the model of care for remote-dwelling women in a midwifery group practice:

So, the women's pregnancies are usually confirmed in the community health centre and the midwife and/or nurse lets me know and a midwife from our practice is allocated. Each midwife goes through the notes of the women in her care by using the electronic health record system- so we'd pick up anyone who has rheumatic heart disease. We have one obstetrician who does a clinic with us one afternoon a week, so she sees all women who have come into town to wait for their baby or who require to be seen by an obstetrician. At the end of that weekly clinic, we have a telephone case conference with a health centre on a rotational basis with all of the staff who are able to be present. So, our Aboriginal health staff as well as midwives and the obstetrician and the midwife and/or doctor who's in the community. The obstetrician gets up the patient's record and we go through everything and check everything and she

orders treatment or interventions, where necessary. So, it means that women have early obstetric input into their care. And we work in collaboration of course with the RHD and cardiac teams and there's also an obstetric Registrar, who does some outreach visits to some of the communities. So during that time, when we have the teleconference, we also discuss other aspects of the person's care outside the clinical care. So, if they've got family issues, if they have other social or emotional concerns that are known to the clinic, then we discuss that. And how perhaps we can meet the needs of the woman when they come in. [CL04 Midwife]

Interviewer: You've got a strong conduit role, haven't you…
Interviewee: Yeah. Yeah. [CL04 Midwife in Aboriginal Mothers and Babies service]

So I think the standout thing for me is that there's just this grand continuity that every week I phone the community health centre for an … It also provides that opportunity for them to have some support. So, all that builds up a relationship and an open communication. [CL04 Midwife in Aboriginal Mothers and Babies service]

One regional hospital midwife commented that collaboration underpinned good outcomes (which included the ideal of a woman birthing as close to home as possible). But, she continued to reflect on barriers to that ideal being achieved, including underlying chronic disease and the 'causes of the causes' of social determinants. She concluded that these were not 'under her jurisdiction':

What works well? Look, it's always nice when you can get a woman cleared to birth as close to home as possible. And when the collaboration goes well. And I think the RHD coordinator is really good here. I think there's also difficulty when you're trying to manage a chronic disease in a hospital acute setting. I don't think people actually understand that if they actually got a woman well, either preconception and antenatally, how much better their pregnancy will be… But then again I don't think until you're going to address poverty, housing and domestic violence and alcohol, I think they're the four things that need addressing before we're going to see significant outcomes. And that doesn't actually fall in anyone's jurisdiction. Like, not mine. [CL11]

Working together: navigating paths, accountability, problem-solving. Workarounds

Navigating pathways and negotiating systems to get the right level of care can require occasional mediation to get the right collaboration. One RHD program described where a woman with moderate RHD seen by an outreach service was being transferred to a tertiary centre 2000km away for antenatal care, with the outreach midwife, and regional centre obstetric and cardiac specialists unaware of her transfer.

> *Information was coming back from the tertiary hospital and they made a decision in the end, after many meetings, emails, phone calls, that this woman with moderate RHD could be cared for regionally instead of the transfers, with all the travel involved, being away from her family: there was no need for it. [SP04]*

One participant with extensive experience in maternal and Aboriginal health in regional and remote setting called for improved accountability between and within community health services. She described her experience of silo-ed care and blocks to collaborating on program initiatives which served to compromise optimal care for women:

> *Take out the politics. Unfortunately, there are a lot of people out there with chips on their shoulder. And I think it gets in the way of having those conversations because they come in with an agenda. And they can't push those things to the side. I look at [community health service] and how much money there is for funding different projects that went no-where. It annoys me— I mean where's your accountability?* [CL10]

This participant continued to describe how she worked 'under the radar' with a peer in different community health service to work around the strictures that prevented the two services working together.

Agile approaches from committed clinicians who worked together to problem-solve was central for one midwife in what she saw as a successful Aboriginal Mothers and Babies service:

> *We have midwives who are flexible, adaptable. They're committed to what they do. And so they will go that extra mile. And along with the Aboriginal health staff I mean they are fantastic… They problem solve like there's no tomorrow sometimes.* [CL04 Midwife in Aboriginal Mothers and Babies service]

Reference to Clinical Guidelines and evidence based care

There was not unanimous reference to the RF/RHD Australian Guidelines[6] by those working in clinical settings. Most clinicians who did refer to the RHD Guidelines included these after mentioning others, with subsequent prompting. Guidelines that were referred to include the CARPA Women's Business Manual[391], Infectious diseases guidelines, Therapeutic Guidelines for Administration of Antibiotics, South Australian Perinatal Practice Guidelines, NSW Health policy guidelines on RF/RHD, as well as the RHDAustralia apps for phone and tablets.

> *There are no clear guidelines. And I've asked the question if, you know, at what stage of pregnancy should they have had that echo. Say you've seen them Week 8 of pregnancy, early on, and you've identified a murmur, when should they have had their echo. And I've just been told as early as possible. So I don't know what the standard is and if there is a standard.* [CL05 Clinician in tertiary centre with high risk population]

> *What I use a lot with ARF or RHD, is the RHDAustralia app on the iPad or the iPhone… and if I don't happen to have access to those I use the RHD Guidelines on the internet. And also CARPA Guidelines[391]. And then if it's something that falls outside of all of that then I usually speak to cardiologists...* [CL05 Obstetrician in tertiary centre with high risk population]

> *I refer to the Therapeutic Guidelines. That would be the chief one. And there's also… Oh… The red book. You know the red… Red and Gold? Yeah. It's Rheumatic Heart Disease – the official one*. [CL02 Obstetrician in regional hospital with high risk RHD population]

> *…for the community primary care, The Women's Business Manual[391] is there. Not just for heart disease alone, for every problem in pregnancy.* [CL05 Obstetrician in regional centre with high risk population]

> *What Guidelines for RHD women do I refer to? The 'CL12 Guidelines'. We have basic [principles] for everyone with cardiac disease … Whether they're having cardiac monitoring or not. ... There's the stock standard sort of monitoring. Saturation monitoring if they need it. ECG monitoring if they need it. Fluid balance monitoring and… and then we watch them closer after the birth for that first 24 hours. [CL12 Midwife in tertiary urban centre]*

I think we need to really understand what… And start to promote and encourage the use of the Guidelines in terms of earlier screening. That sort of stuff. [SP03 RHD Program coordinator]

Co-morbidities. Other chronic diseases

Competing priorities with other chronic conditions was referenced by several clinicians. Addressing other conditions particularly diabetes, renal disease, syphilis, and so on meant that RHD was not 'on the radar' [CL10,CL13]

But when you're in the smaller communities like that you're in the field, you've got to do everything. Yeah. It's not like— You can't focus on one thing. You've got to focus on everything that's happening and, you know, all the sick people, diabetes, everything. Which makes it really hard. [CL13 Aboriginal health practitioner]

Certainly RHD is one of our top priorities.. And I see our job as preventing serious complications for that RHD patient where they then need surgery. Whenever I'm talking with health staff in clinics. And supporting our RHD coordinators, in that message. We have an added responsibility to try and ensure when people get their LA Bicillin and also the echos. Although it's tricky around access to echos because we know who needs the echos, it's really just around them actually getting seen; about having access to enough echo services to get through the list.[CL03]

Similarly, addressing the need for individualised care parallels with other conditions that was responsive to women and evidence-based:

Of course it's not just RHD, it's across other diseases. What's the evidence-base, the best care for that person? Getting that woman to explain what they think is wrong with them and what they want to do. Understand what their options are. Being able to provide the education to the family, as well as that person. So that they can support her in that process. Finding out what the barriers are and helping them access the services they need. [CL08]

You may not come across RHD as commonly but when you do, you need to give it as much importance as you do diabetes. … I mean, every midwife has come across a diabetic with insulin needs and so on. Rheumatic heart disease needs that same attention.[CL10]

Cardiac care assessment & monitoring. Referral pathways. Risk assessment

> *…if it's a matter of doing something elective and then none of this has been done, it may… May result in the patient being sent to [regional tertiary centre] as an emergency, if the anaesthetist's not happy. So we like to do all our homework so that everything runs smoothly for the patient.* [CL02 Obstetrician in regional hospital with high risk RHD population]

Similar to many of the themes, there were multiple factors perceived that improved early diagnosis and models of care. The RHD program, improved resourcing and communication was described by one, supporting improved risk assessment:

> *I know since the Register began, women are identified earlier in their pregnancy. So, they've been able to access services a bit more quickly. The outreach midwife role started alongside the RHD position back in 2009. And they work closely in streamlining care, echo, cardiology appointments. Yeah, I think we just have much more improved communication between the obstetric team, cardiology team. And that's helped improve outcomes - in both ways. In getting high risk clients sent to [tertiary centre]. But also not just sending them over because the obstetric team have panicked and said, "Ooo! They've got RHD. Send them straight [to tertiary centre]", when they're very mild and they're quite capable of staying in their community and delivering surrounded by family and friends.* [SP04 RHD Program coordinator]

In the NT, system-level changes have seen the expansion in cardiac services (and funding) in the Top End of NT including incorporating RHD in pregnancy into standards and measures for cardiac care and health.

> *I'm heavily involved with cardiac health and currently chair the cardiac outreach rehabilitation committee. And attend the cardiac operational committee for Top End and the cardiac reference group, which is sort of the peak NT cardiac health group. Apparently. I think. Which has been involved in overseeing the expansion in cardiac services in the Top End with the increased funding from 2008. So I'm sitting in on behalf of my managers on those reference groups. And just recently, we've passed with a couple of others in organising cardiac health forums; one in the Top End and one in Central Australia around standards and measures in cardiac care and health for NT. And part of that was RHD. And I think… And one of the proposals is that we make sure that there are incorporated.* [CL03 Clinician]

Continuity of care. Conduit. Opportune point in time care. Trust. Juggling resources

The importance of team collaboration and the connections with family and community was stressed:

> *We recall our patients from 21 days, onwards, to make sure that they receive their bicillin every 28 days, what will happen is that Joan [RHD Aboriginal health practitioner] will do home visiting. Often it will be the older females in the family - Mum, Aunty, whoever, who's very quick to let Joan and the other health workers know when someone's expecting and get them in for all the appropriate tests. And then the women generally start volunteering and asking questions. So, generally it is a family affair. So what happens is, we'll either link them in with the maternity services. Or, they'll come up and present themselves. And then the obstetric team, nursing and health workers, will contact our program. Because we work very closely with cardiology and make sure that echoes are up to date, and so forth. And deliver the bicillin because often what we've found is that… That the women are more comfortable continuing within the same clinic setting to receive their bicillin.* [SP04 RHD program coordinator]

Achieving continuity of care with the right clinical staff at the right time was often challenging:

> *There are a number of factors that determine who goes [to appointments, to meetings]. Firstly, who's available. We often have women in labour as well so we have to stretch ourselves between women in the labour ward as well as those who are coming for appointments. The other thing that determines it is, what is the appointment about. So, if it's a serious clinical issue then it's obviously desirable that a midwife would go. Very often it will be with one of the Aboriginal health workers as well. Or it may just be one of the Aboriginal staff who goes. Yeah. So that we look at each situation and determine (a) who's available and (b) who's most appropriate.* [CL04 Midwife in Aboriginal Mothers and Babies service]

… but, it was seen as a crucial part of the coordination of the care that the midwife attended meetings, appointments including those not directly related to the pregnancy [CL04] [CL07]

I attended appointments with a young girl, who had RHD who wasn't happy to go to the cardiologist by herself so I went with her and I learned why she was reluctant to go by herself. It wasn't a very nice consult. Yeah. The cardiologist was a little demeaning. He wasn't happy that I was there, supporting her. Then the same day we went to see her physician, who'd known her since she was a baby 'cause of the RHD. And, you know, he was just so different, so lovely. it was just such a stark contrast. That girl ended up having to go to Adelaide and having a heart operation while she was pregnant, this girl, she was quite unstable so… It was a pretty big issue for her. Unfortunately, she's now died. [CL07]

Reproductive health, preconception care and planning

And when we discharge a post-nate, there should be a follow-up plan. So it should be, so and so wants an Implanon. I can put that Implanon in or I can get the midwives to put the Implanon in. That's great but some of them say they don't want it but it's like, well that's now, should be the responsibility of the clinic that you need to make sure that she does get the contraception that she wants or you do you have another conversation about it. [CL06 Obstetrician in tertiary centre with high risk population]

So, some of the things that we have been trying to do, particularly around young women, is try and get health providers to have those discussions around reproductive health for young women. So talking about contraception. And particularly the women who have been diagnosed as Priority 1 or 2. So, there would be, I would say, on the Register, in terms of Priority 1 and 2 patients, there would be about 150 altogether. And I would say the vast majority of those, so… And these are just rough figures, about 80 of them would be women in that reproductive age. So, what we try and do is get the health sites to have that conversation around contraception and planned pregnancy. It still is not an exact science and I would say that it… It's done as well as it possibly can be but there's a lot of room for improvement. So… So, yeah. So we try to influence those discussions health providers have around reproductive choices. [SP02]

The biggest [indiscernible] is for Implanon, actually. Yeah. So, most of them do, I mean, agree on that Implanon, for contraception. So that gives them about two, three years break to see how things are going and that. You know? So… I think contraception is a very, very important part, considering they've gone through the pregnancy with all these issues, for them. Because again, you can

have postpartum and OK if they can't [indiscernible]… You have another added burden of a pregnancy on a heart which has already been tested. So… I think that contraception is a good issue. Is a good planning to do. And that, I think, you know, you keep advising how next step will be the conception after delivery and… Quite a few— Quite a few of them are susceptible— Almost all, I would think have gone through some kind of a contraception to space out the next pregnancy.[CL05]

Diagnosis - under-dx, missed, late dx RHD, Identifying & notifying women with RHD-P. Screening.

Strategies to pick up RHD among women in a prevalence region were described:

With RHD in pregnancy, the focus should be on identifying it. "Beware! Has this person got any history suggestive of rheumatic heart disease?" *If you think this woman might possibly have it, get an echo. And refer. Discuss. Ring someone. And we've got a culture where ringing people is generally pretty OK. Or you have a main obstetrician who responds very quickly to emails and all the people know that that's how you contact her.* [CR02]

The same clinician went on to comment

One participant described her experiences where RHD was not monitored, often with little consequence in the first pregnancies, until …

Yeah, until she flatlines intrapartum in pregnancy number three, usually when her RHD hasn't been picked up or addressed antepartum. [CL14]

Having a specific RHD and pregnancy clinical checklist was not referred to, but flags to identify women were used in one Aboriginal Mothers and Babies service:

Because we're fortunate enough to have an obstetrician, and others, to work with, then we don't have a checklist ourselves. We don't have a sticker system on the notes for women who have RHD but, we do have usually ['RHD'] in big letters on the handheld record … So that's our visual thing. And of course there are the alerts in PCEHRs. [CL04 Midwife in Aboriginal Mothers and Babies service]

But protocol-ising things is what makes things happen in health care. Unless you have the tick box that they've had their VDRL test or HIV histology done, the busy clinician is not going to think of that. Similarly trying to detect rheumatic fever in pregnancy, they're trying to think about blood pressure and such – but they need to have the tickbox for RF/RHD - it's what makes things happen. [CR01 NT]

Another midwife discussed a shift in awareness in how RHD was flagged and discussed with their clients:

All the Aboriginal health workers and midwives now feel really comfortable about talking about to the women, "Have you thought about RHD?" *You know, before we wouldn't have even thought of it. Now there's that little bit of knowledge where they can investigate and look and know that the things you look for an echo in the previous notes. Look at some pathology. Look at the history. Look at the family history. Yeah. A lot more investigation I think than we did previously.* [CL08 Midwife working Midwife in Aboriginal Mothers and Babies service]

An obstetrician described care pathways for women that fostered continuity of care in an outreach model that she and a colleague had extended at a regional hospital. They contrasted the experience of establishing this specialist outreach service rather than the previous system that used visiting specialists based over 2000km away.

We included some more communities after I joined - we were very passionate about this. I will see the woman in, say, in one of the communities and then in the women's centre at the Aboriginal medical service and then she comes for delivery to (regional) hospital so, you know, you do get to know your patients. [CL05 Obstetrician regional centre with outreach]

One midwife - who had worked in remote Australia and was establishing an Aboriginal Mothers and Babies service in a regional centre - described a potentially catastrophic event:

A woman came in: pregnancy number four. She was monitoring her own health. I first met her when she was about 26 weeks' pregnant coming into hospital. There was RHD in her history but the impact that was having on her pregnancy wasn't acknowledged. It was quite traumatic. I remember her

tummy. It was full and hard and difficult to palp the baby. She had a chest x-ray and the Resident doctor was saying it was an exacerbation of asthma. I said, "Listen to her. It's cardiac - I think she is drowning from heart failure". And he looked at me and he said, "It's got nothing to do with her heart …" and "I'm the doctor", basically. I remember feeling so desperate: I went up to Maternity. The visiting obstetrician came downstairs with me, wiped the floor with the Resident, got a bedside ultrasound. And then a visiting physician assessed her cardiac function, and she was shipped down to the tertiary centre straight away. I remember when I rang her saying, "Oh my God! I can breathe now". *Yeah, she spent the next six weeks in Coronary Care, went across to birth, came back to Coronary Care. And, I think, you know, we could have lost her.* [CL10 Midwife in Aboriginal Mothers and Babies service]

Women who did not have a good shared understanding of the impact of RHD, particularly in pregnancy, were paradoxically less likely to seek care -and more likely to need emergency interventions. Getting the right level of care in a way that promoted understanding was described by one RHD Program coordinator:

Pregnancy and delivery are incredibly medicalised for Aboriginal women living in rural and remote areas. And so what tends to happen is that many women have to deliver away from family, away from community and away from Country and I think that adds an extra complexity. …Also, I believe, make them more reluctant to seek medical attention early. So I think there is quite a lot of work that needs to be done in terms of appropriate patient information that's made available around pregnancy. And also a heightened awareness for service providers as well. Particularly remote area nurses who are expected to be multi-skilled around the importance of information given to women at every stage. Unfortunately, this is an area that we [RHD program] haven't specifically focussed on. What we have tried to do, as a program is work more around support for the providers to at least ensure that women are safe. [SP02]

The health information system deficits described earlier had direct impact on referral pathways:

I find it ironic 'cause the NT is often held up as a poster child of eHealth which is appalling. We really rely on the obstetricians to refer patients to us. If they have any engagement in antenatal care, we usually find out about them, and they get referred to us. So, if they slip through that net, the only way we'll find

out about them is through the RHDAustralia alert system that comes up on our electronic medical record, if you want to call it that. [CL01 Clinician]

Screening. High risk populations. History-taking. Echocardiograms

The need for earlier echocardiograms was mentioned by several participants who called for a low threshold to conduct this test in undiagnosed high-risk women where indicated:

> *Understanding that if they're presenting in pregnancy and they're overdue for their echo or they haven't had an echo for the last 18 months, perhaps that should be a standard, that we implement certain clinical care arrangements that wouldn't necessarily form part of the standard care plan for pregnant… For antenatal patients.* [SP03 RHD Program coordinator]

The concept of an echocardiogram screening program for pregnant women was considered in one high prevalence area, which would entail pregnant women having echocardiogram at the same time point as an ultrasound. Logistical and operational barriers and the usual juggling of funding priorities was a consideration in this being implemented, and the word 'potential' was used a lot. [CR02]

While the benefits of training Aboriginal health workers to perform these echocardiograms were considered, having clinicians with existing ultrasonography skills was thought a priority to make it most viable at the outset:

> *The reason we chose them is that they would potentially be interested and be trained relatively quickly. There's this personal gain for the people involved and not much effort. Whereas if you choose someone who has basically never used an ultrasound, then, the whole process of training people is going to be more logistically complex… But the point of this was to screen pregnant women on the agenda. And then, from there we could go to, "What we might do next and who else could be trained?". So, it was a pragmatic sort of thing.* [CR02 Clinician researcher in regional high-prevalence area][10]

Regardless, a consistent theme was the call for a lower threshold for conducting echocardiograms for pregnant high-risk women [SP03, (others):

[10] As of early 2019, there are no known echocardiogram screening programs for pregnant women that are active in Australia, although one is being considered in Darwin.

> *But what does and should happen and could happen is a much lower threshold for doing echoes. And we're going to start training a few people to do quick look at valve-type screening echoes.* [CR02 Clinician researcher in regional high-prevalence area]

High risk pregnancy and birth - clinical care

Indigenous health

Lessons from other models of care, diseases

Secondary prophylaxis

One of the threads running through interviews, particularly with those clinicians who did not regularly have women under their care with RHD, was the need for more explicit care instructions that were immediately available such as secondary prophylaxis :

> *But it's the duty of care with the cardiologist where there was something there. OK, what is it? Because what does that mean? And is it rheumatic? And I don't think I saw that written in any of the echo reports that I've been given. It never says, "This is rheumatic heart disease. Remember that you need to give [secondary] prophylaxis".* [CL14]

Shared understanding. Cultural constructs and contexts.

Transfer. Planning, strategies, impact. Location of giving birth

Participant perspectives were mixed about the impact of transfer:

> *Maybe I'm living in La-La-land but I think that it generally works well. It's really important in terms of outcome to recognise that some women need an appropriate service that has more resources or abilities than we can provide [in regional tertiary centre]. So we've had a couple who've been transferred interstate*. [CL04 Midwife in Aboriginal Mothers and Babies service]

Where it was depicted as particularly traumatic, it was often associated with other aspects such as late diagnosis and complications leading to emergency transfer - often vast distances from home, confusion or lack of knowledge about RHD severity, overlaid with lack of accompanying family support, and concerns about other children.

> *Particularly if they have severe disease, it certainly… If they don't have the information, or they're not given information, or the information is not*

acceptable to them and only they are confronted with multiple procedures, multiple flights, multiple trips away from home and country, it can sour their experience. [SP02]

Using data to inform better practice was mentioned by some clinicians:

We were invited to a review of the top Aboriginal health facilities to discuss on why they think they're doing good. A lot of that's to do with our audits which is showing how well we're doing. But we always have a meeting afterwards with the report to discuss what we haven't done very well and what we can do about fixing it.[CL07 Midwife in Aboriginal health service]

Access to services

Barriers to services ranged from practical considerations about transport, distance, accommodation, resourcing. There was discussion also about broader underlying issues such as poor infrastructure, competing family needs, language barriers to shared understandings of disease and accessing treatment, and system-level blocks particularly for women with multiple communities.

Coming to town - accommodation for mum, kids. Coming to hospital - appropriate buildings

What would make care for pregnant women with RHD better? Tangible things include improving the quality of accommodation. You've been to [town hostel]. That hostel is horrible. I wouldn't send my dog there. That's considered appropriate accommodation for people from the bush? [CL06 Obstetrician in tertiary centre with high risk population]

Balancing family needs was described as an important factor that impacting on women who may need to travel vast distances for examinations such as echos or specialist appointments, and would impact on women feeling that they couldn't travel away from community: (CR02, CL03]

… But there are particular issues for significant rheumatic heart disease because, you know, it is often managed a long way from home. Women with RHD who need transfer often already have a few children. Who's going to look after the other children when a woman gets transferred, things like that. That was a major obstacle for one woman. In terms of going to [the tertiary centre a few thousand kilometres from home] to stay for a while to get assessed and

treated and then having to stay on … She refused to stay because she didn't feel her partner would look after them properly. [CR02]

And also if they're still breastfeeding but the child is over a certain age, I think it's over one, the child's not allowed to travel with the parent. [CL03]

Another commented on what they saw as the lack of foresight in terms of infrastructure design in a hospital where over half its patients were Aboriginal:

And this hospital is an utterly inappropriate building. Aboriginal patients want to be outside or… on ground level. And what have we got? We've got a high-rise with walled-off outside areas. No safe zone outside and a footpath outside the front of the hospital to sit on. [SR01]

Understanding what? Language and Interpreters

Few health providers (in regions that included high numbers of Aboriginal women with English as a second up to fifth or sixth language) referred to use of interpreters, and those who did acknowledged that there were gaps in service:

We rarely use interpreters. Perhaps we should more. But we rarely do. [CL04 Midwife in Aboriginal Mothers and Babies service Northern Australia]

Others recognised deficiencies and reflected on some of the reasons, but commented that maternity services in their hospital tended to be more responsive:

The problem with the episodic care, and the opportunistic care, is we don't often have good interpreters or an interpreter booked and we sometimes rely on family members which is not ideal. But that's a universal problem, as you can probably understand, up here. It's often having a coordinated response. So, having the right interpreter booked at the right time. When we're meeting with a woman at a booked time, it's done reasonably well. Certainly better in the obstetric setting than any other setting I've seen. I think 'cause the midwives are better attuned to it 'cause they're seeing these people in community. And I think… I just think in obstetric service has a better grasp on that than surgical services.[CL01]

One clinician contrasted their current experience of working with interpreters compared to that with medical interpreters in urban settings, highlighting other issues beyond word interpretation:

> *What I do get frustrated with is the quality of the Indigenous interpreting service sometimes. So previously I was at [an urban] hospital with a very high Mandarin and Greek speaking community so you had a very professional interpreting… Whereas here, you can explain something that's quite complex and use fairly straight language and you can say an entire sentence and it gets summarised down to about two words and you just think, "There's a lot getting lost in translation here" and… I don't know if that's due to cultural nuance or what it is but you can feel very frustrated.* [CL01]

One practitioner who worked in a remote community spoke of work-arounds, as well as suggesting that more resources in the local language would benefit women:

> *Ah, yep. So we have no trained interpreters. And I think we wrongly rely on family and Aboriginal health practitioners for complex discussions. So I don't think we could really ensure that people are understanding what we're saying but… I s'pose we could use, on the phone, the interpreters. We don't do that. And I don't know what the access to that is actually. Yeah. There are resources in the local language for first trimester screening and I think we've got the opportunity to develop more resources in the local language because there's so many who speak it here.* [CL03]

Similar difficulties were described in often fraught situations where non-English-speaking women were transferred from the Pacific region to a tertiary centre for (usually emergency) care with limited ability to interpret due to dialect. [CL09]

> *And she was terrified, clearly terrified yesterday, when she was going into theatre.* [CL09]

Transient women – impact on access

The logistics impact of providing optimal care where women move between communities and towns and jurisdictions highlights difficulties in sharing information and resultant impact on continuity of care.

Findings from a study of secondary prophylaxis adherence found that there were typically three homelands and three locations where a person may go [CR01]. Workarounds based on established collaborations helped in maternity care:

> *Trying to keep up with people that sort of float around communities is hard. We keep up pretty well with the antenatal clients because the midwives are a*

bit obsessed about them and if there's a midwife in the community, you know you've got a person that you can call and talk to. It doesn't always work for everything else. And, sometimes it falls down in communities where they haven't got a midwife. Also, we have a regular meeting for midwives from the hospital and around the region to discuss issues and make sure we're all on the same page and all doing the same things. Plus any other interested people like the diabetic educator, community health nurse come sometimes. It makes a difference. So clients don't get lost in the system. [CL07]

Getting there: transport access and logistics. Tyranny of distance

Travel and coordinating appointments were considered logistic problems that took up much time, but were part of the package that supported continuity of care:

You know, we need to simply ask, "How are you going to get to the appointment?" Transport's a real problem. So if you're four hours from your appointment, and you haven't got a car or it's not registered or there's no money until payday for petrol, how do you get there? And then you haven't got anyone to get your children from the school or you're thinking, "Oh… I've got to get there because no-one's going to be with them tonight". It's a huge expectation. Sometimes if the Aboriginal health worker can take the person to the appointment, they feel safer. They feel like there's someone there advocating for them. And things are put in place so that worry's gone. And helping them arrange with Aunty or whoever, to pick the kids up [CL08]

One midwife described that the contract for echocardiograms with a private provider in her regional hospital dictated specified tests would be done on the weekend.

I'd say, "Why are we getting girls in for a Saturday?". *It's 'cause they're having an echo. And then we'd wonder why they didn't turn up unless [the RHD coordinator] ran around to pick them up. On a Saturday. Yep. The contract has changed now.* [CL11]

Another spoke about the echocardiogram machine being broken [CL07] in a high-risk regional centre: consequently women needed transfer some 300km away for their echo. (This was noted also with frustration by another clinician not interviewed for this study. When discussing an appropriate time to visit the hospital during the RHD-P research project data collection, they opened the conversation by drily asking if I could provide an echo machine).

The impact of severe weather created another layer particularly in remote communities:

> *We're meant to have three cardiology visits - I'm not sure if we will this year 'cause we've had cyclones which disrupted things a bit.*[CL03]

The tyranny of distance and seeming arbitrariness based on location shaped delivery of care trajectories that nudged the bizarre. Intra-jurisdictional systems did not talk with each other: *'… and when you're talking about the Centre and the Top End, you're talking about two different entities which is completely insane.'* [CL03]

Border

A woman who lives in one remote community near the border of Northern Territory and Queensland may visit up to five different hospitals straddling two (possibly three) jurisdictions in the course of her pregnancy depending on her state of health:

> *The obs checks are in Mt Isa [Queensland] so that's where they come for ultrasounds and so on. But the midwife for her community antenatal care is based in Alice Springs [Northern Territory]. And then they fly here to Isa to give birth. So, midwifery care is in one jurisdiction, the tests and birth are in another – about 1500km apart. Then, they'll go to Townsville if they're not well which is the referral centre. Or Brisbane [2100km away], depending on the severity. Or, if they are in Alice at the time and get complications, they'd probably go to Adelaide [South Australia].* [CL011]

Dental care

Access to dental care was referred to by one participant. This standard element of cardiac care[192], apart from addressing specific dental health needs, prevents potential worsening of cardiac function through infections such as endocarditis:

> *Well, one of the biggest issues we've had with RHD ladies was getting their teeth done. It sounds ridiculous, I know, but our dental care here has really been ad hoc over the last few years. Nobody wants to come here and work. It is getting better because now we're actually got funding so we can pay people to go to private dentists.* [CL07]

Women with RHD

Who are the women - country of origin if not Indigenous. Refugee women

Discussion was almost entirely focused on Aboriginal and/or Torres Strait Islander women for most of those interviewed but a small number included reference to non-Indigenous women, based on their own experience:

> *In fact, [previous to working in northern Australia] most of the rheumatic heart patients I've managed, from a purely clinical point of view, would have been South East Asians [in Sydney]. So they're another group that has a high incidence of rheumatic disease.* [CL01 Clinician working in urban tertiary centre in high prevalence region]

Another spoke of a woman with a mechanical valve from the Pacific region transferred to Australia for care, who had gone into heart failure and was being ventilated, while her newborn baby was in neonatal intensive care.

> *So here's a person living remotely where there's not been access to anticoagulation monitoring. And this is her third pregnancy. Her other two babies died at 20 weeks or so. She's really had the worst experience. In terms of everything, you know. There's no one been with her here. Finally (her country's government) has allowed her partner to be with her because she's so unwell.* [CL09].

This extreme situation paralleled that of Aboriginal women from remote communities with significant illness[20].

Care seeking

> *Women do have an awareness that their heart function can be compromised with a pregnancy, I think, a lot of them do understand that. And, they want the best outcome for the baby and themselves. So, they're very quick to let you know that they're pregnant.* [SP03 RHD Program coordinator]

One obstetrician who reflected on the improved care she considered had occurred – associated with improved continuity of skilled staff, better resourcing particularly with hospital-based cardiology, the RHD program, and the community-based Aboriginal women's health service which meant that women needed to go to the hospital less – talked about the 'Town X methodology' for models of care:

I don't think there's any fixed formula which works. So, you could be going to Coles and then you will see your patient and then she looks at you guilty. But, you know, also there is "The Bush Telegraph". Like you pass information to some family that, you know, you really want to see somebody because you are worried. And then, a woman who you've been chasing for months turns up and says, "Oh, I would really quickly want to see you", you know? How do you describe the methodology for that one?! The thing is to not give up. Just because it could've been five times 'did not attend', does not mean it's going to be 'did not attend' forever. [CL05 Obstetrician]

The irony that often women at the most risk of RHD and with the most need to have early diagnosis, monitoring and management were more likely to be the ones who did not seek early care was commented [CL05, list others]

One participant reflected on some of the reasons why women may give low priority to their care:

Cause I… just think if you're in a domestic violence situation with alcohol misuse, who's going to think about a monthly injection? [CL11]

Building relationships. Cultural safety, right communication

And for women to have … their own midwife and her two colleagues, that they know are here for them when they come in to wait for their baby or to have any appointments, is a hugely important confidence builder. I think it builds confidence in the system, that it's a system that is actually caring for them. But it also builds up a sense of trust that where the woman knows that whatever problem she might have, then we will try to assist. [CL04 Midwife in Aboriginal Mothers and Babies service]

The use of mobile phones also helps keep the coordination happening. Women are given the mobile phone number of their midwife. And the midwife has their mobile phone number. So if an appointment is missed we can find out why, remake it or whatever we need to do. [CL04 Midwife in Aboriginal Mothers and Babies service]

Educational resources - availability, appropriateness, language

Information and educational resources that promoted a shared understanding with women of their disease and its impact in pregnancy drew on a skilful approach that wasn't rushed and was appropriate to that woman.

It was Joan [RHD Aboriginal health practitioner] who alerted me, 'cause I was ignorant, that they didn't understand what I was saying at all. And that's why the heart models are invaluable. And education takes time. It can't be done quickly. You can't rush that education 'cause it is important. But you do sadly have to sort out a lot— To get that one-on-one quality time, you do have to sort out a lot of social issues. And once that's under control then you can sit down calmly and explain why we're doing what we're doing. And sometimes we can't solve all those social issues. You know, we can't solve it but we can make it a little bit more functional. If you know what I mean? [SP03 RHD Program coordinator]

The thing I find most offensive in relation to the way that we deal with Indigenous patients is the way that we do dumb it down. Which is crap. There are times when you do need to treat children as children. I mean if you've got a 13 year old, yes, you've got to deal with the fact that they're a 13 year old. You've also got to deal with the fact that they're pregnant. There's layers on layers of complexity. And in these complex Western medical constructs we talk about things that are difficult to understand for someone who's got your own cultural view of the world and who speaks your language when they have no medical knowledge – let alone when they don't share that. [CL06]

How the education is delivered was considered as important as the content:

All of our education is done opportunistically, really. A lot of it is done in the car. …And, as you would know, the response to formal education on a structured basis isn't necessarily appropriate in these circumstances. We don't have that first visit to the MGP, "This will happen and this will happen and this will happen". It is less structured than that. [CL04 Midwife in Aboriginal maternal health service]

Well it'd be nice to see some easy read literature for the women and their families. And I am looking forward to the online learning package. Yeah. As a NUM I just think that I've got many things running through my head at any one time. [CL11 Midwife]

Shared understanding of management and planning.

Several interviewees talked of the gap between what was said in conversations with women and what was understood.

> *You've got a pregnant woman who's got a mechanical valve. Should she change to Clexane? But the thing is has she actually been taking the Warfarin anyway? So she may or may not have been taking her Warfarin and that in itself is problematic. So if you're then going to anticoagulate with Clexane, you've got to talk about twice daily injections. If she's going to do that at home, where is she going to store her Clexane? Does she live in a house with a fridge? Does she live in a house with 16 other people, who have also got access to that fridge and half of those are kids?...* [CL06 Obstetrician in regional tertiary centre with outreach]

> *And we can talk until the cows come home about informed consent. We can talk about health literacy. We can talk about all this type of stuff. It's not there. A number of years ago we introduced the worst consent form in the world in this hospital. It was designed by lawyers. It is dreadful. But it has all these things. And it says, "Yes. Yes. I understand. I understand" 3— 15 different times. And I maintain that probably more than 90% of my patients do not understand what they are signing.* [CL06 Obstetrician in regional tertiary centre with outreach]

This disconnect could apply to Aboriginal health practitioners as well.

> *We get so used to the jargon that we don't realise that 90% of people have no idea what that means that… Yeah. But, I mean, even yesterday we had a meeting with our staff and health workers.. and I was looking around at everyone thinking, "I don't think the health workers even understand these words", you know, like, "prophylaxis" and… Even things like that, you know. I think that they kind of switched off after a while 'cause of the language. Get to know how much they understand and how much they don't understand and sometimes you get really surprised when you find out that they've listened to [indiscernible] which you think is really… Totally in a way you never expected them to but… Yeah. Yeah.*

Understanding of RHD disease. Health literacy for women

> *We have a strong woman worker who doesn't have a medical background. We have an Aboriginal health practitioner who has a very strong maternal and child health and disease base understanding. The level of knowledge of midwives who, when they first come here… needs always to be built on. But what I know we do very well is to talk to women about the effect of rheumatic fever. And what we can suggest in the hope of preventing it. So particularly things around sores. And that's really something that we do talk about as part of that care of the baby.* [CL04 Midwife in Aboriginal Mothers and Babies service]

> *When they say they've got holes in their heart you investigate a bit more. Find out what they've been told. We ask…* "Had you previously had a sore throat? Had you had Bicillin injections?" *You know. And they say,* "Oh, yeah, yeah, yeah, I was really sick. I was in hospital for a long time but I didn't really understand what they were telling me". *So, we find out more. If the diagnosis ends up being RHD we talk about that.* "Look, have you heard about RHD? It's a bug that can attack your valves of your heart and make you sick. If you take these special antibiotics that can help. These needles don't harm the baby". *The reassurance is important because women get scared for them and their baby. The reassurance is important to help make sense of it.* "Oh, so, I caught a bug and it's affecting my heart". *Yeah.* "And that's why I've had sore joints and other things going on." *Yeah.* "So I need that medicine to make me better so I'll go and have my medicine." *Yeah.* [CL08 Midwife in Aboriginal Mothers and Babies service]

One obstetrician reflected on skills that extend beyond clinical skills and language that extends beyond words: the cultural constructs and contexts that shape communication and knowledge and impact on shared understanding:

> *But yeah, when we're training someone to do medicine, we basically teach them a new language. And we have a way of communicating between us which allows us to impart information in a certain way, with the cultural construct within (that) language. So, this is what we do with medical students and we turn them into doctors. That then gets overlayed by peoples' own cultural background: from an Indian background or from South East Asia or an Aboriginal Registrar but we all talk the same language. We have our own*

things that we lay on top of that, but we are still all talking that same language. And it all surrounds the construct of "this is what western medicine does". But then as soon as you put that construct into circumstances where you've got somebody who doesn't speak your language - and that's most of your patients [– it's a different world]. Trying to get that across to trainees, to young doctors or old doctors for that matter, is not an easy task. [CL06 Obstetrician in regional tertiary centre with outreach]

The obstetrician continues on how women's priority-setting and decision-making about health was mediated by [women's] life situation, family and community as well as cultural constructs:

Then on top of that, you're dealing with a whole different context and cultural construct of health, wellbeing; what's important; what's not important; how we do certain things. Why women want to be on Country and not in town; why they won't come in town and sit around and have blood sugars done or whatever when they have important things you need to do or there's five funerals backed up. [Well], that's a very hard thing for medical staff to wrap their heads around. Medical staff make assumptions based on what they've said to women. "I have explained this". Well, you may have said the words (to the woman). But what do those words mean in relation to that other person? We're travelling along parallel pathways. And that's difficult for a lot of doctors. [CL06 Obstetrician in regional tertiary centre with outreach]

The obstetrician described blurred boundaries, tensions between process and outcome:

Especially when new staff come in, that there's sort of this attitude of … paternalism towards these women or they're chasing them around or they're doing certain things and it's like "Well, we've got to make a choice. We are providing a service and we want certain outcomes. If we want certain outcomes then we have to friggin' do it". We try education within our own construct. And then you get all the "Oh, but it's disempowering", "They need to make their own choices" and it's like, "Well….they're making choices about what? They don't really know what they're making choices about. And we don't know what they're making choices about. We don't know what the choices really are". [CL06 Obstetrician in regional tertiary centre with outreach]

I had a patient who was pregnant. She was in ICU with a pericardial effusion. She signed herself out of ICU, to go and pick up her kids, to take them to an appropriate place where they could be looked after and she came back again. But this is what, you know, this is a woman who… You can imagine. [mimicking laboured breathing]. You know, can't breathe, has got a pericardial effusion. She did the right business and then she came back. So it's not that people don't understand that they're sick. They know that they're sick. This is the stuff that a lot of doctors find very challenging. [CL06 Obstetrician in regional tertiary centre with outreach]

You try and negotiate your way through what [the woman] needs to do versus what we see as being we need to do in terms of trying to maximise her chances of having a slightly less complicated pregnancy, as distinct from a disaster. And that's not easy for people to do 'cause it's not 'best practice'. [CL06 Obstetrician in regional tertiary centre with outreach]

So, I think patients engaging in their own health is the biggest challenge. I think in all specialties, not only just pregnancy alone. Yeah. I think they are immersed in so many other issues, you know, housing, food, domestic violence. I don't— sometimes I think pregnancy and rheumatic heart disease is a small, small part of their life. [CL05 Obstetrician]

Family, community, environment, support

If you live in a community, and people see you outside of work, and they understand that you understand the challenges facing rural and remote Australians, and Indigenous Australians, they are a lot more accepting. When I first came here, they would say to me, "Oh… Do you just come here with the doctor?". "No, I actually live here". And then over the years, taking children to sporting events, down at the football. The patients would, you know… They would come up, and talk to me about different things. You'd see them down the street. If we were out fishing, or— You'd run into people and they gradually learned that, yeah, I was actually part of the community. I'm not just a health professional, who comes in, dictates the terms but lives a completely different life to what they experience. Yeah - and it's been wonderful from my perspective too, to actually see the reality of it, and act as that link between high grade services and see the reality for people who live out here. [SP03 RHD Program coordinator]

The time that I have seen it work really well is when there's been really good involvement from the family, especially they have to go to Adelaide or somewhere for an operation. I saw one girl from bush once who apparently everybody had tried really, really hard to get to go to Adelaide for her heart operation. And she just had a baby. I didn't really know her very well but I just kept going and seeing her every week and eventually a family member said, "Why don't we see if her Aunty will go with her?". So Aunty came in from bush. And she went off and had her operation and she came back and well, she just looked like a new person. And I don't really feel like it was me that really achieved it. We found the right Aunty, that's what we found. [CL07]

Secondary prophylaxis - fear of impact on baby in pregnancy

And that they're often very concerned about having the bicillin. And if that will impinge on the baby. Yeah, so they're very quick to ask those questions. [SP03 RHD Program coordinator]

One of our grandmothers described taking her grandchild for an echocardiograph, that when they said, "No, no, it's OK. You can see there that there's a bug in your system that is there and it is around your heart. Except by having injections and things you will be OK. And if you have those injections every 20, 28 days then everything's going to be OK you just need to have those injections to get rid of the bugs". It helped make sense of it all.

We know that some health centres do very well at the secondary prophylaxis. And some do not. My [experience at senior policy and government levels] suggests that some of it could very well be related to a high turnover of staff. [CL04 Midwife in Aboriginal Mothers and Babies service]

The big picture. Politics of RHD. Advocacy. Other RHD research

That the care for women with RHD during pregnancy was thought to span beyond clinical treatment was evident [CL01, SP01, others]

We're starting to, unpack issues related to RHD: that's not just about medical management. And it's really highlighted some institutional racism. And how we need to be doing more in cultural competence. And expanding our own worldview on how a pregnant woman might be experiencing her day-to-day life. That it's not the norm and that something can be done about it. It's a little bit like skin sores, scabies, snotty noses and weeping eyes and puffy ears in

Indigenous communities. Actually these things are not normal. [SP01 RHD Program]

Appendix 6: Published articles

Study one, Chapter 4

Received: 14 January 2019 | Revised: 24 April 2019 | Accepted: 25 April 2019

DOI: 10.1111/birt.12435

SYSTEMATIC REVIEW

BIRTH ISSUES IN PERINATAL CARE WILEY

Standardizing clinical care measures of rheumatic heart disease in pregnancy: A qualitative synthesis

Geraldine Vaughan[1] | **Angela Dawson**[1] | **Michael J. Peek**[2] | **Jonathan R. Carapetis**[3,4] | **Elizabeth A. Sullivan**[1,5]

[1]Faculty of Health, Australian Centre for Public and Population Health Research (ACPPHR), University of Technology Sydney, Sydney, New South Wales, Australia

[2]The Australian National University and Centenary Hospital for Women and Children, The Canberra Hospital, Canberra, Australian Capital Territory, Australia

[3]Telethon Kids Institute, University of Western Australia, Nedlands, Western Australia, Australia

[4]Perth Children's Hospital, Nedlands, Western Australia, Australia

[5]Faculty of Health and Medicine, University of Newcastle, Callaghan, New South Wales, Australia

Correspondence
Geraldine Vaughan, Faculty of Health, Australian Centre for Public and Population Health Research (ACPPHR), University of Technology Sydney, Sydney, Australia.
Email: Geraldine.vaughan@uts.edu.au

Funding information
This work was supported by the Australian National Health and Medical Research Council (NHMRC) project grant #1024206 and NHMRC Postgraduate Scholarship #11332944; University of Technology Sydney Chancellor's Research Scholarship; and END RHD Centre of Research Excellence, Telethon Kids Institute, University of Western Australia. Sincere thanks go to the facilitators and participants of the Twelve Weeks to Publication Program funded by Faculty of Health, University of Technology Sydney.

Abstract

Background: Rheumatic heart disease (RHD) is a preventable cardiac condition that escalates risk in pregnancy. Models of care informed by evidence-based clinical guidelines are essential to optimal health outcomes. There are no published reviews that systematically explore approaches to care provision for pregnant women with RHD and examine reported measures. The review objective was to improve understanding of how attributes of care for these women are reported and how they align with guidelines.

Methods: A search of 13 databases was supported by hand-searching. Papers that met inclusion criteria were appraised using CASP/JBI checklists. A content analysis of extracted data from the findings sections of included papers was undertaken, informed by attributes of quality care identified previously from existing guidelines.

Results: The 43 included studies were predominantly conducted in tertiary care centers of low-income and middle-income countries. Cardiac guidelines were referred to in 25 of 43 studies. Poorer outcomes were associated with higher risk scores (detailed in 36 of 41 quantitative studies). Indicators associated with increased risk include anticoagulation during pregnancy (28 of 41 reported) and late booking (gestation documented in 15 of 41 studies). Limited access to cardiac interventions was discussed (19 of 43) in the context of poorer outcomes. Conversely, early assessment and access to regular multidisciplinary care were emphasized in promoting optimal outcomes for women and their babies.

Conclusions: Despite often complex care requirements in challenging environments, pregnancy provides an opportunity to strengthen health system responses and address whole-of-life health for women with RHD. A standard set of core indicators is proposed to more accurately benchmark care pathways, outcomes, and burden.

KEYWORDS
access, and evaluation, best practice, health care quality, pregnancy, rheumatic heart disease, social determinants of health, systematic review

Abbreviations: CARPREG, CARdiac disease in PREGnancy risk score; CDiP, cardiac disease in pregnancy; NYHA, New York Heart Association functional class (I-IV); RHD, rheumatic heart disease; RHD-P, RHD in pregnancy.

1 | INTRODUCTION

Rheumatic heart disease (RHD) is a preventable disease of inequity. It is twice as common in women,[1-4] creating added risk in pregnancy. There are many challenges to providing optimal care for women with RHD, particularly in low-income and middle-income countries. Service provision is limited by poorly resourced expertise and facilities with barriers of distance and cost. There is often deficient awareness for women and health services of RHD and its impact in pregnancy.

Consequently, the higher prevalence of RHD in pregnancy (RHD-P) in low-income and middle-income countries is matched by poorer outcomes than in high-income countries, with documented maternal mortality rates of up to 37%.[5] Its burden is also high among vulnerable populations in upper-income countries. In Australia, Aboriginal and Torres Strait Islander women are over five times more likely to die from RHD,[6] with RHD-P rates for Aboriginal Northern Territory women up to 63 times those of non-Indigenous women.[7] Inequitable outcomes are also seen in Māori and Pasifika women[8] and First Nation populations in North America.[9,10] There are growing numbers of women with RHD in high-income countries as migration from resource-poor countries increases.[11,12]

There are no known systematic reviews that describe approaches to care and associated reporting measures for women with RHD-P globally. A review of the burden of antenatal cardiac disease in South Africa has a strong focus on RHD.[13] Guidelines refer to all cardiovascular pathologies in pregnancy[14] or are referenced in non-pregnancy–specific cardiac valvular[15-17] or RHD-specific guidelines.[4]

Reporting measures for studies of cardiac disease in pregnancy are currently in development[18] as part of the Core Outcomes in Women's and Newborn Health (CROWN) initiative,[19,20] but there is no known equivalent for RHD-P, which has specific risks related to its epidemiology.

Although clinical pathways can vary considerably according to the severity of RHD, principles of care that promote optimal maternal and baby outcomes include early diagnosis; preconception care including surgery and other interventions where required; early antenatal assessment including echocardiogram; access to specialized centers and treatment for high-risk women; and collaborative individualized care across disciplines and sectors.[4,14,21]

The purpose of this study was to systematically examine descriptions of care provision and associated outcomes for women with RHD-P in order to improve the understanding of how attributes of care are reported and how they align with guidelines.

2 | METHODS

Because of the lack of internationally accepted RHD-P measures, we reviewed relevant models of care and associated reporting measures referred to in clinical guidelines to conceptualize existing measures in a framework. We found no specific guidelines for RHD-P. Guidelines were chosen that addressed all cardiac diseases in pregnancy[14] and RHD with some reference to pregnancy.[4]

The scope was further broadened to include cardiovascular care standards in primary health settings for Australian Aboriginal and Torres Strait Islander peoples.[21] This guideline outlines elements of care across the continuum of risk and disease, with a focus on reducing disparity in access and outcomes, applicable for most populations where RHD is disproportionate.

Reporting measures relevant for women with RHD-P were identified and grouped in three categories to provide an analytic tool with which to interrogate the literature (Figure 1). These included the following: clinical information and reporting; risk in pregnancy; and RHD through the life-course. This framework served to guide the analysis of data gathered for the systematic review presented in this paper.

2.1 | Data sources and search protocol

A structured search of peer-reviewed research literature identified studies that described clinical care and measures for women with RHD-P. Data were extracted from the reported results of included studies and examined using a content analytic process,[22] directed by the framework of reporting measures (Figure 1).

The study was registered with the International Prospective Register of Systematic Reviews (PROSPERO #CRD42018059849).

Searches on PubMed, MEDLINE, EMBASE, CINAHL, Nursing and Allied Health Database, ATSIhealth, Indigenous Collection, Rural and Remote Health Database, ETG Complete, ISI Web of Science, Public Library of Science, and Trip Pro Databases were supported by hand-searching. The search strategy incorporated a combination of free term text items and Medical Subject Headings (MeSH): ("rheumatic heart" or "rheumatic fever" or "valvular heart disease") and ("pregnancy" or "pregnancy complications" or "pregnancy, high-risk" or "pregnancy complications, cardiovascular" "maternal") and ("models of care" or "guideline*" or "health service" or "maternal health services" or "primary health care" or "practice guideline" or "guideline adherence" or "health services accessibility" or "health care"). Inclusion criteria included all English-language peer-reviewed studies after 1994 in any setting or country with reference to RHD-P and attributes of care (Table 1).

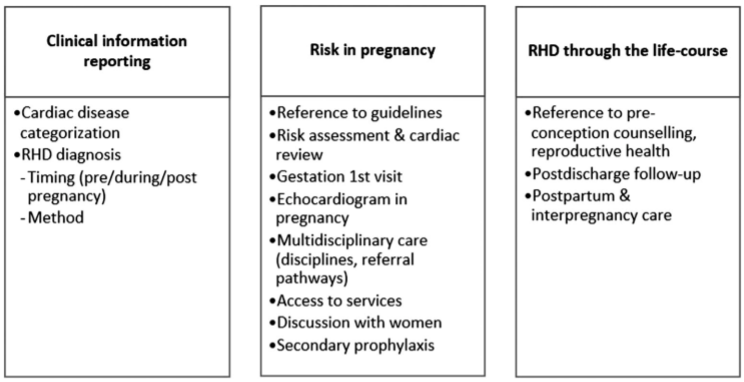


FIGURE 1 Framework of reporting measures for women with RHD-P

TABLE 1 Inclusion/exclusion criteria for content analysis of studies with reference to RHD-P

Included	Excluded
1995-2018	Pre-1995
English language	Non-English
Any setting in any country	None
Any study of women with cardiac disease with reference to RHD and pregnancy and attributes of care	Conference abstracts Opinion pieces/editorials Guidelines/reviews Systematic reviews Studies of biomedical treatments/interventions for women with RHD that do not refer to models of care in pregnancy

The PICOS (Population, Interventions, Comparators, Outcomes, and Study design)[23] framework guided the review question: *In studies that reference pregnant women with RHD, what core reporting measures are used to describe models of care?*

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines[24] informed the review. Screening used Endnote™ bibliographic and Covidence™ review tools. Critical appraisal referenced CASP and JBI checklists,[25,26] and the quality appraisal is summarized in Figure 4 as a four-tier grading. Differing judgments on inclusion were resolved by consensus, or, where no consensus was achieved, by a third reviewer. Reasons for excluding studies were clearly documented (Figure 2).

2.2 | Data extraction and content analysis

A data extraction tool was developed using Microsoft Excel™. Visual mapping used Tableau™ v2018.2.0 analytic software. Study characteristics included (Table 2 and Figure 3) country, World Bank income category, study design, setting/s and population, and documenting maternal mortality. Data were coded against the reporting framework and associated measures (Figure 1).

The study was exempt from ethics approval because the research was not conducted with humans or animals and used publicly available data.

3 | RESULTS

3.1 | General characteristics and quality appraisal

The most common types of study design were cohort (19) and case series (20), with two qualitative[27,28] studies, one cross-sectional[29] and one longitudinal screening study.[30] There was considerable heterogeneity in the methodologies, levels of evidence, and reporting measures of these predominantly retrospective studies. Individual study characteristics are outlined in Table 2. Reflecting the overall burden of RHD, the majority of the 43 studies from 18 countries were from India (8), South Africa (6), Pakistan (4), and Thailand (4), with one multicountry (predominantly Egypt) study (Figure 3). Most were published after 2004, paralleling a resurged clinical and research interest.[31] The distribution of studies by country and World Bank income category is detailed in Figure 3.

All studies were conducted in tertiary care settings with access to cardiac (or dedicated obstetric-cardiac) care, as well as primary health settings[27,28,30] and regional centers.[30,32] Maternal mortality ranged from 0% (16/42) to 37%.[5] Between 1% and 4% of women died in nearly half (20) of the studies. One study found significantly lower mortality rates in its index population (10%) compared with referred women (32%).[32]

Study designs affected the quality and were subject to high levels of bias, especially the case series. Referral and other selection biases as acknowledged in several papers[30,32-43]

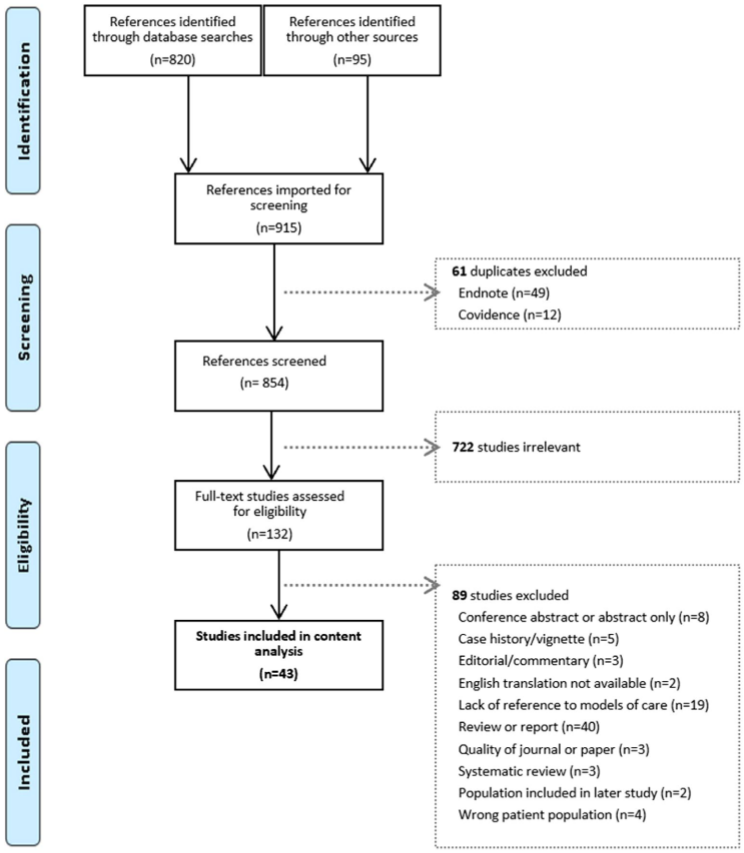


FIGURE 2 PRISMA diagram of studies with reference to RHD-P

were particularly related to the predominantly single-site tertiary-care-level settings.

Study periods ranged from 6 months to 21 years, with five of unspecified periods.[39,44-47] Long study periods (from 10 to 21 years in 16 studies) were noted to impact on protocols, which changed in response to therapeutic advances during that time.[33] Figure 4 provides the quality appraisal overview and maps studies against reporting measures. The studies were assessed as low (9), medium (12), medium-high (21), and high (1) quality, respectively. Key reporting measures from the framework (Figure 1) were poorly documented.

3.2 | Clinical information reporting

The percentage of the study population with RHD ranged from 100% (11 studies of women with RHD or mitral stenosis) to 3% in a high-income country,[11] with most comprising over 55% of the study population (Table 2). Six studies[29,46,48-51] from countries with an otherwise medium-to-high burden of RHD did not give a breakdown of underlying pathology of mitral stenosis or all valvular heart diseases (Table 2 and Figure 4). Mitral stenosis in women during their reproductive years is usually of rheumatic origin[14,52] and was used as a proxy for RHD where causation was unspecified.

Heart disease in low-income and middle-income countries is commonly diagnosed in pregnancy on development of severe symptoms.[5,13,32,44,53] However, 18 of the 41 quantitative studies did not specify timing of diagnosis. Others referred to late diagnosis in the context of poorer outcomes and health system shortcomings.[35,40,41,49,51,54-56] Diagnosis during pregnancy/postpartum ranged from 1%[57] to 97%[30] in a longitudinal screening study, with eight studies above 20% and four above 40%. In one high-income country, four women (of 95 pregnancies) were diagnosed with RHD after

TABLE 2 Characteristics of studies with reference to RHD-P

	Study	Setting/ type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
1	Abdel-Hady (2005)[57]	TCC-HRPC	n = 86 RHD = 90%	PCS	Egypt	Assess maternal/perinatal CDiP outcome.	1%*
2	Ahmed (2015)[48]	TCC-HRPC	n = 101 RHD = N/S MS = 100%	PCS	Pakistan	Evaluate MS feto-maternal outcomes and patient-specific management plan.	2%
3	Asghar (2005)[44]	TCC-HRPC	n = 50 RHD = 66%	PCS	Pakistan	Assess maternal/fetal outcome of CDiP.	0%*
4	Avila (2003)[33]	TCC-HRPC	n = 1000 RHD = 56%	RCS	Brazil	Experiences and outcomes of CDiP in referral center.	2%
5	Barbosa (2000)[34]	TCC-CDM	n = 45 RHD = 100%	RC	Brazil	Identify characteristics of complications of MS in pregnancy.	2%*
6	Beaton (2018)	PLS, CR	n = 58 RHD = 88%	PLS	Uganda	Determine the prevalence of maternal heart disease through active case finding and its attributable risk to adverse pregnancy outcomes.	2%*
7	Belton (2017)[28]	TCC-HRPC, CR	n = 8 RHD = 100%	Qualitative, Yarning	Australia	Study RHD-P health literacy and health services responses.	N/A (none)
8	Bhatla (2003)[56]	TCC-HRPC	n = 207 RHD = 88%	RC	India	Evaluate CDiP maternal/fetal outcome in developing country.	0%
9	Bhutta (2003)[65]	TCC-HRPC	n = 170 RHD = 91%	PCS	Pakistan	Determine CDiP outcomes postcardiac surgery.	0%
10	Chang (2018)[27]	TCC	n = 50 n = 25 RHD = 100%	Mixed methods	Uganda	Understand factors/attitudes toward reproductive health and disease in women with RHD.	N/A (none)
11	Chhetri (2014)[35]	TCC-HRPC	n = 53 RHD = 89%	PCS	Nepal	Investigate prevalence, characteristics, and outcomes of CDiP.	4%*
12	Chumpathong (2014)[36]	TCC-HRPC	n = 175 RHD = 66%	RC	Thailand	Evaluate CARPREG predicting cardiac/obstetric/neonatal complications.	3%
13	Curtis (2009)[11]	TCC-HRPC	n = 177 RHD = 3%	RCS	United Kingdom	Describe CDiP, review guideline adherence, and identify suboptimal management.	2%
14	Desai (2000)[49]	TCC-HRPC	n = 208 RHD = N/S MS = 100%	PCS	South Africa	Evaluate management/outcomes of MS in pregnancy.	0%
15	Diao (2011)[5]	TCC-HRPC	n = 50 RHD = 92%	RCS	Senegal	CDiP maternal/fetal outcomes in a low-income country.	37%*

(Continues)

TABLE 2 (Continued)

	Study	Setting/ type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
16	Faiz (2003)[50]	TCC-HRPC	n = 126 RHD = N/S MVHD = 95%	RCS	Saudi Arabia	Review MVHD during pregnancy: incidence, outcome.	0%
17	Fu (2015)[37]	TCC-CDM	n = 1086 RHD = 15%	RC	China	Identify heart failure risk during pregnancy women with preexisting disease	1%*
18	Jatavan (2011)[59]	TCC-HRPC	n = 125 RHD = 49%	RC	Thailand	Determine outcomes of CDiP.	0%
19	Kaluarachchi (1995)[66]	TCC-HRPC	n = 166 RHD = 70%	PCS	Sri Lanka	Evaluate CDiP pattern and outcome.	2%
20	Kanwar (2018)[67]	TCC-HRPC	n = 66 RHD = 77%	PC	India	Identify feto-maternal CDiP predictor complications/ outcomes ≤ 28v>28 weeks.	6%
21	Konar (2012)[45]	TCC-HRPC	n = 281 RHD = 69%	PCS	India	Evaluate CDiP and maternal/perinatal outcome.	1%
22	Kovavisarach (2007)[60]	TCC-HRPC	n = 196 RHD = 55% (period 3)	RC	Thailand	Assess prevalence, demographics, and maternal/perinatal outcomes of CDiP (3 study periods).	3%
23	Madazli (2010)[68]	TCC-HRPC	n = 144 RHD = 87%	RC	Turkey	Evaluate maternal/fetal outcome of CDiP in developing country.	0%
24	Malhotra (2004)[46]	TCC-HRPC	n = 312 RHD = N/S VHD = 100%	RC	India	Compare pregnancy outcomes of women with VHD to healthy women.	0.6%
25	Martins (2016)[54]	TCC-HRPC	n = 132 RHD = 62%	RC	Brazil	Determine CDiP risk factors associated with maternal/ neonatal complications.	3%
26	Michaelson-Cohen (2011)[38]	TCC-HRPC	n = 175 RHD = 41%	PC	Israel	Assess CDiP outcome.	0%
27	Nqayana (2008)[47]	TCC-CDM	n = 95 RHD = 81%	RCS	South Africa	Review CDiP in developing country.	0%
28	Pratibha (2014)[53]	TCC-HRPC	n = 200 RHD = 100%	RCS	India	Study pregnancy outcomes of RHD-P and evaluate perinatal outcomes of percutaneous balloon mitral valvuloplasty during pregnancy.	1%*
29	Puri (2013)[39]	TCC-HRPC	n = 97 RHD = 70%	RC	India	Assess CDiP and associated maternal/fetal complications.	3%

(Continues)

TABLE 2 (Continued)

	Study	Setting/ type of care	Study population	Study design	Country(s)	Objectives	Maternal mortality (rounded to nearest percent)
30	Rahman (2000)[61]	TCC-HRPC	n = 274 RHD = 76%	RCS	Saudi Arabia	Review CDiP outcomes.	0%
31	Rezk (2018)[69]	TCC-HRPC	n = 204 RHD = 100%	PC	Egypt	Assess cardiac/obstetric outcome of RHD-P and predictors of poor outcome.	0%*
32	Sartain (2012)[58]	TCC-HRPC	n = 95 RHD = 100%	RC	Australia	Determine maternal-cardiac complications/outcomes in patients with RHD.	0%*
33	Sawhney (2003)[55]	TCC-CDM	n = 500 RHD = 100%	RC	India	Study maternal/perinatal outcomes of RHD-P.	2%
34	Schoon (2001)[32]	TCC-HRPC	n = 42 index + 25 referred RHD = 33%	RCS	South Africa	Document CDiP mortality/morbidity and compare complicated vs uncomplicated.	18%
35	Schoon (1997)[51]	TCC/ regional-HRPC	n = 164 RHD = N/S	RCS	South Africa	Describe maternal outcome of CDiP.	10%
36	Silversides (2003)[72]	TCC-CDMs x2	n = 80 RHD = 100%	PC	Canada	Define predictors of maternal-cardiac complications in women with MS	0%*
37	Sliwa (2014)[40]	TCC-CDM	n = 225 RHD = 25%	PC	South Africa	Investigate spectrum of disease and maternal/fetal outcome in CDM.	4%
38	Soma-Pillay (2008)[41]	TCC-CDM	n = 189 RHD = 64%	RCS	South Africa	Assess CDiP profile and maternal/fetal outcome, and identify risk categories.	3%
39	Stangl (2008)[42]	TCC-HRPC	n = 93 RHD = 7.5%	RC	Germany	Analyze risks in low/high-risk women with CDiP.	0%*
40	Subbaiah (2013)[70]	TCC-HRPC	n = 100 RHD = 64%	RC	India	Analyze CDiP and maternal/fetal outcome.	0%*
41	Thanajiraprapa (2010)[71]	TCC-HRPC	n = 193 RHD = 69%	RCS	Thailand	Identify complications of CDiP.	1%
42	Van Hagen (2018)[43]	TCCs. Multiple sites/countries	n = 390 RHD = 100%	RC	Multiple countries	Assess maternal/fetal outcomes in women with MVHD.	1%*
43	Wasim (2008)[29]	TCC-HRPC	n = 160 RHD = N/S	Cross-sectional descriptive	Pakistan	Assess CDiP and feto-maternal outcomes.	4%

Abbreviations: CDiP, cardiac disease in pregnancy; CDM, dedicated cardiac/maternity clinic; CR, community setting and/or regional center; HRPC, high-risk multidisciplinary pregnancy clinic; MS, mitral stenosis; MVHD, mitral valvular heart disease; PC, prospective cohort; PCS, prospective case series; PLS, prospective longitudinal screening; RC, retrospective cohort; RCS, retrospective case series; TCC, tertiary care center; VHD, valvular heart disease.

Maternal mortality: *Asterisked percentages indicate RHD only.

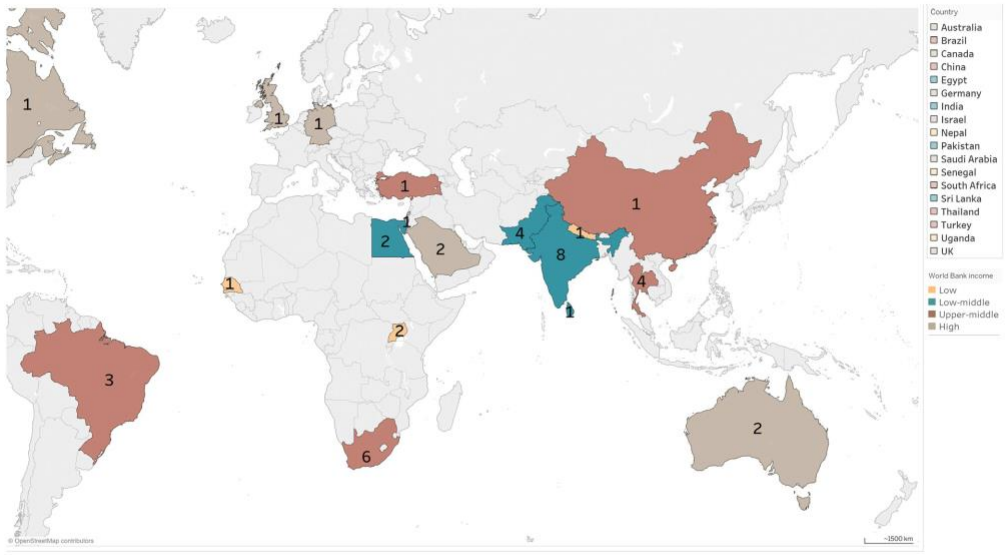


FIGURE 3 Number of studies referencing women with RHD-P: by country and World Bank income category

developing peripartum acute pulmonary edema.[58] One found 7% of women diagnosed postpartum,[39] but this was poorly documented overall.

Thirty-six studies specified echocardiographic review during pregnancy, although only four referenced diagnostic criteria.[30,43,48,58] Six studies did not specify RHD diagnosis confirmed by echocardiography, nor its use during pregnancy.[5,46,53,59-61]

3.3 | Models of care and risk in pregnancy

There was limited or no reference made to guidelines related to the care of pregnant women with cardiac disease in 16 studies.

The majority of the 36 quantitative studies that specified a cardiac risk score used the New York Heart Association (NYHA) classification (I-IV) of functional capacity.[62] Pregnancy-specific scores referenced CARdiac disease in PREGnancy (CARPREG)[63] cardiac events risk index, modified CARPREG,[58] and modified World Health Organization (mWHO) risk classifications.[14,64] A referral algorithm was developed for suspected and known cardiovascular disease in a low-resource setting.[40]

Poorer maternal and fetal outcomes were associated with higher risk scores (NYHA > II,[29,35,38,42,43,45-47,50,53-57,59,60,65-71] NYHA > I with mitral stenosis,[43] mWHO > 1,[40] CARPREG[36,54,67,72]/modified CARPREG[58] > 0, or study-specific factors such as mitral stenosis and anticoagulation therapy leading to increased maternal risks of heart failure, pulmonary hypertension, thromboembolic episodes, atrial fibrillation, and death[30,32,37,41,46,48,49,51]). The CARPREG index underestimated cardiac events in low-risk women but overestimated it in CARPREG > 0 in one study, possibly reflecting late diagnoses in pregnancy.[54] The quality of care and avoidable factors associated with near-miss morbidity was assessed in two papers,[32,41] whereas others described gaps between guideline recommendations and clinical implementation leading to compromised care.[11,43]

Late booking and/or infrequent antenatal care hampered early diagnosis and treatment[5,49,59] and was associated with poorer cardiac and perinatal outcomes,[32,40,41,49,51,67,69,70] yet the gestational age at first antenatal visit was reported in only 15 of 41 quantitative studies.

Medical management (such as beta-blockers, digoxin, and/or diuretics) and percutaneous balloon mitral valvuloplasty[73] (hereafter valvuloplasty) in refractory cases of mitral stenosis generally improved outcomes[33,34,56] where reported. However, studies emphasized the challenges of providing optimal care in resource-challenged environments, including appropriate clinician skills, access to medication, valvuloplasty and surgery, health system shortcomings, and sociocultural factors,[27,30,32,34,35,40,41,43,44,46,49,51,54-56,59] with one (where 37% of women with rheumatic valvular disease died) noting that valvuloplasty facilities were simply unavailable at their tertiary center.[5] The multicountry study found

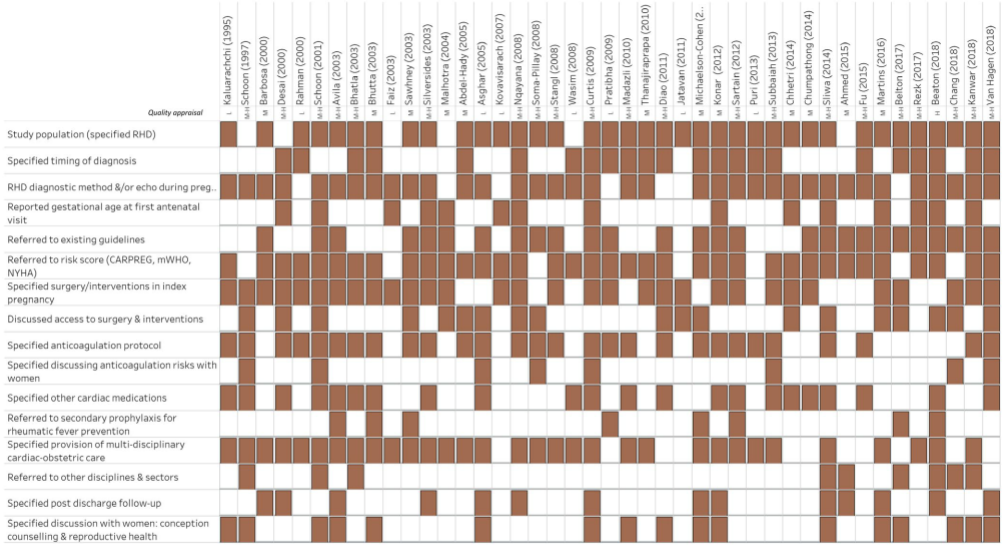


FIGURE 4 Studies with reference to RHD-P: core reporting measures and quality appraisal by study. Legend: Visual representation of specified reporting measures (outlined in Figure 1 framework) according to study. Shaded square indicates the reporting measure was specified in the study; blank square indicates it was not. Quality appraisal assessed as L (low), M (medium), M-H (medium-high), or H (high)

a greater number of valvular interventions in high-income countries despite more women in low-income settings having severe mitral stenosis.[43]

Despite the potentially catastrophic maternal-fetal risks associated with the use of anticoagulation in pregnancy, the regimen was detailed in only half of the 28 quantitative studies that specified the number of women on therapy. Nine referred to discussion with women in the context of complex decisions surrounding choice of regimen which balanced the maternal risk of thromboembolism using low molecular weight heparin against the increased fetal risk of warfarin use. Studies discussed late booking affecting the anticoagulation regimen[47,51,66,70] with lack of adherence to protocol (or access to treatment) detailed as an important risk factor for morbidity and mortality.[41,47,51,53,57] Warfarin embryopathy/fetopathy was likely underestimated in studies where postmortems or detailed examinations were not performed.[41,65]

Secondary prophylaxis (usually 3-4 weekly bicillin injections), where indicated, prevents rheumatic fever recurrence and is safe during pregnancy. Its use was referred to in only seven quantitative studies.

Most studies emphasized the need for multidisciplinary care in discussion and/or recommendations although somewhat fewer (32) specified its provision in their study. This was highlighted in one study that found obstetric-cardiac individualized review determined according to the risk-promoted optimal outcomes despite its low-resource setting.[40] Others similarly pointed to early multidisciplinary evaluation and management contributing to few or no maternal deaths in otherwise high-risk women.[38,47,49,50,56,58,61,65,68,70,71]

Vaginal birth is recommended for women with valvular heart disease unless contraindicated by severe cardiac morbidity[14] or obstetric complications. Cesarean rates varied enormously from <10%[44,46,50,61] to 75%,[69] with several studies above 40%[35,41-43,47,57,68] and higher again in groups stratified by risk or poorer outcomes.[42,43,57,68,69]

There was limited reference to care outside the index pregnancy period. Those that did noted the continued heightened risk of morbidity and mortality,[40,43] and another detailed an increased need for cardiac intervention in the first year after delivery.[11]

Papers that called for improved clinician training in primary health settings to support cardiac disease detection/referral[48,51,67] were mostly (5/6) published since 2014. Gaps in awareness among primary health care nurses (and women) were associated with delayed referrals[40] and consistent with other studies that found women received contradictory advice and limited education.[27,28] Language-appropriate health education that promoted a shared understanding was largely absent for Aboriginal women with RHD.[28]

3.4 | RHD through the life-course

Twenty-six studies did not specify the provision of conception counseling and reproductive health in their setting, with one listing it under management standards not followed.[11] Women can perceive risk to be over with the end of pregnancy,[65] underscoring the significance of postnatal counseling.

Emerging themes in a qualitative study of women's experiences with RHD included misconceptions about side effects of contraceptives; lack of agency in reproductive decision-making; and stigma related to financial and perceived reproductive limitations.[27]

4 | DISCUSSION

The aim of this review was to synthesize the literature and map reported measures against a framework drawn from guidelines related to models of care for RHD-P. Our study found gaps in the three framework categories of clinical reporting, risk in pregnancy, and RHD through the life-course.

A recent overview of RHD strategies emphasizes the imperative for accurate, current data to inform policy and measure trends.[1] Poor reporting of measures related to cardiac pathology and diagnoses precludes a true assessment of the burden of RHD-P and changing epidemiology. In turn, this is limited by the capacity of health services to diagnose cases. Women with subclinical or milder forms of disease or fatal events before admission are likely to be missed in low-income settings.[40] The community-based screening study found <4% of women with RHD were aware of their diagnosis of prepregnancy.[30] There are no known studies of the impact of RHD-P from countries that have among the highest reported rates of RHD in the world,[74] including the Pacifica[75,76] and Oceanic regions.[77] A population-based study conducted in the high-income countries of Australia and New Zealand (currently under review) shows similarly high rates among Māori and Pasifika women.[8,78]

Reporting gaps are consistent with a South African systematic review of antenatal heart disease, which recommended minimum criteria including diagnosis, reference population, cardiac profile, and outcomes.[13]

Although the lack of diagnostic reference to echocardiography is partly explained by study periods, resource limitations of facilities and expertise no doubt also have an impact on this. However, increasingly portable technologies and successful screening programs strengthen the argument for earlier review in primary care settings.[30,79,80] Standardization has improved with the 2012 echocardiographic diagnostic criteria.[81]

The high risk of anticoagulation in pregnancy requires better reporting in any study of women with RHD. A recent meta-analysis of anticoagulation in women with mechanical heart valves found fetal risk was similar between women taking <5 mg warfarin daily to those on low molecular weight heparin.[82] These findings underscore the need for granularity of reporting prescribed regimens—including level of adherence and women's access to treatment.

Increasing calls to improve the scale-up of and access to surgery/interventions in low-income countries[83,84] reflect service deficiencies highlighted in studies.

There were few studies that followed RHD-P care trajectories and outcomes outside tertiary centers. A small but growing number of initiatives such as the landmark RHD screening study[30] harness specialist resources in community settings to improve early diagnosis of RHD and care for women. These are embedded in collaborative cross-sectoral approaches,[80] drawing on successful strategies developed in other chronic disease models and supported by strengthened health systems.[30,85] They potentially obviate the need for emergency-driven, costly tertiary care[32]—and in turn support improved outcomes for this disease which is preventable at many levels.[4] Such principles can equally apply to vulnerable populations in high-income countries.[86] However, executing these models in practice is often tested by the plethora of structural, political, and economic barriers to implementation that are part of the RHD landscape.

The overall lack of reference to postdischarge care (including recommended follow-up) suggests likely underreporting of complications. Three of four RHD-related maternal deaths reported in the multicountry study were up to 6 months' postpartum,[43] consistent with (often-avoidable) factors and risks reported in other studies of late maternal death.[87-89]

Existing literature on preconception and reproductive health care is predominantly focused on congenital heart disease. There is a growing body of evidence of the role of preconception care in optimizing general health and risk awareness in marginalized communities,[90] highly relevant for women with RHD.[3]

There are no RHD-P–specific guidelines. Selected reporting measures were drawn from cardiac disease in pregnancy guidelines that referenced RHD,[14] the Australia-specific RHD guidelines[4,91] (an updated edition of which includes a substantially enhanced section on women and RHD[91]), and cardiovascular standards for Aboriginal and Torres Strait Islander peoples.[21] Guidelines are themselves mostly based on case series and observational studies. However, we believe the included reporting measures reflect fundamental principles of care for vulnerable populations where RHD is prevalent, particularly in relation to maternal health. RHD-specific research to test the evidence is required to strengthen the rigor of recommendations, better understand the effects of pregnancy, and choose the best individualized plan for ongoing care.

We propose the reviewed framework of measures (Figure 1) addressing the categories of clinical information reporting;

models of care and risk in pregnancy; and RHD through the life-course as a core outcome set for women with RHD-P, adapted to local cultural, social, and economic contexts.[92] A Delphi method review[93,94] to evaluate an extended set with neonatal outcomes (with global stakeholders including health services and women in high-prevalence settings) will further strengthen recommendations for adoption.

4.1 | Strengths and limitations

This review was constrained by the heterogeneity and design of included studies, with most subject to substantial bias (particularly referral) and reporting inconsistencies. Study sites were predominantly tertiary centers, providing care particularly for those with severe RHD who were able to access specialist care. However, these observational studies provide the best available current evidence and insight in determining models of care associated with optimal maternal outcomes.

What was reported (or not) may not reflect actual practice. In the absence of specific reference to a care attribute, it was assumed that it was not addressed, which may or may not be true. This may be particularly relevant for aspects such as conception counseling.[11]

4.2 | Conclusions

Rheumatic heart disease has been described as providing a model for strengthening health systems to address other cardiovascular diseases in limited-resource countries. This framework is especially pertinent for women with RHD, where best-practice models of care in a strengthened maternal health system are often congruent with those that support women with RHD.

This qualitative synthesis highlights gaps of what is reported in the literature, with consequent underestimation of burden and weakened ability to action strategies based on findings. We propose a Delphi testing of the reporting framework detailed in this paper and adoption of a core outcome set to support data consistency and comparability of studies, strengthen knowledge and awareness of burden (clinical and social), and improve benchmarking of care for women with RHD.

ORCID

Geraldine Vaughan https://orcid.org/0000-0002-0132-9946
Angela Dawson https://orcid.org/0000-0003-0926-2202
Michael J. Peek https://orcid.org/0000-0001-6055-5129
Jonathan R. Carapetis https://orcid.org/0000-0002-1182-9792
Elizabeth A. Sullivan https://orcid.org/0000-0002-8718-2753

REFERENCES

1. Abouzeid M, Wyber R, La Vincente S, et al. Time to tackle rheumatic heart disease: data needed to drive global policy dialogues. *Glob Public Health*. 2018;14:1-13.
2. Zühlke LJ, Steer AC. Estimates of the global burden of rheumatic heart disease. *Glob Heart*. 2013;8(3):189-195.
3. Zühlke L, Karthikeyan G, Engel ME, et al. Clinical outcomes in 3343 children and adults with rheumatic heart disease from 14 low- and middle-income countries: two-year follow-up of the global Rheumatic Heart Disease Registry (the REMEDY Study). *Circulation*. 2016;134(19):1456-1466.
4. RHDAustralia (ARF/RHD writing group), National Heart Foundation of Australia, Cardiac Society of Australia and New Zealand. *Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease*, 2nd edn. 2012. 978-0-9587722-9-7 (paperback); 978-0-9587722-5-9 (online).
5. Diao M, Kane A, Ndiaye MB, et al. Pregnancy in women with heart disease in sub-Saharan Africa. *Arch Cardiovasc Dis*. 2011;104(6–7):370-374.
6. AMHMAC. Aboriginal and Torres Strait Islander Health Performance Framework 2017 Report. Canberra 2017.
7. Vaughan G, Tune K, Peek MJ, et al. Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia. *Int Health*. 2018;10(6):480-489.
8. Farquhar C, McLintock C, Mahony F, et al. Rheumatic heart disease in pregnancy: a prospective New Zealand cohort: ep13. 44. *BJOG*. 2015;122:271.
9. Bocking N, Matsumoto C-L, Loewen K, et al. High Incidence of invasive Group A streptococcal infections in remote Indigenous communities in Northwestern Ontario, Canada. *Open Forum Infect Dis*. 2016;4(1):ofw243.
10. Zühlke LJ, Beaton A, Engel ME, et al. Group A streptococcus, acute rheumatic fever and rheumatic heart disease: epidemiology and clinical considerations. *Curr Treat Options Cardiovasc Med*. 2017;19(2):15.
11. Curtis SL, Marsden-Williams J, Sullivan C, et al. Current trends in the management of heart disease in pregnancy. *Int J Cardiol*. 2009;133(1):62-69.
12. Van Hanegem N, Miltenburg AS, Zwart JJ, Bloemenkamp K, Van Roosmalen J. Severe acute maternal morbidity in asylum seekers: a two-year nationwide cohort study in the Netherlands. *Acta Obstet Gynecol Scand*. 2011;90(9):1010-1016.
13. Watkins D, Sebitloane M, Engel M, Mayosi B. The burden of antenatal heart disease in South Africa: a systematic review. *BMC Cardiovasc Disor*. 2012;12(1):23.
14. Regitz-Zagrosek V, Roos-Hesselink J, Bauersachs J, et al. 2018 ESC Guidelines for the management of cardiovascular diseases during pregnancy. *Eur Heart J*. 2018;39(34):3165-3241.
15. Baumgartner H, Falk V, Bax JJ, et al. 2017 ESC/EACTS Guidelines for the management of valvular heart disease. *Eur Heart J*. 2017;38(36):2739-2791.
16. Nishimura R, Otto C, Bonow RO, et al. 2017 AHA/ACC Focused Update of the 2014 AHA/ACC Guideline for the Management of Patients With Valvular Heart Disease: A Report of the American College of Cardiology/American Heart Association Task Force on Clinical Practice Guidelines. 2017(1524–4539 (Electronic)).
17. Nishimura RA, Otto CM, Bonow RO, et al. 2014 AHA/ACC guideline for the management of patients with valvular heart

disease: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines. *J Thoracic Cardiovasc Surg*. 2014;148(1):e1-e132.
18. D'Souza R, Silversides C, Sermer M, et al. COSCarP study (Core outcome sets for studies on cardiac disease in pregnancy). 2018. http://www.comet-initiative.org/studies/details/834
19. Duffy J, Rolph R, Gale C, et al. Core outcome sets in women's and newborn health: a systematic review. *BJOG*. 2017;124(10):1481-1489.
20. Geirsson RT, Khan K. The CROWN Initiative: journal editors invite researchers to develop core outcomes in women's health. *Acta Obstet Gynecol Scand*. 2014;93(9):845-847.
21. Brown A, O'Shea R, Mott K, McBride K, Lawson T, Jennings G. Essential service standards for equitable national cardiovascular care for aboriginal and torres strait islander people. *Heart Lung Circ*. 2015;24:126-141.
22. Hsieh H-F, Shannon S. Three approaches to qualitative content analysis. *Qual Health Res*. 2005;15(9):1277-1288.
23. Schardt C, Adams MB, Owens T, Keitz S, Fontelo P. Utilization of the PICO framework to improve searching PubMed for clinical questions. *BMC Med Inform Decis Mak*. 2007;7(1):16.
24. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med*. 2009;6(7):e1000097.
25. Critical Appraisal Skills Programme (CASP). CASP Appraisal Checklists. 2014; https://casp-uk.net/casp-tools-checklists/
26. Johanna Briggs Institute. Critical Appraisal Checklists. 2017; http://joannabriggs.org/research/critical-appraisal-tools.html
27. Chang AY, Nabbaale J, Nalubwama H, et al. Motivations of women in Uganda living with rheumatic heart disease: a mixed methods study of experiences in stigma, childbearing, anticoagulation, and contraception. *PLoS ONE*. 2018;13(3):e0194030.
28. Belton S, Kruske S, Jackson Pulver L, et al. Rheumatic heart disease in pregnancy: how can health services adapt to the needs of Indigenous women? A qualitative study. *Aust N Z J Obstet Gynaecol*. 2018;58(4):425-431.
29. Wasim T, Amer W, Majrroh A, Siddiq S. Foetomaternal outcome of pregnancy with cardiac disease. *J Pak Med Assn*. 2008;58(4):175-178.
30. Beaton A, Okello E, Scheel A, et al. Impact of heart disease on maternal, fetal and neonatal outcomes in a low-resource setting. *Heart*. 2019;105(10):755-760.
31. Carapetis J. The stark reality of rheumatic heart disease. *Eur Heart J*. 2015;36(18):1070-1073.
32. Schoon M. Severe morbidity and mortality associated with cardiac disease during pregnancy in the Free State Public Health Service. PhD, UoFS. 2001.
33. Avila WS, Rossi EG, Ramires J, et al. Pregnancy in patients with heart disease: experience with 1,000 cases. *Clin Cardiol*. 2003;26(3):135-142.
34. Barbosa P, Lopes AA, Feitosa GS, et al. Prognostic factors of rheumatic mitral stenosis during pregnancy and puerperium. *Arq Bras Cardiol*. 2000;75(3):215-224.
35. Chhetri S, Shrestha N, Pilgrim T. Pregnancy complicated by heart disease in Nepal. *Heart Asia*. 2014;6(1):26-29.
36. Chumpathong S, Sirithaweesit C, Pechpaisit N, et al. Predictors for complications in pregnant women with heart disease, a retrospective study. *J Med Assoc Thai*. 2014;97(7):730-735.
37. Fu Q, Lin J. Risk factors for heart failure during pregnancy among Chinese women with cardiac disease. *Int J Gynaecol Obstet*. 2015;130(3):266-269.
38. Michaelson-Cohen R, Elstein D, Ioscovich A, et al. Severe heart disease complicating pregnancy does not preclude a favourable pregnancy outcome: 15 years' experience in a single centre. *J Obstet Gynaecol*. 2011;31(7):597-602.
39. Puri S, Bharti A, Mohan B, Bindal V, Verma S. Maternal heart disease and pregnancy outcomes. *JK Sci*. 2013;15(1):7-10.
40. Sliwa K, Libhaber E, Elliott C, et al. Spectrum of cardiac disease in maternity in a low-resource cohort in South Africa. *Heart*. 2014;100(24):1967-1974.
41. Soma-Pillay P, MacDonald A, Mathivha T, Bakker J, Mackintosh M. Cardiac disease in pregnancy: a 4-year audit at Pretoria Academic Hospital. *S Afr Med J*. 2008;98(7):553-556.
42. Stangl V, Schad J, Gossing G, Borges A, Baumann G, Stangl K. Maternal heart disease and pregnancy outcome: a single-centre experience. *Eur J Heart Failure*. 2008;10(9):855-860.
43. van Hagen IM, Thorne SA, Taha N, et al. Pregnancy outcomes in women with rheumatic mitral valve disease. *Circulation*. 2018;137(8):806-816.
44. Asghar F, Kokab H. Evaluation and outcome of pregnancy complicated by heart disease. *J Pak Med Assn*. 2005;55(10):416-419.
45. Konar H, Chudhuri S. Pregnancy complicated by maternal heart disease: a review of 281 women. *J Obstet Gynecol India*. 2012;62(3):301-306.
46. Malhotra M, Sharma J, Tripathii R, Arora P, Arora R. Maternal and fetal outcome in valvular heart disease. *Int J Gynecol Obstet*. 2004;84:11-16.
47. Nqayana T, Moodley J, Naidoo D. Cardiac disease in pregnancy. *Cardiovasc J Afr*. 2008;19(3):145-151.
48. Ahmed N, Kausar H, Ali RL. Fetomaternal outcome of pregnancy with mitral stenosis. *Pak J Med Sci*. 2015;31(3):643-647.
49. Desai D, Adanlawo M, Naidoo D, Moodley J, Kleinschmidt I. Mitral stenosis in pregnancy: a four-year experience at King Edward VIII Hospital, Durban, South Africa. *BJOG*. 2000;107(8):953-958.
50. Faiz S, Al-Meshari A, Sporrong B. Pregnancy and valvular heart disease. *Saudi Med J*. 2003;24(10):1098-1101.
51. Schoon M, Bam R, Wolmarans L. Cardiac disease during pregnancy – a free state perspective on maternal morbidity and mortality. *Cardiovasc J Afr*. 1997;87(1):C19-C22.
52. Iung B. Pregnancy-related cardiac complications: A consequence of the burden of rheumatic heart disease in sub-Saharan Africa. *Arch Cardiovasc Dis*. 2011;104(6):367-369.
53. Pratibha D, Kiranmai D, Vemuri U, Rani V, Vani N. Pregnancy outcome in chronic rheumatic heart disease. *J Obstet Gynecol India*. 2009;59(1):41-46.
54. Martins L, Freire C, Capurucu C, Nunes M, Rezende C. Risk prediction of cardiovascular complications in pregnant women with heart disease. *Arq Bras Cardiol*. 2016;106(4):289-296.
55. Sawhney H, Aggarwal N, Suri V, Vasishta K, Sharma Y, Grover A. Maternal and perinatal outcome in rheumatic heart disease. *Int J Gynecol Obstet*. 2003;80(1):9-14.
56. Bhatla N, Lal S, Behera G, et al. Cardiac disease in pregnancy. *Int J Gynaecol Obstet*. 2003;82(2):153-159.
57. Abdel-Hady E, El-Shamy M, El-Rifai A, Goda H, Abdel-Samad A, Moussa S. Maternal and perinatal outcome of

pregnancies complicated by cardiac disease. *Int J Gynaecol Obstet*. 2005;90(1):21-25.
58. Sartain J, Anderson N, Barry J, Boyd P, Howat P. Rheumatic heart disease in pregnancy: cardiac and obstetric outcomes. *Intern Med J*. 2012;42(9):978-984.
59. Jatavan T, Luewan S, Tongsong T. Outcomes of pregnancy complicated by heart disease at Maharaj Nakorn Chiang Mai Hospital. *J Med Assoc Thai*. 2011;94(10):1159-1163.
60. Kovavisarach E, Nualplot P. Outcome of pregnancy among parturients complicated with heart disease in Rajavithi Hospital. *J Med Assoc Thai*. 2007;90(11):2253-2259.
61. Rahman J, Rahman M, Al-Suleiman S, Al-Jama F. Pregnancy complicated by maternal cardiac disease: a review of 274 patients. *J Obstet Gynaecol*. 2000;20(3):242-245.
62. The Criteria Committee of the New York Heart Association (NYHA). 1994 Revisions to classification of functional capacity and objective assessment of patients with diseases of the heart: NYHA functional class. In: Dolgin M, ed. *Nomenclature and Criteria for Diagnosis of Diseases of the Heart and Great Vessels*, 9th edn. Boston, MA: Little, Brown & Co; 1994:253-256.
63. Siu S, Sermer M, Colman J, et al. Prospective multicenter study of pregnancy outcomes in women with heart disease. *Circulation*. 2001;104:515-521.
64. Regitz-Zagrosek V, Blomstrom Lundqvist C, Borghi C, et al. ESC Guidelines on the management of cardiovascular diseases during pregnancy. *Eur Heart J*. 2011;32:3147-3197.
65. Bhutta S, Aziz S, Korejo R. Pregnancy following cardiac surgery. *J Pak Med Assn*. 2003;53(9):407-413.
66. Kaluarachchi A, Seneviratne H. Heart disease in pregnancy – evaluation of disease pattern and outcome in Sri Lanka. *J Obstet Gynaecol*. 1995;15(1):9-14.
67. Kanwar R, Sharma M, Marwah S, Mittal P, Kumari N. Heart disease in pregnancy-evaluation of spectrum, association of predictors with obstetric outcome and need for comprehensive medical care. *J Clin Diagn Res*. 2018;12(1):QC20-QC24.
68. Madazli R, Sal V, Cift T, Guralp O, Goymen A. Pregnancy outcomes in women with heart disease. *Arch Gynecol Obstet*. 2009;281(1):29-34.
69. Rezk M, Elkilani O, Shaheen A, Gamal A, Badr H. Maternal hemodynamic changes and predictors of poor obstetric outcome in women with rheumatic heart disease: a five-year observational study. *J Matern-Fetal Neonat Med*. 2018;31(12):1542-1547.
70. Subbaiah M, Sharma V, Kumar S, et al. Heart disease in pregnancy: cardiac and obstetric outcomes. *Arch Gynecol Obstet*. 2013;288(1):23-27.
71. Thanajiraprapa T, Phupong V. Pregnancy complications in women with heart disease. *J Matern-Fetal Neonat Med*. 2010;23(10):1200-1204.
72. Silversides C, Colman J, Sermer M, Siu S. Cardiac risk in pregnant women with rheumatic mitral stenosis. *Am J Cardiol*. 2003;91(11):1382-1385.
73. Nobuyoshi M, Arita T, Shirai S-I, et al. Percutaneous balloon mitral valvuloplasty: a review. *Circulation*. 2009;119(8):e211-e219.
74. Watkins DA, Johnson CO, Colquhoun SM, et al. Global, regional, and national burden of rheumatic heart disease, 1990-2015. *New Engl J Med*. 2017;377(8):713-722.
75. Mirabel M, Tafflet M, Noël B, et al. Newly diagnosed rheumatic heart disease among indigenous populations in the Pacific. *Heart*. 2015;101(23):1901-1906.
76. Colquhoun S. Global epidemiology, prevention and control of rheumatic heart disease with a focus on the Pacific Islands region Darwin, Australia: Global Health Division, Menzies School of Health Research, Charles Darwin University; 2015.
77. Davis K, Remenyi B, Draper AD, et al. Rheumatic heart disease in Timor-Leste school students: an echocardiography-based prevalence study. *Med J Australia*. 2018;208:303-307.
78. Sullivan E, Vaughan G, Jackson Pulver L. Perinatal outcomes of women with rheumatic heart disease. *J Paediatr Child Health*. 2016;52(S2):1-131.
79. Saxena A. Rheumatic heart disease screening by "point-of-care" echocardiography: an acceptable alternative in resource limited settings? *Transl Pediatr*. 2015;4(3):210-213.
80. Beaton A, Okello E, Destigter K, Scheel A, Perlman L, Sable C. Impact of rheumatic heart disease on maternal outcomes in pregnancy: Leveraging existing infrastructure to address a critical knowledge gap. *Glob Heart*. 2016;11(2 SUPPL. 1):e75.
81. Reményi BO, Wilson N, Steer A, et al. World Heart Federation criteria for echocardiographic diagnosis of rheumatic heart disease—an evidence-based guideline. *Nat Rev Cardiol*. 2012;9:297-309.
82. Steinberg ZL, Dominguez-Islas CP, Otto CM, Stout KK, Krieger EV. Maternal and fetal outcomes of anticoagulation in pregnant women with mechanical heart valves. *J Am Coll Cardiol*. 2017;69(22):2681-2691.
83. Forcillo J, Watkins DA, Brooks A, et al. Making cardiac surgery feasible in African countries: experience from Namibia, Uganda, and Zambia. *J Thoracic Cardiovasc Surg*. 2019, in press. https://doi.org/10.1016/j.jtcvs.2019.01.054
84. Zilla P, Bolman RM, Yacoub MH. The Cape Town declaration on access to cardiac surgery in the developing world. *S Afr Med J*. 2018;108:702-704.
85. Fauveau V, Sherratt D, de Bernis L. Human resources for maternal health: multi-purpose or specialists? *Hum Resour Health*. 2008;6(1):21.
86. Lee I, Purbrick B, Barzi F, et al. Cohort profile: the Pregnancy and Neonatal Diabetes Outcomes in Remote Australia (PANDORA) study. *Int J Epidemiol*. 2018;47:1045-1046.
87. Knight M, Nair M, Tuffnell D, et al. Saving lives, improving mothers' care—surveillance of maternal deaths in the UK 2012-14 and lessons learned to inform maternity care from the UK and Ireland Confidential Enquiries into Maternal Deaths and Morbidity 2009-14. National Perinatal Epidemiology Unit, University of Oxford;2016.
88. Hameed A, Morton C, Moore A. Improving health care response to cardiovascular disease in pregnancy and postpartum. Developed under contract #11-10006 with the California Department of Public Health, Maternal, Child and Adolescent Health Division. 2017.
89. Sliwa K, Azibani F, Baard J, et al. Reducing late maternal death due to cardiovascular disease—a pragmatic pilot study. *Int J Cardiol*. 2018;272:70-76.
90. Zühlke L, Acquah L. Pre-conception counselling for key cardiovascular conditions in Africa: optimising pregnancy outcomes. *Cardiovasc J Afr*. 2016;27(2):79-83.
91. RHDAustralia. Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease (3rd edition) (Under development). 2019.

92. Kennedy HP, Cheyney M, Dahlen HG, et al. Asking different questions: a call to action for research to improve the quality of care for every woman, every child. *Birth*. 2018;45(3):222-231.
93. Powell C. The Delphi technique: myths and realities. *J Adv Nurs*. 2003;41(4):376-382.
94. Boulkedid R, Abdoul H, Loustau M, Sibony O, Alberti C. Using and reporting the Delphi method for selecting healthcare quality indicators: a systematic review. *PLoS ONE*. 2011;6(6):e20476.

How to cite this article: Vaughan G, Dawson A, Peek MJ, Carapetis JR, Sullivan EA. Standardizing clinical care measures of rheumatic heart disease in pregnancy: A qualitative synthesis. *Birth*. 2019;00:1–14. https://doi.org/10.1111/birt.12435

Int Health 2018; **10**: 480–489
doi:10.1093/inthealth/ihy048 Advance Access publication 25 July 2018

ORIGINAL ARTICLE

Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia

Geraldine Vaughan[a,*], Kylie Tune[b,c], Michael J. Peek[d,e], Lisa Jackson Pulver[f], Bo Remenyi[b,c], Suzanne Belton[b,g] and Elizabeth A. Sullivan[a]

[a]University of Technology Sydney, Faculty of Health, Australian Centre for Public and Population Health Research (ACPPHR), Sydney, Australia; [b]Menzies School of Health Research, Darwin, Australia; [c]The Royal Darwin Hospital, NT Cardiac, Darwin, Australia; [d]The Australian National University and Centenary Hospital for Women and Children, Canberra, Australia; [e]The Canberra Hospital, Canberra, Australia; [f]Western Sydney University, Parramatta, Australia; [g]Primary Health Network, Darwin, Australia

*Corresponding author. Tel: +61 402 905 220; E-mail: Geraldine.vaughan@uts.edu.au

Received 3 January 2018; revised 29 May 2018; editorial decision 1 June 2018; accepted 2 June 2018

Background: The global burden of rheumatic heart disease (RHD) is two-to-four times higher in women, with a heightened risk in pregnancy. In Australia, RHD is found predominantly among Aboriginal and Torres Strait Islander peoples.

Methods: This paper reviews processes developed to identify pregnant Australian women with RHD during a 2-year population-based study using the Australasian Maternity Outcomes Surveillance System (AMOSS). It evaluates strategies developed to enhance reporting and discusses implications for patient care and public health.

Results: AMOSS maternity coordinators across 262 Australian sites reported cases. An extended network across cardiac, Aboriginal and primary healthcare strengthened surveillance and awareness. The network notified 495 potential cases, of which 192 were confirmed. Seventy-eight per cent were Aboriginal and/or Torres Strait Islander women, with a prevalence of 22 per 1000 in the Northern Territory.

Discussion: Effective surveillance was challenged by a lack of diagnostic certainty, incompatible health information systems and varying clinical awareness among health professionals. Optimal outcomes for pregnant women with RHD demand timely diagnosis and access to collaborative care.

Conclusion: The strategies employed by this study highlight gaps in reporting processes and the opportunity pregnancy provides for diagnosis and re/engagement with health services to support better continuity of care and promote improved outcomes.

Keywords: Health information systems, Health services, Indigenous health, Pregnancy, Public health, Rheumatic heart disease

Introduction

Rheumatic heart disease (RHD) is a condition of paradox in the high-income countries of Australia and New Zealand (ANZ), rare overall, but common in disadvantaged populations, with the burden of RHD among Indigenous and Pacifica peoples in ANZ among the highest documented rates in the world.[1–3]

This non-communicable disease of inequity is a serious sequela of (usually repeated) episodes of the Group A streptococcus infection of rheumatic fever (RF), resulting in chronic damage to heart valves. Two-to-four times as many women as men are diagnosed with RHD.[2,4,5] Together with other high-income countries, the overall incidence of RHD in Australia dropped dramatically in the second half the twentieth century (Figure 1). However, for Aboriginal and/or Torres Strait Islander women, RHD is among the leading 20 causes of fatal burden (years of life lost) and among the 20 specific diseases contributing to the gap in total burden (disability-adjusted life years (DALYs)), with a rate ratio of 6.9 Indigenous compared with non-Indigenous Australian women.[2]

An increased cardiac workload in pregnancy can unmask undiagnosed RHD and exacerbate clinical symptoms in women with known disease. The risk of poorer maternal and perinatal outcomes escalates, particularly for women requiring anticoagulant

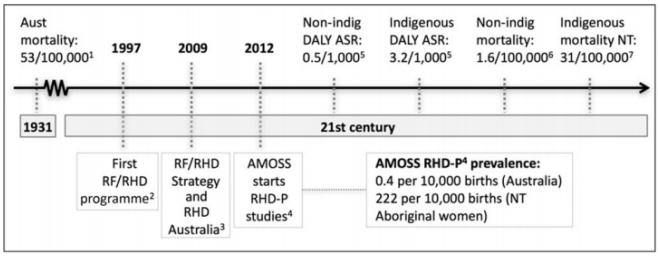


Figure 1. RHD prevalence and surveillance timeline in Australia. [1]'All-female Australia age-standardized RHD death rate' (Mortality and Morbidity: Cardiovascular disease: 20th century trends, 2002). Section 127 of the Australian Constitution excluded Aboriginals from official statistics until 1967. The first regular collection of Aboriginal health data began in 1957 (NT). Jurisdictional legislation did not allow for registration of vital statistics by race until after 1979.[32] [2]First Rheumatic Fever (RF)/RHD control programme established Northern Territory (NT) jurisdiction of Australia.[33] [3]Register and control programmes in three jurisdictions (five of eight jurisdictions by 2016); National coordination to develop resources and data collection system through RHDAustralia.[34] [4]2012–2016: Rheumatic heart disease in pregnancy. National Health and Medical Research Council (NHMRC) project grant #1024206. An AMOSS study. [5]Australian Institute of Health and Welfare (AIHW). Australian Burden of Disease Study: Impact and causes of illness and death in Aboriginal and Torres Strait Islander people 2011. Canberra, Australia: AIHW, 2016. BOD 7. [6]Table 1.1 'Underlying cause of death, All causes, Australia' (All-female age-standardized).[35] [7]Table 12.5 'Underlying causes of death, leading causes by Aboriginal and Torres Strait Islander status, NSW, Qld, SA, WA and NT, 2010–2014' (All-female age-standardized). In: 3303.0 Causes of Death, Australia, 2014. ABS 2016. **Notes**: DALY, ASR, disability adjusted life years age-standardized rate; all rates except AMOSS RHD-P prevalence are for females with RHD.

therapy and for women with mitral stenosis.[6] The burden of RHD in pregnancy is under-researched—the majority of studies examine severe disease in non-pregnant adults, all-cardiac disease in pregnancy or single-site studies, mostly in high-prevalence countries of sub-Saharan Africa, Asia and South America.[7–10] There are no known national population-based studies of RHD in pregnant women.

Commencing in 2013, a 2-year surveillance and descriptive study of the prevalence, management and outcomes of rheumatic heart disease in pregnancy (RHD-P) across ANZ was undertaken against a backdrop of increased advocacy and growing recognition of the burden—both global and country-specific—of RHD.[11–14]

Objective

The Australasian Maternity Outcomes Surveillance System (AMOSS) RHD in pregnancy study aimed to identify all pregnant women with RHD who presented at any of 284 participating AMOSS maternity sites across Australia (n=262) and New Zealand (n=22) during 2013 and 2014, for inclusion in a descriptive study of clinical backgrounds, models of care and cardiac, obstetric and perinatal outcomes. The objective of this paper is to identify the challenges of surveillance in the Australian arm of the study, and the strategies developed to strengthen reporting by—and improve awareness among—health services.

Materials and methods

Inclusion

The AMOSS study used World Heart Federation (WHF) criteria based on echocardiographic diagnosis of RHD[15] to identify women for inclusion. Pregnant women (20 or more weeks' gestation) were included if they had confirmed RHD based on their most recent echocardiogram report, or a historic echocardiogram where the most recent echocardiogram report was not available. Because a case was defined during pregnancy, women could potentially be included more than once during the 24-mo study period. Women were excluded if they had a miscarriage or termination of pregnancy before 20 weeks' gestation, or if the baby's date of birth was outside the study period (1 January 2013 to 31 December 2014).

Reporting

The study was conducted under the umbrella of the AMOSS.[1] This hospital-based surveillance and research system was established across ANZ in 2009 in response to the lack of national information on the incidence, risk factors, management and sequelae of rare and serious conditions in pregnancy. Volunteer site coordinators at participating AMOSS maternity sites (predominantly midwives and obstetricians—'AMOSS data collectors') report and complete web-based surveys on nominated conditions over defined study periods. De-identified data are entered from case notes.

Negative reporting was employed; monthly emails were sent to AMOSS data collectors during 2013–2014 asking whether or not they had any women under their care with RHD in the preceding month. This established AMOSS reporting system (Figure 2) was supplemented by additional notification methods in the RHD-P study (Figure 2 and Table 1), although the AMOSS remained the central notification point. This extensive network of stakeholders helped optimize surveillance and strengthen awareness of the study.

Specific approaches to case ascertainment were developed according to jurisdiction and site. AMOSS maternity site data collectors primarily used perinatal data and health information systems. The AMOSS project coordinators from the study team

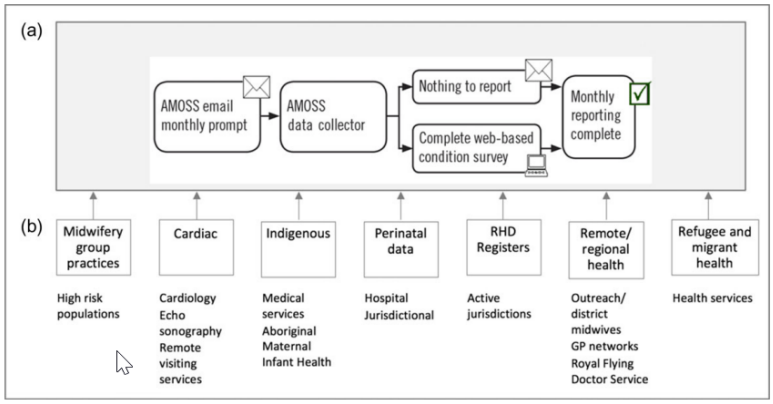


Figure 2. AMOSS surveillance and research platform (262 maternity units in Australia). Usual AMOSS reporting cycle (a) supplemented by RHD-P enhanced network (b).

Table 1. Confirmed cases of women giving birth with RHD reported according to source[a]

Confirmed cases: notification source	n	% of total 192 cases
AMOSS		
AMOSS participating maternity site data coordinator (sole notification)	106	55%
All Australian cases: AMOSS participating maternity site data coordinator or NT AMOSS coordinator (multiple notification with other sources possible)	181	94%
NT cases: AMOSS NT coordinator (multiple notification with other sources possible)	59	31%
Other sources (non-AMOSS coordinator)		
Midwifery group practice/antenatal care/remote area or district midwife/remote medical officer/general practitioner	22	11%
Obstetrician/obstetric physician/obstetric registrar	7	4%
Aboriginal maternal and infant care/Aboriginal medical service/Indigenous Cardiac Outreach Program	6	3%
Cardiologist/cardiac nurse/anaesthetic/echocardiogram technician/other specialist service	8	4%
RHD registers (four active jurisdictions during study)	26	14%
Total	**192**	

[a]More than one source notification possible, percentages are of the total 192 cases.

queried additional systems in high prevalence regions, including remote/primary health information systems and RHD control registers, using the broad search terms 'cardiac' and 'rheumatic' in the search criteria, and reviewing individual case notes. Similar enhanced case note reviews of perinatal data systems were conducted by the study team at three major tertiary sites outside the NT. In the NT, which has the highest reported rates of RHD in Australia among Aboriginal women, a dedicated project coordinator conducted a validation study. Where the echocardiogram report was inconclusive in this jurisdiction, the actual echocardiogram was reviewed by a cardiologist.

In New South Wales (NSW), through consultation with Western NSW Aboriginal Maternal Infant Health Service and the 'ObstetriX/eMaternity' perinatal data working group, questions for women at the antenatal booking visit were revised to include detailed prompts on RF/RHD history, including whether the woman remembered having regular intramuscular injections (prompt for secondary prophylaxis) as a child, as well as her heart history.

Following case confirmation, AMOSS site data collectors or the project coordinators completed web-based surveys, covering demographic, general medical/obstetric history, pregnancy pathway, and maternal obstetric/cardiac and perinatal outcomes. The authors

revised the study protocol during a 2-mo pilot phase to send echocardiogram reports directly to the study team for entry, in order to confirm case inclusion, reduce resource burden and achieve optimal consistency. In addition to the NT, AMOSS project coordinators supported case note review and completion of surveys in other high prevalence regions, particularly the Kimberley region of (north) Western Australia and far north Queensland, and/or where the resource burden meant that onsite staff required assistance.

Specific data items identified whether the woman had been reported in a previous pregnancy during the study period, as well as number of months since the last pregnancy.

Duplication was checked through a tiered process, including continual monitoring of the reporting database, probabilistic methods (using a series of concatenated data fields) and checking with the AMOSS site data collector. The survey was completed at the site where the woman gave birth.

Ethics and consultative processes

Ethics approval requests under the aegis of the AMOSS were submitted to 32 Australian ethics committees and over 200 affiliated governance sites.[1] Subsequent amendments repeated this process as the study protocol was revised during the pilot phase. Access to sites for case note review and/or data entry directly by the research team was also approved where requested by the AMOSS site coordinator and for all NT. De-identified data were collected and reported at an aggregate level only. No consent was required for this quantitative arm of the RHD-P study. Aboriginal health services and Aboriginal Maternal Infant Health Services endorsed the study through letters of support.

Formal and informal consultative processes were established and continued throughout the research project in order to build awareness of the study (and more generally about the impact of RHD-P), optimize notification processes and provide avenues for dissemination of findings (Figure 2).

An Advisory Group comprised Australia and New Zealand investigators, the study team, invited representatives from Aboriginal, cardiac and maternity services, public health, jurisdictional and RHD organizations, and related policy bodies.

Results

Surveillance

The AMOSS network of 262 Australian sites notified of 495 potential cases (Figure 3a). Of these, 246 did not meet the inclusion criteria. There was no evidence of RF or RHD in 99 cases (identified predominantly through the perinatal data system search), and in 147 cases there was RF only or a valvular RHD lesion that had resolved. An additional 32 women gave birth outside the study period, and eight women who miscarried or who had a surgical termination of pregnancy before 20 weeks' gestation were excluded. No data were received for four cases and 13 cases were duplicate notifications. A total of 192 pregnancies of Australian women with confirmed RHD (according to WHF criteria for echocardiographic diagnosis of RHD) who gave birth (≥20 weeks' gestation) were included in the Australian arm of the study.

In the NT, where the validation study was performed, 126 of 211 cases reviewed did not meet inclusion criteria. Sixty-eight cases reviewed were excluded due to lack of evidence of RF or RHD, and a further 58 had either RF only or resolved RHD (Figure 3b).

Data collection processes

Notification and data collection processes varied according to jurisdiction, geography and site.

Notification

Cases were principally notified by data collectors at participating AMOSS maternity units or the NT-based AMOSS coordinator (181/192 94% eligible women), supported by other notifications such as Midwifery Group Practices, antenatal clinics and cardiac, community, remote, primary health and Aboriginal health services, including the Indigenous Cardiac Outreach Project (Table 1, with multiple notifications possible). In the NT, notification sources were documented by health group and location (Figure 4).

The RHD-P study partnered with jurisdictional RHD control programmes to promote register functions and purpose, with registers assisting in notification. Additionally, RHD programme staff in some jurisdictions had developed care plans and notification prompts that aided reporting.

A total of 106 cases (55%) were notified solely by the AMOSS maternity site data collector.

Where transfer of care occurred, a post-hoc review of data to ascertain any duplication of cases was conducted. Two women were lost to follow-up when they moved away in early pregnancy from the maternity site where they attended the antenatal 'booking' visit.

Data collection

Case note reviews entailed querying up to seven distinct information systems, in addition to reviewing paper-based files in sites where hybrid electronic-paper systems were active. In the high prevalence NT, the seven information domains included two discrete primary healthcare information systems that did not integrate with each other, and a privately run cardiac practice that provides services to the predominantly government-funded NT health system (Figure 5). Jurisdictional RHD registers are separate entities from each other, which may or may not articulate with the hospital and primary health information systems.

Prevalence

The Australian rate of RHD in pregnancy was 4.3 per 10,000 women giving birth. Of the confirmed 192 cases from six jurisdictions included in the study, overall rates ranged from 0.7 per 10,000 women giving birth in Victoria to 74.3 per 10,000 women giving birth in the NT (Figure 6). There were no confirmed cases in two jurisdictions. Rates among Aboriginal and/or Torres Strait Islander women ranged from 7.0 per 10,000 women giving birth in NSW to 222.2 per 10,000 in the NT

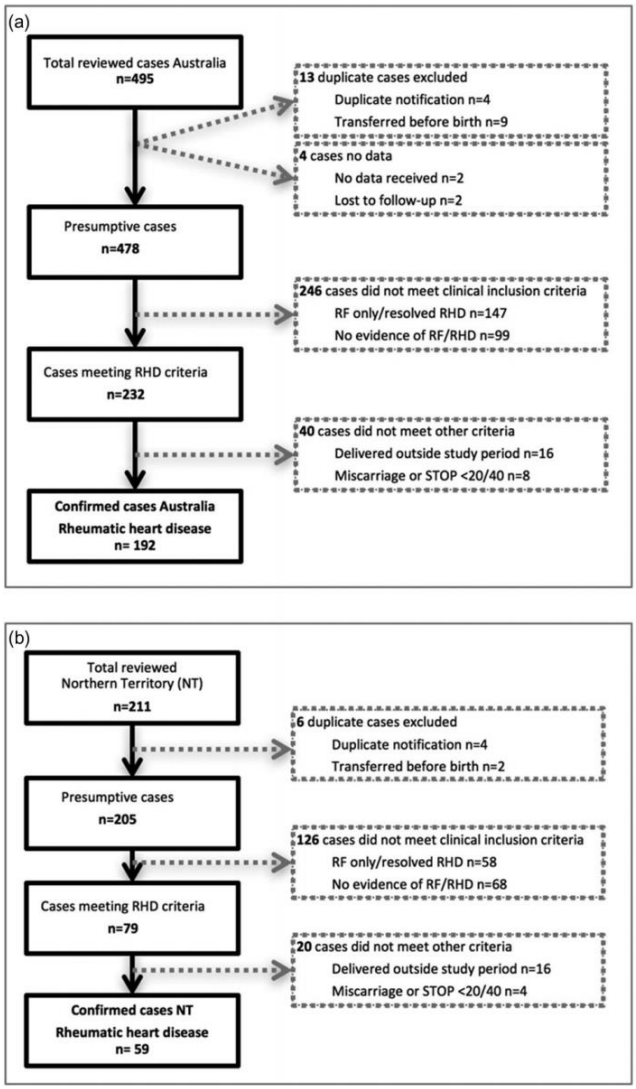


Figure 3. Surveillance of pregnant women with RHD: (a) Australia and (b) Northern Territory 2013–2014.

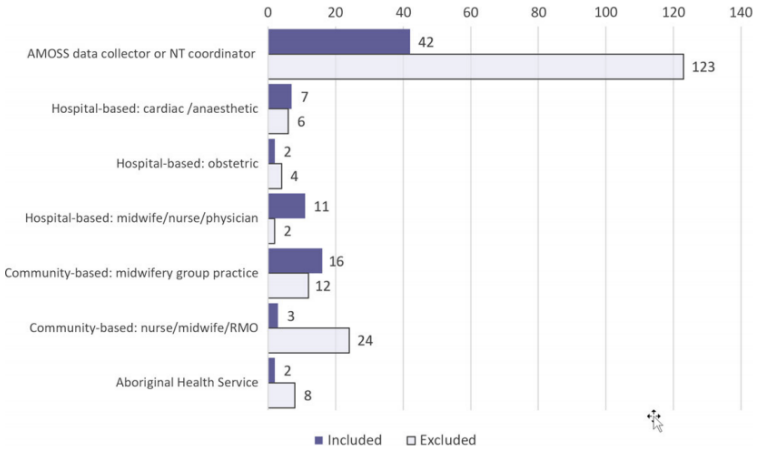


Figure 4. AMOSS surveillance and research platform (262 maternity units in Australia. NT breakdown of reported cases by health group/location and inclusion.

(Figure 6). Overall, 78% (n=150) of Australian women included in the study were Aboriginal and/or Torres Strait Islander. The NT had 59 (31%) of cases, 99% of whom were Aboriginal women.

Discussion

As well as providing multidisciplinary clinical care for women with known RHD, pregnancy provides the opportunity to identify newly diagnosed cases of RHD, re-engage with women who may have 'dropped out' of care as they transition from paediatric to adult specialist cardiac services, and provide language-appropriate health education that promotes a shared understanding of the impact of RHD and its implications in pregnancy.[16] However, this study highlights challenges to effective provision of care across several tiers, relating to case ascertainment, burden of reporting and health information systems. These factors impacted on a group of at-risk women with complex health needs and high rates of co-morbidities. Strategies developed to help support case notification underscored the value of an extended reporting network across health sectors, and reinforce the value of collaborative care.

Case ascertainment

High false positive rates (Figures 3 and 4) are consistent with preliminary active case finding work being undertaken by jurisdictional RHD Registers (personal communication) and the 'End RHD' Centre for Research Excellence[17]), which suggests significant over-ascertainment of hospital cases of RHD through ICD 10 coding, including valvular heart disease of unspecific origin. In the NT, where the validation study was conducted, a shortened case survey data collection was performed of the 58 women excluded from the study with either RF only or resolved RHD, with the aim of examining the burden on NT health services of being incorrectly diagnosed with RHD according to WHF criteria.

As well as over-reporting, under-reporting is assumed to have occurred of women with mild asymptomatic RHD without a case history or resultant complications who did not have an echocardiogram and were not reported. A number of women with a history of RF were reported, but definitive diagnosis or exclusion of RHD could not occur due to lack of supporting evidence (by echocardiogram/cardiac review), including five women born overseas.

The AMOSS system is based on reported cases of women with RHD admitted to maternity units at 20 weeks' gestation or more: yet RHD is associated with higher rates of miscarriage, as well as perinatal death.[18] In the NT review, an estimated 7% of otherwise eligible women with RHD miscarried, or were recommended for termination of pregnancy due to their cardiac condition. Thus, the true prevalence of RHD-P is higher than documented in this study.

Burden of reporting

The AMOSS system researches morbidities in pregnancy with an (overall) estimated prevalence of less than 1:1000 women giving birth – completed studies included – for example, H1N1 influenza, amniotic fluid embolism, vasa previa. Smaller maternity sites typically report an AMOSS condition once every several years, if at all. However, in this study, there was a significant burden on data collectors in high prevalence regions, where sites with 200–300 births per year had eight or more cases of women with RHD. This site burden was compounded by the

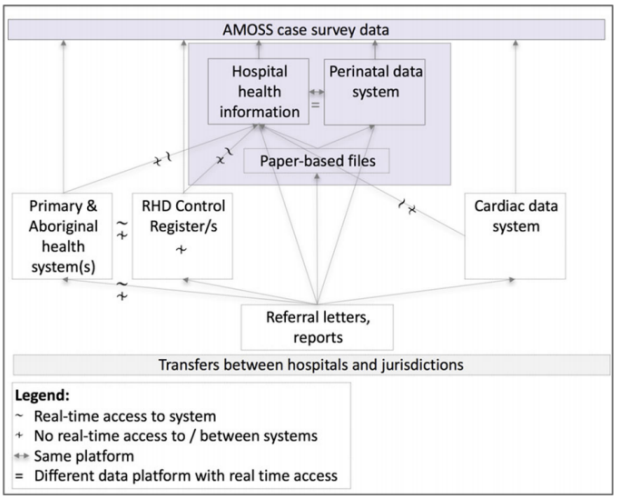


Figure 5. Data sources: surveillance and data collection.

work involved in information retrieval (Figure 5 and the next section).

Health information systems

Our study found a lack of cohesion in standard data collection systems, including multiple systems, gaps in accurate case ascertainment, and effective transfer of information between and across health services and jurisdictions. There were multiple sources of data, with some sites employing a mix of electronic and paper-based record systems, various electronic data systems in remote and primary healthcare networks that did not articulate, and gaps in sharing that information across networks, including Aboriginal health services.

There was a significant variation in the amount and quality of information provided in echocardiogram reporting used to determine inclusion. Reports ranged from detailed records to handwritten excerpts. They were often not included in patient notes where women were transferred or had the echocardiogram performed by a private provider, or were unavailable for women born outside of Australia. Additionally, the lack of standardized reference values in echocardiogram reporting impacts on data integrity and clinical decision-making,[19] particularly for pregnant women.

The World Health Organization Roadmap for Action[20] notes that 'Only by disaggregating and analysing data can populations in need of health services be identified and included in informed policies and programmes'. Our study highlights the need for improved RHD annual reporting and analysis at a national level with disaggregation by gender together with Indigenous identification and age.

The National Data Collection System (NDCS; RHDAustralia-managed during the study period) is the central repository for the collection and reporting of RF and RHD data, yet it only reports on data from the jurisdictional register-based control programmes for RF/RHD (four out of a total of eight Australian jurisdictions during the study period).[21] Pregnancy status was a recommended data item in the NDCS recommended dataset in 2011. However, no jurisdictions currently monitor pregnant status within RHD Registers. This sharing of health information would be further strengthened by including pregnant status as a data item in RHD Registers.

Strategies to improve surveillance and build awareness

Streamlined, multi-tiered surveillance processes were established in this study to identify pregnant women with RHD across ANZ. While the AMOSS system remains a timely and reliable primary source of notification of (overall) rare conditions in pregnancy, these multi-tiered processes demonstrated that employing additional reporting sources can provide an effective surveillance adjunct,[22] and a useful augmentation strategy to better inform the research of health risk, diagnosis, management and pregnancy outcomes of women with RHD. This is particularly significant for RHD, where a missed diagnosis or lack of awareness can have such a detrimental impact in pregnancy.

The call for multi-disciplinary care that straddles specialist cardiac and obstetric disciplines for pregnant women with cardiac disease is not new.[6,23] However, this study highlighted the

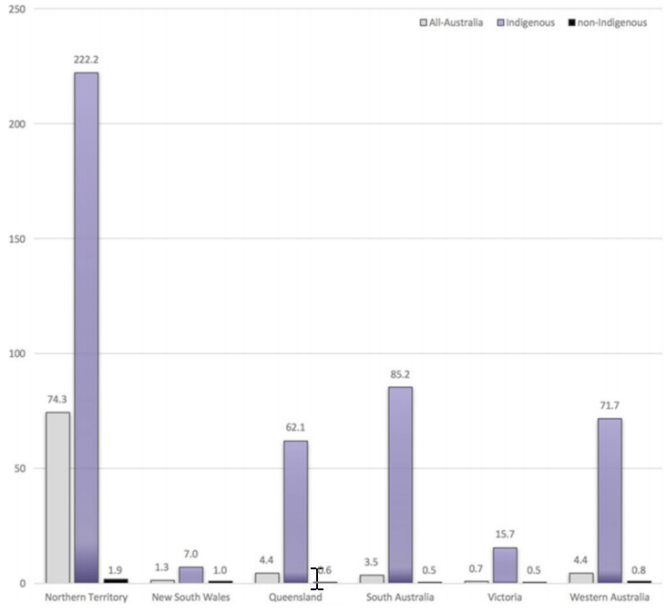


Figure 6. Rates per 10 000 Australian women giving birth with RHD by Australian jurisdiction 2013–2014.

benefit – and imperative – of building integrative, diagonal approaches to care[14,24] across all maternity, Aboriginal, and primary and public healthcare services (including RHD programmes), in order to support an optimal pathway of care, particularly early diagnosis and assessment.

This more integrative approach to care is consistent in study findings and reports ranging from chronic disease in Aboriginal peoples to international studies of maternal mortality[25,26] and to global RHD initiatives. Many chronic conditions face similar hurdles in achieving effective health information and communication sharing, particularly at the interface of primary and hospital-based care, and especially with remote Aboriginal and Torres Strait Islander health communities.[27] In their critique of Aboriginal health cardiac rehabilitation, DiGiacomo et al. similarly argue that inadequate referral systems, fragmented health information systems and gaps in coordinated health services create significant barriers to access for Aboriginal patients. They also point to inadequate resourcing of the Aboriginal health worker workforce, instrumental in supporting continuity of culturally competent care and making connections with various health agencies, also emphasized by Kelly et al.[28] Integration of RHD interventions with essential packages of health services, including maternal and child health, is consistent with strategies called for by RHD advocacy groups and initiatives at the global public health level.[11,29,30]

Study limitations

Miscarriage or surgical termination of pregnancy often occurred outside participating AMOSS maternity units. Thus, accurate case ascertainment of all women with pregnancy outcomes at <20 weeks' gestation was not possible across Australia in this cohort of women with a higher risk of pregnancy complications due to their cardiac status.

The study reports on one high-resource country and findings may not be as applicable in low-income countries. These findings do, however, parallel the overall health status of Aboriginal and Torres Strait Islander peoples in Australia. Indigenous identification was probably under-reported and thus the burden of RHD is under-estimated in this population, highlighting the need for health professionals to ask all women whether they identify as being of Aboriginal and/or Torres Strait Islander descent.

Moreover, themes raised by this study related to RHD awareness among maternal and primary health services are consistent with conclusions and recommendations in studies of RHD in pregnancy in endemic regions, and in global RHD strategies.

Conclusions

Optimal outcomes for pregnant women with RHD demand timely diagnosis, access to integrative, appropriate models of

care and health systems. The potential consequences of undiagnosed RHD in a pregnant woman demonstrate the value of enhanced reporting. Our study of the implementation of a population-based study of RHD in pregnancy identifies gaps in reporting and health information at a system level, as well as strategies developed to address these.

Our study findings call for implementation of pregnant status as a data item in jurisdictional RHD registers, specific questions related to RF/RHD history built into perinatal data systems, improved real-time access to RHD register information by hospital sites (and to other registers), improved education to support a more informed health workforce,[31] and a more granular reporting of RHD in national data sets of prevalence and mortality, broken down by gender. The study highlights the need to promote greater awareness among the maternity sector in order to detect RHD, provide early monitoring in pregnancy, and strengthen collaboration between disciplines in the care of pregnant women with this preventable condition.

The AMOSS RHD in pregnancy study investigators (NHMRC #1024206): (Chief) Professors Elizabeth Sullivan, University of Technology Sydney, Faculty of Health, Australian Centre for Public and Population Health Research (ACPPHR), Sydney, Australia, Lisa Jackson Pulver, Western Sydney University, Parramatta, Australia, Jonathan R Carapetis, Telethon Kids Institute, University of Western Australia and Princess Margaret Hospital for Children, Perth, Australia, Dr Warren Walsh, University of New South Wales and Prince of Wales Hospital, Sydney, Australia, Professor Michael Peek, The Australian National University and Centenary Hospital for Women and Children, Canberra, Australia, Dr Claire McLintock, National Women's Health Auckland City Hospital, Auckland, New Zealand, Professor Sue Kruske, Maternal and Child Health Institute for Urban Indigenous Health, Brisbane, Australia, Dr Suzanne Belton, Menzies School of Health Research, Darwin, Australia. (Associate) Professor Alex Brown, South Australian Health & Medical Research Institute (SAHMRI), Adelaide, Australia, A/Professor Elizabeth Comino, Centre for Primary Health Care and Equity, University of New South Wales, Sydney, Australia, Ms Heather D'Antoine, Menzies School of Health Research, Darwin, Australia, Dr Simon Kane, Lyell McEwin Hospital, Adelaide, Australia, Dr Bo Remenyi, Royal Darwin Hospital and NT Cardiac, Darwin, Australia, Professor Juanita Sherwood, University of Sydney, Sydney, Australia, Dr Sujatha Thomas, Royal Darwin Hospital, Darwin, Australia, Geraldine Vaughan, University of Technology Sydney, Faculty of Health, Australian Centre for Public and Population Health Research (ACPPHR), Sydney, Australia.

Authors' contributions: GV was responsible for the study design and implementation, analysis and interpretation of the data, contributed to the writing of this article, and read and approved the final version. KT was responsible for the study design and implementation, and read and approved the final version. MJP, BR and EAS were responsible for the study design, and the analysis and interpretation of the data. LJP was responsible for the study design, analysis and interpretation of the data, contributed to the writing of this article, and read and approved the final version. SB was responsible for the analysis and interpretation of the data, and read and approved the final version.

Acknowledgements: The RHD in pregnancy study (NHMRC #1024206) could not have occurred without the ongoing support of data coordinators at all AMOSS maternity sites, as well as all those working in Aboriginal and remote/primary healthcare; Midwifery Group Practices; Aboriginal Maternal Infant Health Services, particularly Western NSW LHD; cardiac services, including NT Cardiac and the Indigenous Cardiac Outreach Project; jurisdictional RHD Control Registers and RHDAustralia – and many other individuals and groups. Thank you.

Funding and other support: The authors gratefully acknowledge aid from the Australian National Health and Medical Research Council (NHMRC) project grant #1024206 and NHMRC Postgraduate Scholarship #11332944; University of Technology Sydney Chancellor's Research Scholarship; and END RHD Centre of Research Excellence, Telethon Kids Institute, University of Western Australia. Sincere thanks go to the facilitators and participants of the Twelve Weeks to Publication Program funded by Faculty of Health, University of Technology Sydney.

Competing interests: None declared.

Ethical approval: Ethical approval for the Australian arm of the AMOSS RHD in pregnancy study (NHMRC #1024206) was granted by the NSW Population and Health Services Research Ethics Committee (2009/03/144), Menzies School of Health Research and multiple other Human Research Ethics Committees including Aboriginal ethics.

References

1 Australasian Maternity Outcomes Surveillance System (AMOSS). http://www.amoss.com.au/publications [accessed 14 June 2018].
2 Australian Institute of Health and Welfare (AIHW). Australian Burden of Disease Study: Impact and causes of illness and death in Aboriginal and Torres Strait Islander people 2011. Australian Burden of Disease Study series no. 6. Cat no. BOD 7. Canberra: AIHW; 2016.
3 Heart Foundation of New Zealand. New Zealand Guidelines for Rheumatic Fever: Diagnosis, Management and Secondary Prevention of Acute Rheumatic Fever and Rheumatic Heart Disease: 2014 Update. Heart Foundation of New Zealand; 2014.
4 Zühlke L, Karthikeyan G, Engel ME et al. Clinical outcomes in 3343 children and adults with rheumatic heart disease from 14 low- and middle-income countries: two-year follow-up of the Global Rheumatic Heart Disease Registry (the REMEDY Study). Circulation 2016;134(19):1456–66.
5 Doukky R, Abusin SA, Bayissa YA et al. Rheumatic heart disease in modern urban America: a cohort study of immigrant and indigenous patients in Chicago. Int J Cardiol 2014;175(1):178–80.
6 Elkayam U, Goland S, Pieper P et al. High-risk cardiac disease in pregnancy Part I. J Am Coll Cardiol 2016;68(Jul 26(4)):396–410.
7 Elliott C, Sliwa K, Anthony J. Perinatal outcome in pregnant women with heart disease attending a combined obstetric and cardiology clinic in a resource limited country. Int J Gynecol Obst Neonatal Care 2015;2(2):8–15.
8 Abdel-Hady E, El-Shamy M, El-Rifai A et al. Maternal and perinatal outcome of pregnancies complicated by cardiac disease. Int J Gynaecol Obstet 2005;90(1):21–5.
9 Campanharo FF, Cecatti JG, Haddad SM et al. The impact of cardiac diseases during pregnancy on severe maternal morbidity and mortality in Brazil. PLoS One 2015;10(12):e0144385.
10 Sartain JB, Anderson NL, Barry JJ et al. Rheumatic heart disease in pregnancy: cardiac and obstetric outcomes. Intern Med J 2012;42(9):978–84.
11 World Health Organisation (WHO) Executive Board. Rheumatic fever and rheumatic heart disease: Report by the Director-General. Seventy-first World Health Assembly A71/25 Provisional agenda item 12.8. Geneva, Switzerland; 2018.

12 RHD Action. RHD Global Status Report 2015–2017. Geneva: World Heart Federation; 2016.

13 Australian Medical Association (AMA). AMA Report Card on Indigenous Health: a call to action to prevent new cases of rheumatic heart disease in indigenous Australia by 2031. Canberra: AMA; 2016.

14 Watkins DA, Zühlke LJ, Narula J. Moving forward the RHD agenda at global and national levels. Global Heart 2017;12(1):1–2.

15 Reményi B, Wilson N, Steer A et al. World Heart Federation criteria for echocardiographic diagnosis of rheumatic heart disease—an evidence-based guideline. Nat Rev Cardiol 2012;9(5):297–309.

16 Belton S, Kruske S, Jackson Pulver L et al. Rheumatic heart disease in pregnancy: how can health services adapt to the needs of Indigenous women? A qualitative study. Aust NZ J Obstet Gynaecol 2018; 58(4). doi:10.1111/ajo.12744.

17 Katzenellenbogen J, Kruger D, Nedkoff L et al. Over-counting rheumatic heart disease in hospital administrative data: does it matter and what can be done? Heart, Lung Circul 2017;26:S332–3.

18 RHDAustralia (ARF/RHD writing group). National Heart Foundation of Australia and the Cardiac Society of Australia and New Zealand. Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease (2nd edition). Casuarina NT: ARF/RHD; 2012.

19 Lancellotti P, Badano L, Lang R et al. Normal reference ranges for echocardiography: rationale, study design, and methodology (NORRE Study). Eur Heart J Cardiovasc Imag 2013;14(4):303–8.

20 World Health Organisation. Integrating equity, gender, human rights and social determinants into the work of WHO: Roadmap for Action (2014–2019). WHO/FWC/GER/15.2. Geneva: World Health Organization; 2015.

21 Health Policy Analysis. Evaluation of the Commonwealth Rheumatic Fever Strategy – Final report. Canberra, Primary Healthcare Branch, Commonwealth Department of Health; 2017.

22 Knowles R, Smith A, Lynn R et al. Using multiple sources to improve and measure case ascertainment in surveillance studies: 20 years of the British Paediatric Surveillance Unit. J Publ Health (Bangkok) 2006; 28(2):157–65.

23 Mocumbi AO, Sliwa K. Women's cardiovascular health in Africa. Heart 2012;98(6):450–5.

24 Frenk J, Gómez-Dantés O. False dichotomies in global health: the need for integrative thinking. Lancet 2017;389(10069):667–70.

25 Knight M, Nair M, Tuffnell D et al. Saving lives, improving mothers' care—surveillance of maternal deaths in the UK 2012–14 and lessons learned to inform maternity care from the UK and Ireland Confidential Enquiries into Maternal Deaths and Morbidity 2009–14. Oxford: National Perinatal Epidemiology Unit; 2016.

26 Soma-Pillay P, Seabe J, Sliwa K. The importance of cardiovascular pathology contributing to maternal death: Confidential Enquiry into Maternal Deaths in South Africa, 2011–2013. Cardiovasc J Afr 2016; 27(2):60–5.

27 DiGiacomo M, Davidson P, Taylor K et al. Health information system linkage and coordination are critical for increasing access to secondary prevention in Aboriginal health: a qualitative study. Qual Prim Care 2010;18:17–26.

28 Kelly J, Ramage M, Perry D et al. Managing two worlds together. Stage 3: improving Aboriginal patient journeys—cardiac case studies. Melbourne: The Lowitja Institute; 2015.

29 Watkins D, Zühlke L, Engel M et al. Seven key actions to eradicate rheumatic heart disease in Africa: the Addis Ababa communique. Cardiovasc J Afr 2016; 27:184–7.

30 Palafox B, Mocumbi AO, Kumar RK et al. The WHF roadmap for reducing CV morbidity and mortality through prevention and control of RHD. Global Heart 2017;12(1):47–62.

31 RHDAustralia. RHDAustralia. http://www.rhdaustralia.org.au [accessed 15 June 2018].

32 Ruddock P. Aboriginal Health: report from the House of Representatives Standing Committee on Aboriginal Affairs. Canberra: Parliament of the Commonwealth of Australia; 1979.

33 Freund S. Rheumatic heart disease program, in Northern Territory Communicable Diseases Bulletin, vol 4, no 4 Dec. Darwin: Centre for Disease Control, NT; 1997.

34 Commonwealth Department of Health. Health Policy Analysis. Evaluation of the Commonwealth Rheumatic Fever Strategy—Final report. Canberra: Commonwealth Department of Health; 2017.

35 Australian Bureau of Statistics. In: 3303.0 Causes of Death, Australia, 2015, Table 1.1. Canberra: Australian Bureau of Statistics (ABS); 2016.

Appendix 7: Authors' contributions and signatures

Chapter 4: *Standardizing clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis* Birth: Issues in Perinatal Care [early view]

Conceptualization, review design: Geraldine Vaughan (GV), Angela Dawson (AD), Elizabeth Sullivan (EAS). Drafting of manuscript GV, AD, EAS with input from Michael Peek (MP), Jonathan Carapetis (JC). Study selection and data extraction: GV and AD. Analysis and interpretation: GV, EAS, AD. Critical revision of the review for intellectual content: GV, EAS, AD, MP, JC.

Geraldine Vaughan	Production Note: Signature removed prior to publication.
Professor Angela Dawson	Production Note: Signature removed prior to publication.
Professor Michael Peek	Production Note: Signature removed prior to publication.
Professor Jonathan Carapetis	Production Note: Signature removed prior to publication.
Professor Elizabeth Sullivan	Production Note: Signature removed prior to publication.

Chapter 6: ***Review of strategies and lessons learnt implementing a population-based study of rheumatic heart disease in pregnancy in Australia*** **Int Health. 2018;10(6):480-9.**

Geraldine Vaughan (GV) was responsible for the study design and implementation, analysis and interpretation of the data, contributed to the writing of this article, and read and approved the final version. Kylie Tune was responsible for the study design and implementation, and read and approved the final version. Michael Peek, Bo Remenyi and Elizabeth Sullivan were responsible for the study design, and the analysis and interpretation of the data. Lisa Jackson Pulver was responsible for the study design, analysis and interpretation of the data, contributed to the writing of this article, and read and approved the final version. Suzanne Belton was responsible for the analysis and interpretation of the data, and read and approved the final version.

Geraldine Vaughan	Production Note: Signature removed prior to publication.
Kylie Tune	Production Note: Signature removed prior to publication.
Professor Michael Peek	Production Note: Signature removed prior to publication.
Professor Lisa Jackson Pulver	Production Note: Signature removed prior to publication.
Dr Bo Remenyi	Production Note: Signature removed prior to publication.
Dr Suzanne Belton	Production Note: Signature removed prior to publication.
Professor Elizabeth Sullivan	Production Note: Signature removed prior to publication.

Appendix 8: Presentations

Chapter 4

In addition to publication, elements from this study were also presented at the following conferences and meetings:

Cardiac Society of Australia and New Zealand (CSANZ) 3rd Indigenous Cardiovascular Health Conference, 2019, Wellington New Zealand. Vaughan G, Wheeler M, Bootle L, Noonan S, Slade B, Wade V. *Beyond pregnancy: Women with rheumatic heart disease* (Poster presentation)

Perinatal Society of Australia & New Zealand (PSANZ) 2019 annual Congress Gold Coast, Australia. Vaughan GA, Dawson A, Peek M, Carapetis J, Sullivan EA. *Rheumatic heart disease in women: can improving reporting measures support better outcomes?* (Poster presentation)

South Africa Heart Congress Johannesburg October 2018. Vaughan GA, Dawson A, Peek M, Carapetis J, Sullivan EA. *Models of care for pregnant women with rheumatic heart disease: a qualitative evidence synthesis* (Poster presentation)

University of Technology Sydney Graduate Research Forum 2018. Vaughan GA, Dawson A, Peek M, Sullivan EA. *Rheumatic heart disease in women: can improving reporting measures support better outcomes?* (Oral presentation)

Chapter 5

Elements of this study were presented at the following conferences and meetings:

Cardiac Society of Australia and New Zealand (CSANZ) 3rd Indigenous Cardiovascular Health Conference, 2019, Wellington New Zealand. Vaughan, G, Dawson A, Peek MJ, Sullivan EA. *Better care for pregnant women with RHD: what works?* (Oral presentation)

Cardiac Society of Australia and New Zealand (CSANZ) 3rd Indigenous Cardiovascular Health Conference, 2019, Wellington New Zealand. Vaughan G, Belton S, La Vincente S, Bootle L, Sullivan E. *Using research to design educational resources for women with rheumatic heart disease* (Poster presentation)

Perinatal Society of Australia & New Zealand (PSANZ) 2018 Annual Congress Auckland, New Zealand. Vaughan G, Dawson A, Peek MJ, Sullivan EA. *Language of the heart? Health*

services perspectives on care of pregnant women with rheumatic heart disease. (Poster presentation)

Cardiac Society of Australia and New Zealand (CSANZ) Perth August 2017. Vaughan GA, Dawson A, Peek M, Sullivan EA. *Rheumatic heart disease in pregnancy: a health services challenge.* (Oral presentation)

University of Technology Sydney Graduate Research Forum 2017. Vaughan GA, Dawson A, Peek M, Sullivan EA. *Rheumatic heart disease in pregnancy: a health services challenge.* (Oral presentation)

Chapter 6

In addition to publication, elements of this study were also presented at the following conferences and meetings:

Cardiac Society of Australia and New Zealand (CSANZ) Perth August 2017. Vaughan GA, Dawson A, Peek M, Sullivan EA. *Rheumatic heart disease in pregnancy: a health services challenge.* (Oral presentation)

University of Technology Sydney Graduate Research Forum 2017. Vaughan GA, Dawson A, Peek M, Sullivan EA. *Rheumatic heart disease in pregnancy: a health services challenge.* (Oral presentation)

Bibliography

1. RHDAustralia (ARF/RHD writing group). *Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease (3nd edition) (Under development).* RHDAustralia, Heart Foundation of Australia; Cardiac Society of Australia and New Zealand; 2019.
2. NSW Health. *Communicating positively: A guide to appropriate Aboriginal terminology.* Sydney 2004.
3. Australian Indigenous Health*Info*Net. The Australian Indigenous Health*Info*Net guidelines for Aboriginal and Torres Strait Islander terminology. 2018; https://healthinfonet.ecu.edu.au/uploads/resources/30201_30201.pdf.
4. Ministry of Education. *Pasifika Peoples: A note on terminology.* Wellington: NZARE; 2007.
5. RHD Action. *RHD Action Prospectus.* Perth, Australia 2017.
6. RHDAustralia (ARF/RHD writing group). *Australian guideline for prevention, diagnosis and management of acute rheumatic fever and rheumatic heart disease (2nd edition).* RHDAustralia, Heart Foundation of Australia; Cardiac Society of Australia and New Zealand; 2012. 978-0-9587722-9-7 (paperback); 978-0-9587722-5-9 (online).
7. World Health Organisation (WHO). *WHO Model Prescribing Information: Drugs Used in the Treatment of Streptococcal Pharyngitis and Prevention of Rheumatic Fever.* Geneva: WHO; 1999.
8. Zühlke LJ, Beaton A, Engel ME, et al. Group A streptococcus, acute rheumatic fever and rheumatic heart disease: epidemiology and clinical considerations. *Current Treatment Options in Cardiovascular Medicine.* 2017;19(2):15.
9. Wyber R, Taubert K, Marko S, Kaplan EL. Benzathine Penicillin G for the Management of RHD: Concerns About Quality and Access, and Opportunities for Intervention and Improvement. *Global Heart.* 2013;8(3):227-234.
10. Gordon J, Kirlew M, Schreiber Y, et al. Acute rheumatic fever in First Nations communities in northwestern Ontario: Social determinants of health "bite the heart". *Canadian Family Physician Medecin De Famille Canadien.* 2015;61(10):881-886.
11. Doukky R, Abusin SA, Bayissa YA, Kelly RF, Ansari AH. Rheumatic heart disease in modern urban America: A cohort study of immigrant and Indigenous patients in Chicago. *International Journal of Cardiology.* 2014;175(1):178-180.
12. Elkayam U, Bitar F. Valvular heart disease and pregnancy. Part I: native valves. *J Am Coll Cardiol.* 2005;46(July 19;(2)):223-230.
13. Diao M, Kane A, Ndiaye M, et al. Pregnancy in women with heart disease in sub-Saharan Africa. *Arch Cardiovasc Dis.* 2011;104(6-7):370-374.
14. Australian Institute of Health and Welfare (AIHW). *Rheumatic heart disease: all but forgotten in Australia except among Aboriginal and Torres Strait Islander peoples.* Canberra ACT, Australia: AIHW; 2004.
15. Vaughan G, Tune K, Peek M, et al. Rheumatic heart disease in pregnancy: strategies and lessons learnt implementing a population-based study in Australia. *Int Health.* 2018;10(6):480-489.
16. Sullivan E, Vaughan G, Li Z, et al. The high prevalence and impact of rheumatic heart disease in pregnancy in First Nations populations in a high-income setting: a prospective cohort study. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2019;00.

17. WHO Director-General. *Provisional agenda of the seventy-first World Health Assembly. 142nd session.* Geneva 2018.
18. World Health Organisation (WHO). *Global Strategy for Women's, Children's and Adolescents' Health: 2018 monitoring report: current status and strategic priorities.* Geneva: WHO; 2018.
19. World Health Organisation Executive Board. *Rheumatic fever and rheumatic heart disease: Director-General report. Seventy-first WHA A71/25.* Geneva 2018.
20. Belton S, Kruske S, Jackson Pulver L, et al. Rheumatic heart disease in pregnancy: How can health services adapt to the needs of Indigenous women? A qualitative study. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 2018;58(4):425-431.
21. Belton S, Sherwood J, Peek MJ, Vaughan G, Sullivan EA. Yarning about 'that heart problem': RHD in pregnancy. *O&G Magazine.* 2016;18(4):65-67.
22. Wyber R, Katzenellenbogen JM, Pearson G, Gannon M. The rationale for action to end new cases of rheumatic heart disease in Australia. *Medical Journal of Australia.* 2017;207(8):322-323.
23. Katzenellenbogen JM, Ralph AP, Wyber R, Carapetis JR. Rheumatic heart disease: infectious disease origin, chronic care approach. *BMC Health Serv Res.* 2017;17(1):793.
24. United Nations General Assembly. *Global indicator framework for the Sustainable Development Goals and targets of the 2030 Agenda for Sustainable Development.* UN; 2019.
25. Hooker C. History and Social Change in Health and Medicine. In: Bourgeault I, Dingwall R, De Vries R, eds. *The SAGE Handbook of Qualitative Methods in Health Research*. London: SAGE Publications Ltd; 2010.
26. Katz AM, Katz, Phyllis B. Diseases of the heart in the works of Hippocrates. *British Heart Journal.* 1962;24(3):257-264.
27. Traill TA. Valvular Heart Disease and Pregnancy. *Cardiology Clinics.* 2012;30(3):369-381.
28. McKenzie J. *Heart disease and pregnancy.* London: Oxford Medical Publications; 1921.
29. Macdonald A. *The bearings of chronic disease of the heart upon pregnancy, partuition and childbed.* London UK: J&A Churchill; 1878.
30. Mackness G. Heart disease in pregnancy and labour. In: *The Transactions of the Edinburgh Obstetrical Society, Session 1889-1890.* Vol XV. Edinburgh: Oliver and Boyd; 1890.
31. The Criteria Committee of the New York Heart Association. 1994 Revisions to Classification of Functional Capacity and Objective Assessment of Patients With Diseases of the Heart: NYHA Functional class. In: Dolgin M, ed. *Nomenclature and Criteria for Diagnosis of Diseases of the Heart and Great Vessels.* 9th ed. Boston, Mass: Little, Brown & Co; 1994:253-256.
32. Jensen J. *The Heart in Pregnancy.* London: H. Kimpton; 1938.
33. Haig D, Gilchrist AR. Heart disease complicated by pregnancy. *Trans Edin Obst Soc.* 1949.
34. Bunim J, Appel S. A principle for determining prognosis of pregnancy in rheumatic heart disease. *JAMA.* 1950;142.
35. Sutherland AM, Mair JT, Bruce DF. The management of pregnancy associated with cardiac disease. *Glasgow Medical Journal.* 1955;36(12):411.
36. Oram S. Pregnancy and rheumatic heart disease. *Postgraduate Medical Journal.* 1956;32(364):76-84.

37. Hamilton B, Thomson K. *The heart in pregnancy and the childbearing age.* Boston: Little Brown and Co; 1941.
38. Gilchrist A, Murray-Lyon R. Does pregnancy hasten the fatal termination in rheumatic heart disease. *Edinburgh Medical Journal.* 1933;40(12):587.
39. Hamilton BE. Report from the cardiac clinic of the Boston Lying-in Hospital for the first twenty-five years. *American Heart Journal.* 1947;33(5):663-668.
40. Henderson DN. Pregnancy complicated by rheumatic heart disease. *Canadian Medical Association Journal.* 1936;35(4):394-398.
41. McIlroy DL, Rendel O. The Problem of the Damaged Heart In Obstetrical Practice. *BJOG: An International Journal of Obstetrics & Gynaecology.* 1931;38(1):7-7.
42. O'Driscoll MK, Coyle CF, Drury MI. Rheumatic heart disease complicating pregnancy. The remote prospects. *British Medical Journal.* 1962;2(5307):767-768.
43. Szekely P, Turner R, Snaith L. Pregnancy and the changing pattern of rheumatic heart disease. *British Heart Journal.* 1973;35(12):1293-1303.
44. Swift H. Features which suggest public health consideration of rheumatic fever. *Bulletin of the New York Academy of Medicine.* 1940.
45. Gordis L, Lilienfeld A, Rodriguez R. Studies in the epidemiology and preventability of rheumatic fever--II: Socio-economic factors and the incidence of acute attacks. *Journal of Chronic Diseases.* 1969;21(9-10):655-666.
46. Rutstein DD. Need for a public health program in rheumatic fever and rheumatic heart disease. *American Journal of Public Health* 1946;36(5):461-467.
47. Rutstein DD. The war against rheumatic fever; the nation's greatest child killer. *National Council on Rehabilitation Newsletter.* 1947;4(3):3-5.
48. Glover AJ. War-time decline of acute rheumatism. *The Lancet.* 1943;242(6254):51-52.
49. Rutstein DD. Role of the cardiac clinic in the rheumatic program. *JAMA.* 1944;126(8).
50. Gordis L. The virtual disappearance of rheumatic fever in the United States: lessons in the rise and fall of disease. *Circulation.* 1985;72(6):1155-1162.
51. Quinn RW. Comprehensive review of morbidity and mortality trends for rheumatic fever, streptococcal disease, and scarlet fever: the decline of rheumatic fever. *Reviews of Infectious Diseases.* 1989;11(6):928-953.
52. Abajobir AA, Abbafati C, Abbas KM, et al. Global, regional, and national age-sex specific mortality for 264 causes of death, 1980-2016: a systematic analysis for the Global Burden of Disease Study 2016. *The Lancet.* 2017;390(10100):1151-1210.
53. Carapetis JR, and Currie, B.J. Preventing rheumatic heart disease in Australia. *Medical Journal of Australia.* 1998;168: 428-429.
54. Besterman E. The changing face of acute rheumatic fever. *British Heart Journal.* 1970;32(5):579-582.
55. Parnaby MG, Carapetis, J.R. Rheumatic fever in Indigenous Australian children. *Journal of Paediatrics and Child Health.* 2010;46(9):527-533.
56. Hibbard LT. Maternal mortality due to cardiac disease. *Clinical Obstetrics and Gynecology.* 1975;18(3):27-36.
57. McFaul PB, Dornan JC, Lamki H, Boyle D. Pregnancy complicated by maternal heart disease. A review of 519 women. *BJOG: An International Journal of Obstetrics & Gynaecology.* 1988;95(9):861-867.
58. Sachs BP, Brown DA, Driscoll SG, et al. Hemorrhage, infection, toxemia, and cardiac disease, 1954-85: causes for their declining role in maternal mortality. *American Journal of Public Health.* 1988;78(6):671-675.
59. Negus RM. Rheumatic fever in Western Fiji: the female preponderance. *Medical Journal of Australia.* 1971;2(5):251-254.

60. World Bank Country and Lending Groups. Country classification. 2018; https://datahelpdesk.worldbank.org/knowledgebase/articles/906519-world-bank-country-and-lending-groups.
61. World Bank. The World Bank In South Africa. 2019; https://www.worldbank.org/en/country/southafrica/overview.
62. Bergsma L, Delgado S, Kizer E. *Navajo County Community Health Status Assessment.* Navajo Public Health Services District and the University of Arizona Center for Rural Health; 2012.
63. Golnick C, Asay E, Provost E, et al. Innovative primary care delivery in rural Alaska: a review of patient encounters seen by community health aides. *International Journal of Circumpolar Health.* 2012;71(1):18543.
64. Australian Institute of Health and Welfare (AIHW). *Impact and causes of illness and death in Aboriginal and Torres Strait Islander people 2011.* Canberra, Australia: AIHW; 2016.
65. Wilson N. Rheumatic heart disease in Indigenous populations--New Zealand experience. *Heart, Lung and Circulation.* 2010;19:282-288.
66. Slocumb JC, Kunitz SJ. Factors affecting maternal mortality and morbidity among American Indians. *Public Health Reports.* 1977;92(4):349.
67. Bocking N, Matsumoto C-L, Loewen K, et al. High Incidence of invasive Group A streptococcal infections in remote Indigenous communities in Northwestern Ontario, Canada. *Open Forum Infectious Diseases.* 2016;4(1):ofw243-ofw243.
68. Wyber R, Johnson T, Perkins S, et al. *Tools for Implementing RHD Control Programmes (TIPS) Handbook, 2nd edition.* Geneva, Switzerland: RHD Action; 2018.
69. Zühlke L, Karthikeyan G, Engel ME, et al. Clinical outcomes in 3343 children and adults with rheumatic heart disease from 14 low- and middle-income countries: two-year follow-up of the global Rheumatic Heart Disease Registry (the REMEDY Study). *Circulation.* 2016;134(19):1456-1466.
70. Carapetis J, Steer, AC., Mulholland, EK, Weber, M. The global burden of group A streptococcal diseases. *Lancet Infect Dis.* 2005;2005(5):685–694.
71. Watkins D, Zühlke L, Engel M, Mayosi B. Rheumatic fever: neglected again. *Science.* 2009;324(5923):37.
72. Zühlke LJ, Steer AC. Estimates of the global burden of rheumatic heart disease. *Global Heart.* 2013;8(3):189-195.
73. Reményi B, Wilson N, A S, et al. World Heart Federation criteria for echocardiographic diagnosis of rheumatic heart disease—an evidence-based guideline. *Nat Rev Cardiol.* 2012;9:297–309.
74. Remenyi B, Carapetis J, Wyber R, Taubert K, Mayosi BM. Position statement of the World Heart Federation on the prevention and control of rheumatic heart disease. *Nat Rev Cardiol.* 2013;10(5):284-292.
75. Roberts K, Colquhoun S, Steer A, Remenyi B, Carapetis J. Screening for rheumatic heart disease: current approaches and controversies. *Nat Rev Cardiol.* 2013;10(1):49-58.
76. Marijon E, Mirabel M, Celermajer DS, Jouven X. Rheumatic heart disease. *The Lancet.* 2012;379(9819):953-964.
77. Marijon E, Ou P, Celermajer DS, et al. Prevalence of rheumatic heart disease detected by echocardiographic screening. *New England Journal of Medicine.* 2007;357(5):470-476.
78. Wilson N. Cardiology and echocardiography. Inaugural RHD National Advisory Workshop: A Shared Challenge without Borders; 2010; Darwin Australia.
79. Saxena A, Zühlke L, Wilson N. Echocardiographic screening for rheumatic heart disease. *Global Heart.* 2013;8.

80. World Heart Federation (WHF). Rheumatic heart disease: neglected disease of poverty. 2010; http://www.world-heart-federation.org/what-we-do/rheumatic-heart-disease/.
81. Watkins D, Daskalakis A. The economic impact of rheumatic heart disease in developing countries. *The Lancet Global Health.* 2015;3, Supplement 1:S37.
82. Schoon M. Severe morbidity and mortality associated with cardiac disease during pregnancy in the Free State Public Health Service. *PhD, University of the Orange Free State.* 2001.
83. Australian Institute of Health and Welfare (AIHW). *Rheumatic heart disease and acute rheumatic fever in Australia: 1996–2012.* Canberra: Australian Institute of Health and Welfare; 2013.
84. Sani MU, Karaye KM, Borodo MM. Prevalence and pattern of rheumatic heart disease in the Nigerian savannah : an echocardiographic study. Cardiovascular topics. *Cardiovascular Journal of Africa.* 2007;18(5):295-299.
85. Sriha Belguith A, Koubaa Abdelkafi A, El Mhamdi S, et al. Rheumatic heart disease in a developing country: Incidence and trend (Monastir; Tunisia: 2000–2013). *International Journal of Cardiology.* 2017;228:628-632.
86. Sandhu AT, G K, Bolger A, Okello E, Kazi DS. Clinical and economic burden of rheumatic heart disease in low-income nations: Estimating the cost-of-illness in India and Uganda. *Circulation.* 2014;130.
87. Otto H, Saether SG, Banteyrga L, Haugen BO, Skjaerpe T. High prevalence of subclinical rheumatic heart disease in pregnant women in a developing country: an echocardiographic study. *Echocardiography.* 2011;28(10):1049-1053.
88. Beaton A, Okello E, Scheel A, et al. Impact of heart disease on maternal, fetal and neonatal outcomes in a low-resource setting. *Heart.* 2018.
89. Steer AC. Historical aspects of rheumatic fever. *Journal of Paediatrics and Child Health.* 2015;51(1):21-27.
90. Stanhope JM. New Zealand trends in rheumatic fever: 1885-1971. *N Z Med J.* 1975;82(551):297-299.
91. Australian Bureau of Statistics (ABS). *Mortality and Morbidity: Cardiovascular disease: 20th century trends.* Canberra, Australia: ABS; 2002.
92. Maddox K. Some clinical aspects of rheumatic fever. *Medical Journal of Australia.* 1937;1(12):375-377.
93. Clarke JT. Geographical distribution of rheumatic fever. *British Medical Journal.* 1930;1(3615):760-760.
94. Attwood B, Markus A. *The 1967 Referendum or when Aborigines didn't get the vote.* Canberra, Australia 1997.
95. Edmondson KW. Morbidity in the Australian housewife. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 1969;9(3):175-182.
96. Somers K. Cardiology: Africa versus Australia. *Medical Journal of Australia.* 1976;1(3):60-63.
97. Patten BR. Rheumatic fever in the West Kimberley. *Medical Journal of Australia.* 1981;1(2 Suppl):11-15.
98. Australian Institute of Health and Welfare (AIHW). *Cardiovascular disease: Australian facts 2011.* Canberra ACT, Australia: Australian Institute of Health and Welfare; 2011.
99. Marks L. Arnhem Land children suffer world's highest known rates of rheumatic heart disease. *Australian Broadcasting Commission.* 2018.
100. Francis J, Fairhurst H, Hardefeldt H, et al. Echocardiographic screening detects extremely high prevalence of RHD in Australia. *Heart, Lung and Circulation.* 2019;28:S51.

101. Australian Institute of Health and Welfare (AIHW). *Australian Burden of Disease Study: Impact and causes of illness and death in Aboriginal and Torres Strait Islander people 2011.* Canberra, Australia: AIHW; 2016.
102. Davis K, Remenyi B, Draper AD, et al. Rheumatic heart disease in Timor-Leste school students: an echocardiography-based prevalence study. *Medical Journal of Australia.* 2018;208(7):303-307.
103. Shearman RP. Trends in Maternal Mortality in Australia: Relevance in Current Practice. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 1990;30(1):15-17.
104. Walters WAW. Cardiovascular disease contributing to maternal mortality in Victoria. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 1969;9(1):1-6.
105. Sullivan EA, Ford JB, Chambers G, Slaytor EK. Maternal mortality in Australia, 1973–1996. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 2004;44(5):452-457.
106. Xu F, Sullivan E, Madden R, Black D, Jackson Pulver L. Improvement of maternal Aboriginality in NSW birth data. *BMC medical research methodology.* 2012;12:8.
107. Australian Institute of Health and Welfare (AIHW). *National best practice guidelines for collecting Indigenous status in health data sets.* Canberra ACT, Australia: Australian Institute of Health and Welfare; 2010.
108. Australian Institute of Health and Welfare (AIHW). *Maternal deaths in Australia 2012–2014. Cat. no. PER 92.* Canberra, Australia: AIHW; 2017.
109. Australian Institute of Health and Welfare (AIHW). *Maternal deaths in Australia 2016.* Canberra, Australia: AIHW; 2018.
110. Australasian Maternity Outcomes Surveillance System (AMOSS). AMOSS Publications. 2018; http://www.amoss.com.au/publications, 2018.
111. Hanna J, O'Rourke S. Rheumatic fever and chronic rheumatic heart disease in Yarrabah Aboriginal community, North Queensland. *Medical Journal of Australia.* 1993;158(11):793.
112. Neilson G, Streatfield RW, West M, Johnson S, Glavin W, Baird S. Rheumatic fever and chronic rheumatic heart disease in Yarrabah aboriginal community, north Queensland. Establishment of a prophylactic program. *Medical Journal of Australia.* 1993;158(5):316-318.
113. Freund S. Rheumatic heart disease program, in Northern Territory Communicable Diseases Bulletin, vol 4, no 4 Dec. In. Darwin: Centre for Disease Control, NT; 1997.
114. RHDAustralia. RHDAustralia. 2019; http://www.rhdaustralia.org.au. Accessed 30/7/2019, 2019.
115. NSW Health. Acute rheumatic fever and rheumatic heart disease control guideline. 2018; http://www.health.nsw.gov.au/Infectious/controlguideline/Pages/rheumatic-heart-disease.aspx. Accessed 12/6/2018.
116. Heart Foundation of New Zealand. *New Zealand Guidelines for Rheumatic Fever: Diagnosis, Management and Secondary Prevention of Acute Rheumatic Fever and Rheumatic Heart Disease: 2014 Update.* 2014.
117. CSANZ and HF. *New Zealand Guidelines for Rheumatic Fever 1. Diagnosis, Management and Secondary Prevention.* Auckland, New Zealand: The National Heart Foundation of New Zealand and The Cardiac Society of Australia and New Zealand; 2006.
118. Noonan S. Personal communication. In:2011.

119. RHDAustralia. RHDAustralia Champions4change program. 2019; https://www.rhdaustralia.org.au/news/champions4change-sharing-caring-and-inspiring.
120. RHDAustralia. Sharing the heartbeat: short films about RHD and pregnancy. 2017; https://www.rhdaustralia.org.au/resources/sharing-heartbeat-short-film-about-rhd-pregnancy-kriol.
121. RHDAustralia. RHD Midwifery Working Group. In: Menzies School of Health Research; 2019.
122. Marmot M. Inclusion health: addressing the causes of the causes. *The Lancet.* 2017;391(10117):186-188.
123. Bhaumik S. Doctors call for countries to step up the fight against rheumatic heart disease. *BMJ.* 2013;346.
124. Watkins D, Zühlke L, Engel M, et al. Seven key actions to eradicate rheumatic heart disease in Africa: the Addis Ababa communique. *Cardiovascular journal of Africa.* 2016;27(3):184-187.
125. Maurice J. Rheumatic heart disease back in the limelight. *The Lancet.* 2013;382(9898):1085-1086.
126. Australian Medical Association (AMA). *AMA Report Card on Indigenous Health: a call to action to prevent new cases of rheumatic heart disease in Indigenous Australia by 2031.* Canberra: AMA; 2016.
127. Ralph Anna P, de Dassel Jessica L, Kirby A, et al. Improving delivery of secondary prophylaxis for rheumatic heart disease in a high-burden setting: outcome of a stepped-wedge, community, randomized trial. *Journal of the American Heart Association.* 2018;7(14):e009308.
128. de Dassel JL, Ralph AP, Carapetis JR. Controlling acute rheumatic fever and rheumatic heart disease in developing countries: are we getting closer? *Current opinion in pediatrics.* 2015;27(1):116-123.
129. Vekemans J, Gouvea-Reis F, Kim JH, et al. The Path to Group A Streptococcus Vaccines: World Health Organization Research and Development Technology Roadmap and Preferred Product Characteristics. 2019.
130. Hunt G, Wyatt K. Media release: $35 Million for Vaccine to End Rheumatic Heart Disease. In:2019.
131. Watkins DA, Beaton AZ, Carapetis JR, et al. Rheumatic heart disease worldwide: JACC Scientific Expert Panel. *Journal of the American College of Cardiology.* 2018;72(12):1397-1416.
132. Chang A, Nabbaale J, Nalubwama H, et al. Motivations of women in Uganda living with rheumatic heart disease: A mixed methods study of experiences in stigma, childbearing, anticoagulation, and contraception. *PLOSOne.* 2018;13(3).
133. Read C, Mitchell Alison G, de Dassel Jessica L, et al. Qualitative evaluation of a complex intervention to improve rheumatic heart disease secondary prophylaxis. *Journal of the American Heart Association.* 2018;7(14):e009376.
134. Saxena A. Task shifting rheumatic heart disease screening to non-experts. *The Lancet Global Health.* 2016;4(6):e349-e350.
135. Abajobir AA, Abate KH, Abbafati C, et al. Measuring progress and projecting attainment on the basis of past trends of the health-related Sustainable Development Goals in 188 countries: an analysis from the Global Burden of Disease Study 2016. *The Lancet.* 2017;390(10100):1423-1459.
136. Walsh W. Cardiovascular health in Indigenous Australians: a call for action. *Med J Aust.* 2001;175(7):351-353.
137. Walsh WF. Medical Management of Chronic Rheumatic Heart Disease. *Heart, Lung and Circulation.* 2010;19(5–6):289-294.

138. Noonan S, Zurynski Y, Currie B, et al. A national prospective surveillance study of acute rheumatic fever in Australian children. *Pediatric Infectious Disease Journal.* 2013;32(1):e26-e32.
139. Lawrence JG, Carapetis JR, Griffiths K, Edwards K, Condon JR. Acute rheumatic fever and rheumatic heart disease: incidence and progression in the Northern Territory of Australia, 1997 to 2010. *Circulation.* 2013;128(0009-7322):492-501.
140. Brown A, Tonkin, A., White, H., Riddell, T., Brieger, D., Walsh, W., Zeitz, C., Jeremy, R., Kritharides, L. The Cardiac Society inaugural cardiovascular health conference: findings and ways forward. *Heart Lung Circulation.* 2010;10(19):264-268.
141. White H, Walsh W, Brown A, et al. Rheumatic heart disease in Indigenous populations. *Heart, Lung and Circulation.* 2010.
142. Kritharides L, Brown A, Brieger D, et al. Recommendations arising from the Inaugural CSANZ Conference on Indigenous Cardiovascular Health. *Heart, Lung and Circulation.* 2010;19(5–6):269-272.
143. National Health and Medical Research Council (NHMRC). Road Map II: A strategic framework for improving the health of Aboriginal and Torres Strait Islander people through research. In. Canberra, ACT: NHMRC Publications; 2010.
144. National Health and Medical Research Council (NHMRC). Values and Ethics: Guidelines for Ethical Conduct in Aboriginal and Torres Strait Islander Health Research. In. Canberra, ACT: NHMRC Publications; 2003.
145. Homer CSE, Biggs, J., Vaughan, G., Sullivan, E.A. Mapping maternity services in Australia: location, classification and services. *Australian Health Review.* 2011;35(2):222-229.
146. Kruske S, Kildea, S., Barclay, L. Cultural safety and maternity care for Aboriginal and Torres Strait Islander Australians. *Women and Birth.* 2006;19(3):59-88.
147. King J, Sullivan, E. *Maternal deaths in Australia 2000-2002.* Canberra: Australian Institute of Health and Welfare; 2006.
148. Laws P, Sullivan E. *Report on the evaluation of the Perinatal National Minimum Data Set.* Sydney: AIHW National Perinatal Statistics Unit; 2004.
149. Metcalfe A. *Maternal morbidity data in Australia: an assessment of the feasibility of standardised collection.* Canberra: AIHW; 2012.
150. Pollock WE, Sullivan, E., Nelson, S., King, J.,. Capacity to monitor severe maternal morbidity in Australia. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 2008;48(1).
151. Walker J. *Maternity data in Australia: a review of sources and gaps.* Canberra ACT, Australia: Australian Institute of Health and Welfare 2011. Bulletin 87.
152. Sullivan E, Hall, B., King, JF.,. *Maternal deaths in Australia 2003-2005.* Sydney: AIHW National Perinatal Statistics Unit; 2008. Cat. no. PER 42.
153. Boerm T, AbouZhar C, Bos E, Hansen P, Addai E, Low-Beer D. *Monitoring and evaluation of health systems strengthening.* Geneva: WHO, World Bank, GAVI, Global Fund; 2010.
154. Gissler M, Mohangoo A, Blondel B, et al. Perinatal health monitoring in Europe: results from the EURO-PERISTAT project. *Informatics for Health and Social Care.* 2010;35:2(1753-8165 (Electronic)):64-79.
155. Kildea S, Leap N. Risk and childbirth in rural and remote Australia. National Rural Health Conference; 2003; Hobart, Australia.
156. Vaughan G, McLintock C, Sherwood J, et al. Challenges of Surveillance: Rheumatic Heart Disease (RHD) in Pregnancy. *Heart, lung & circulation.* 2013;22:S251.
157. Tibby D, Corpus R, Walters DL. Establishment of an innovative specialist cardiac Indigenous outreach service in rural and remote Queensland. *Heart, Lung and Circulation.* 2010;19(5–6):361-366.

158. Varghese S, Corkill W. Cardiac Coordination in Central Australia. *Heart, Lung and Circulation.* 2019;28:S59.
159. Huffman MD, Galloway JM. Cardiovascular Health in Indigenous Communities: Successful Programs. *Heart, Lung and Circulation.* 2010;19(5–6):351-360.
160. Hayman N. Strategies to Improve Indigenous Access for Urban and Regional Populations to Health Services. *Heart, Lung and Circulation.* 2010;19(5–6):367-371.
161. Jeremy R, Tonkin A, White H, et al. Improving cardiovascular care for Indigenous populations. *Heart, Lung and Circulation.* 2010;19(5–6):344-350.
162. Josif CM, Barclay L, Kruske S, Kildea S. 'No more strangers': Investigating the experiences of women, midwives and others during the establishment of a new model of maternity care for remote dwelling aboriginal women in northern Australia. *Midwifery.* 2014;30(March):317-323.
163. Maguire G, Nelson C. Acute rheumatic fever and rheumatic heart disease: an insight into Aboriginal health disadvantage and remote Australia. *Med J Aust.* 2006;184(10).
164. Kildea S, Kruske S, Barclay L, Tracy S. 'Closing the Gap': how maternity services can contribute to reducing poor maternal infant health outcomes for Aboriginal and Torres Strait Islander women. *Rural and remote health.* 2010;10(3):1383.
165. Jackson-Pulver L. *An argument on culture safety in health service delivery: towards better health outcomes for Aboriginal peoples* [Unpublished]. Sydney: School of Public Health, The University of Sydney, University of Sydney; 2003.
166. Wilson G. *What do Aboriginal women think is good antenatal care?* Alice Springs: Cooperative Research Centre for Aboriginal Health (CRCAH); 2009.
167. Lawrence M, with Dodd, Z., Mohor, S., Dunn, S., de Crespigny, C., Power, C. & MacKean, L.,. *Improving the patient journey: achieving positive outcomes for remote Aboriginal cardiac patients.* Darwin: Cooperative Research Centre for Aboriginal Health; 2009.
168. Kildea S, Hickey S, Nelson C, et al. Birthing on Country (in Our Community): a case study of engaging stakeholders and developing a best-practice Indigenous maternity service in an urban setting. *Australian Health Review.* 2018;42(2):230-238.
169. CATSINaM, ACM, CRANAplus. Birthing on Country Position Statement. 2017; http://catsinam.org.au/static/uploads/files/birthing-on-country-position-statement-endorsed-march-2016-wfaxpyhvmxrw.pdf. Accessed 24/9/2019.
170. Kildea S, Gao Y, Hickey S, et al. Reducing preterm birth amongst Aboriginal and Torres Strait Islander babies: A prospective cohort study, Brisbane, Australia. *EClinical Medicine, published by The Lancet.* 2019;12:43-51.
171. Harrington Z, Thomas, D.P., Currie, B.J.,, Bulkanhawuy J. Challenging perceptions of non-compliance with rheumatic fever prophylaxis in a remote Aboriginal community. *Med J Aust.* 2006;184(10).
172. Davidson P, Abott, P., Davison, J., DiGiacomo, M. Improving medication uptake in Aboriginal and Torres Strait Islander Peoples. *Heart, Lung and Circulation.* 2010;19:372-377.
173. Nappaljari Jone J. Birthing: Aboriginal Women. National Native Title Conference 2011; 1 June 2011, 2011; Brisbane.
174. Couchie C, Sanderson S. A Report on Best Practices for Returning Birth to Rural and Remote Aboriginal Communities. *J Obstet Gynaecol Can.* 2007;29(3):250–254.
175. Kildea S. *Birthing business in the bush: it's time to listen*: Centre for Family Health and Midwifery, University of Technology; 2005.
176. Arnold J, De Costa C, Howat PW. Timing of transfer for pregnant women from Queensland Cape York communities to Cairns for birthing. *Med J Aust.* 2009;190:3.

177. Van Mook W, Peeters, L. Severe cardiac disease in pregnancy, part II: impact of congenital and acquired cardiac diseases during pregnancy. *Current Opinion in Critical Care.* 2005;11(5).
178. Douglas VK. Childbirth among the Canadian Inuit: A review of the clinical and cultural literature. *International Journal of Circumpolar Health.* 2012;65.
179. National Aboriginal Health Organisation (NAHO). *Exploring models for quality maternity care in First Nations and Inuit communities: a preliminary needs assessment.* Ottawa, Canada: National Aboriginal Health Organisation; 2006.
180. Hodnett Ellen D, Downe S, Walsh D. Alternative versus conventional institutional settings for birth. In. *Cochrane Database of Systematic Reviews*: John Wiley & Sons, Ltd; 2012.
181. Hodnett Ellen D. Continuity of caregivers for care during pregnancy and childbirth. In. *Cochrane Database of Systematic Reviews*: John Wiley & Sons, Ltd; 2008.
182. Van Wagner V, Epoo B, Nastapoka J, Harney E. Reclaiming Birth, Health, and Community: Midwifery in the Inuit Villages of Nunavik, Canada. *The Journal of Midwifery & Women's Health.* 2007;52(4):384-391.
183. WHO. Ottawa Charter for Health Promotion, 1986. Paper presented at: First International Conference on Health Promotion, Ottawa, Canada, 17–21 November 19861986; Ottawa.
184. National Health and Medical Research Council (NHMRC). *Better informed health care through better clinical guidelines: An NHMRC Draft Discussion Paper.* Canberra 2015.
185. Cochrane Australia. *Living Evidence for Australian health care.* Melbourne 2018.
186. AIHW. *Women and heart disease: Cardiovascular profile of women in Australia.* AIHW and Heart Foundation; 2010.
187. Brennand J, Northridge R, Scott H, Walker N, on behalf of the Royal College of Physicians and Surgeons of Glasgow. *Addressing the heart of the issue: Standards of good clinical practice in the shared obstetric and cardiology care of women of childbearing age.* Glasgow: RCPS Glasgow; 2016.
188. Elkayam U, Bitar F. Valvular heart disease and pregnancy: Part II: prosthetic valves. *Journal of the American College of Cardiology.* 2005;46(3):403-410.
189. National Health and Medical Research Council (NHMRC). Australian Clinical Practice Guidelines. In. Canberra, ACT: NHMRC Publications; 2018.
190. Reiner Ž. Guidelines, position papers and critical reviews: differences and similarities. *European Journal of Preventive Cardiology.* 2012;20(1):3-5.
191. Australian Commission on Safety and Quality in Health Care. Overview of the Clinical Care Standards. 2018; https://www.safetyandquality.gov.au/our-work/clinical-care-standards/overview-of-the-clinical-care-standards/. Accessed 8/8/2018, 2018.
192. Brown A, O'Shea R, Mott K, McBride K, Lawson T, Jennings G. Essential Service Standards for Equitable National Cardiovascular Care for Aboriginal and Torres Strait Islander People. *Heart Lung Circ.* 2015;24(Feb (2)).
193. Nishimura RA, Otto CM, Bonow RO, et al. 2014 AHA/ACC guideline for the management of patients with valvular heart disease: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines. *The Journal of thoracic and cardiovascular surgery.* 2014;148(1):e1-e132.
194. Nishimura R, Otto C, Bonow RO, et al. 2017 AHA/ACC Focused Update of the 2014 AHA/ACC Guideline for the Management of Patients With Valvular Heart Disease: A Report of the American College of Cardiology/American Heart Association Task Force on Clinical Practice Guidelines. 2017(1524-4539 (Electronic)).

195. Noonan S, Wade V. Integrating Cultural Safety Into Disease Management Guidelines. *Heart, Lung and Circulation.* 2019;28:S48-S49.
196. Regitz-Zagrosek V, Roos-Hesselink J, Bauersachs J, et al. 2018 ESC Guidelines for the management of cardiovascular diseases during pregnancy. *Eur Heart J.* 2018;39(34).
197. Royal College of Obstetricians and Gynaecologists (RCOG). *Cardiac Disease and Pregnancy.* UK: RCOG; 2011.
198. Royal Australia and New Zealand College of Obstetrics and Gynaecology (RANZCOG). College Statements & Guidelines. 2018; https://www.ranzcog.edu.au/Statements-Guidelines. Accessed 8/8/2018, 2018.
199. Palafox B, Mocumbi AO, Kumar RK, et al. The WHF Roadmap for Reducing CV Morbidity and Mortality Through Prevention and Control of RHD. *Global Heart.* 2017;12(1):47-62.
200. Department of Health. Clinical Practice Guidelines: Pregnancy Care. In. Canberra: Australian Government; 2018.
201. World Health Organization (WHO). *WHO recommendations on antenatal care for a positive pregnancy experience.* Geneva 2016.
202. Elkayam U, Goland S, Pieper P, Silversides C. High-risk cardiac disease in pregnancy Part I. *J Am Coll Cardiol.* 2016;68(Jul 26(4)):396-410.
203. Mocumbi AO, Sliwa K. Women's cardiovascular health in Africa. *Heart.* 2012;98(6):450-455.
204. Essop MR, Nkomo VT. Rheumatic and nonrheumatic valvular heart disease: Epidemiology, management, and prevention in Africa. *Circulation.* 2005;112(23):3584-3591.
205. Anthony J, Osman A, Sani MU. Valvular heart disease in pregnancy. *Cardiovascular Journal of Africa.* 2016;27(2):111-118.
206. RHD Action; School of Medicine CWRU. *Bridging the treatment gap for RHD in Sub-Saharan Africa: a Uganda—Case Western Reserve University Partnership.* 2018.
207. Zühlke L, Acquah L. Pre-conception counselling for key cardiovascular conditions in Africa: optimising pregnancy outcomes. *Cardiovasc J Afr.* 2016;27(2):79-83.
208. Marijon E, Ou, P., Celermajer, DS., Ferreira, B., Mocumbi, AO., Sidi, D., Jouven, X. Echocardiographic screening for rheumatic heart disease. *Bulletin of the World Health Organization (BLT)* 2008; http://www.who.int/bulletin/volumes/86/2/07-046680/en/.
209. Ilton M. The challenge and controversies of Indigenous rheumatic heart disease: the NT experience. Chronic Disease Network Conference 2008 - Health at the Heart of Australia 200; 2008; Alice Springs.
210. Zoellner J, Curry R, Johnson M. The contribution of heart disease to maternal mortality. *Current Opinion in Obstetrics & Gynecology.* 2013;25(2):91-97.
211. Hameed A KI, Tummala PP, Wani OR, Canetti M, Akhter MW, Goodwin I, Zapadinsky N, Elkayam U. The effect of valvular heart disease on maternal and fetal outcome of pregnancy. *Journal of the American College of Cardiology.* 2001;37(3).
212. Siu S, Sermer, M., Colman, J, Alvarez, N., Merier, L-A., Morton, BC., Kells, CM., Bergin, L., Kiess, M., Marchotte, F., Taylor, D., Gordon, EP., Spears, JC, Tam, J., Amankwah, K., Smallhorn, JF., Farine, D., Sorensen, S. Prospective multicenter study of pregnancy outcomes in women with heart disease. *Circulation.* 2001;104:515-521.
213. McLintock C. Anticoagulant therapy in pregnant women with mechanical prosthetic heart valves: no easy option. *Thrombosis Research.* 2011;127(S3):S56-S60.
214. Freire C, Ribeiro A. Cardiac disease and maternity in Africa. *Heart.* 2014;100(24):1901-1902.

215. Warnes CA, Williams RG, Bashore TM, et al. ACC/AHA 2008 Guidelines for the Management of Adults With Congenital Heart Disease: Executive Summary. *Circulation.* 2008;118(23):2395.
216. Hays L. Transition to Adult Congenital Heart Disease Care: A Review. *Journal of Pediatric Nursing.* 2015;30(5):e63-e69.
217. Hameed A, Morton C, Moore A. *Improving health care response to cardiovascular disease in pregnancy and postpartum.* Developed under contract #11-10006 with the California Department of Public Health, Maternal, Child and Adolescent Health Division; 2017.
218. Sliwa K, Johnson M, Zilla P, Roos-Hesselink J. Management of valvular disease in pregnancy: a global perspective. *European Heart Journal.* 2015;36(18):1078-1089.
219. Knight M, Nair M, Tuffnell D, et al. *Saving Lives, Improving Mothers' Care - Surveillance of maternal deaths in the UK 2012-14 and lessons learned to inform maternity care from the UK and Ireland Confidential Enquiries into Maternal Deaths and Morbidity 2009-14.* NPEU, UOxford; 2016.
220. European Society of Cardiology. *Expert consensus document on management of cardiovascular diseases during pregnancy.* London UK: ESC; 2003.
221. Sliwa K, Libhaber E, Elliott C, et al. Spectrum of cardiac disease in maternity in a low-resource cohort in South Africa. *Heart.* 2014;100(24).
222. Nqayana T, Moodley J, Naidoo D. Cardiac disease in pregnancy. *Cardiovasc J Afr.* 2008;19(3):145 - 151.
223. Hanania G. Management of anticoagulants during pregnancy. *Heart.* 2001;86(2):125-126.
224. WHO. *Rheumatic fever and rheumatic heart disease.* Geneva, Switzerland: World Health Organisation; 2004.
225. Raffaele De Caterina SH, Wallentin L, Agnelli G, et al. Anticoagulants in heart disease: current status and perspectives. *European heart journal.* 2007;28:880-913.
226. Siu SS, Colman, J. Heart disease and pregnancy. *Heart.* 2001;85(6):710-715.
227. Westhoff-Bleck M, Hilfiker-Kleiner D, Gunter HH, Schieffer E, Drexler H. Management of heart diseases in pregnancy: rheumatic and congenital heart disease, myocardial infarction and post partum cardiomyopathy. *Internist (Berl).* 2008;49(7):805-810.
228. Madazli R, Sal V, Cift T, Guralp O, Goymen A. Pregnancy outcomes in women with heart disease. *Arch Gynecol Obstet.* 2009;281(1):29 - 34.
229. Cabral Castaneda F, Karchmer S, Aguilera Perez R, Villareal Munoz E, Ruiz Anguas J. Perinatal perspective in heart disease and pregnancy: review of 1169 cases of pregnancy. *Ginecol Obstet Mex.* 1997;65.
230. Subbaiah M, Sharma V, Kumar S, et al. Heart disease in pregnancy: cardiac and obstetric outcomes. *Arch Gynecol Obstet.* 2013;288(1):23-27.
231. Bhatla N, Lal S, Behera G, et al. Cardiac disease in pregnancy. *Int J Gynaecol Obstet.* 2003;82(2):153 - 159.
232. Silversides C, Colman J, Sermer M, Siu S. Cardiac risk in pregnant women with rheumatic mitral stenosis. *Am J Cardiol.* 2003;91(11):1382 - 1385.
233. Bass E. Management of heart disease in pregnancy. *O and G Magazine.* 2008;10(3):21-24.
234. Reimold S, Rutherford, MB. Valvular Disease in Pregnancy. *New England Journal of Medicine.* 2003;349:52-59.
235. Desai D, Adanlawo M, Naidoo D, Moodley J, Kleinschmidt I. Mitral stenosis in pregnancy: a four-year experience at King Edward VIII Hospital, Durban, South Africa. *BJOG.* 2000;107(8):953 - 958.

236. Stangl V, Schad, J., Gossing, G., Borges, A., Baumann, G., Stangl, K. Maternal heart disease and pregnancy outcome: A single-centre experience. *Eur J Heart Failure.* 2008;10(9):855-860.
237. Jatavan T, Luewan S, Tongsong T. Outcomes of pregnancy complicated by heart disease at Maharaj Nakorn Chiang Mai Hospital. *Journal of the Medical Association of Thailand = Chotmaihet thangphaet.* 2011;94(10):1159-1163.
238. Sartain J. Obstetric patients with rheumatic heart disease. *O&G Magazine.* 2008;10(3):18-20.
239. Sartain J, Anderson N, Barry J, Boyd P, Howat P. Rheumatic heart disease in pregnancy: cardiac and obstetric outcomes. *Intern Med J.* 2012;42(9):978-984.
240. Regitz-Zagrosek V, Blomstrom Lundqvist C, Borghi C, et al. *ESC Guidelines on the management of cardiovascular diseases during pregnancy.* European Heart Journal; 2011.
241. van Hagen IM, Boersma E, Johnson MR, et al. Global cardiac risk assessment in the Registry Of Pregnancy And Cardiac disease: results of a registry from the European Society of Cardiology. *European Journal of Heart Failure.* 2016;18(5):523-533.
242. Drenthen W, Boersma E, Balci A, et al. Predictors of pregnancy complications in women with congenital heart disease. *European Heart Journal.* 2010;31(17):2124-2132.
243. Sliwa K. Dealing with late maternal death due to cardiovascular disease. *SA Heart.* 2015;12(3).
244. Department of Health South Africa. *Saving mothers 2014-2016: seventh triennial report on confidential enquiries into maternal deaths in South Africa:.* Pretoria, South Africa: DoH SA; 2018.
245. Naidoo DP, Shein K. The implications of HIV infection on the management of valvular heart disease in Southern Africa. *SA Heart.* 2009;6(2).
246. Fitzpatrick A, Mohammadi F, Jesudason S. Managing pregnancy in chronic kidney disease: improving outcomes for mother and baby. *International Journal of Women's Health.* 2016;8:273-285.
247. Australian Institute of Health and Welfare (AIHW). *Chronic kidney disease in Aboriginal and Torres Strait Islander people 2011.* Canberra Australia: AIHW; 2011.
248. Lindquist A, Noor N, Sullivan E, Knight M. The impact of socioeconomic position on severe maternal morbidity outcomes among women in Australia: a national case-control study. *BJOG.* 2014.
249. Lindquist A, Kurinczuk JJ, Redshaw M, Knight M. Experiences, utilisation and outcomes of maternity care in England among women from different socio-economic groups: findings from the 2010 National Maternity Survey. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2014:n/a-n/a.
250. Sliwa K, Md P, Fesc F, et al. Impact of Socioeconomic Status, Ethnicity, and Urbanization on Risk Factor Profiles of Cardiovascular Disease in Africa. *Circulation.* 2016;133(12):1199-1208.
251. Tanya Nagahawatte N, Goldenberg RL. Poverty, Maternal Health, and Adverse Pregnancy Outcomes. *Annals of the New York Academy of Sciences.* 2008;1136(1):80-85.
252. Elassy SM, Elmidany AA, Elbawab HY. Urgent cardiac surgery during pregnancy: a continuous challenge. *Ann Thorac Surg.* 2014;97(5):1624-1629.
253. Mischke K. Management of pregnant women with artificial heart valves: inconsistency in ESC publications. *European Heart Journal.* 2007;28(19):2419-2420.
254. McLintock C. Anticoagulant options in pregnancy for women with mechanical valves. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2017;124(9):1421-1421.

255. Mazibuko B, Ramnarain H, Moodley J. An audit of pregnant women with prosthetic heart valves at a tertiary hospital in South Africa: a five-year experience. *Cardiovascular Journal of Africa.* 2012;23(4).
256. Tounsi A, Abid D, Abid L, et al. Prosthetic mechanic valve thrombosis in pregnancy: a single center study of 7 cases. *European Heart Journal.* 2013;34(suppl 1).
257. D'Souza R, Silversides CK, McLintock C. Optimal anticoagulation for pregnant women with mechanical heart valves. Paper presented at: Seminars in thrombosis and hemostasis; 2016, 2016.
258. Vause S, Clarke B, Tower CL, Hay CRM, Knight M. Pregnancy outcomes in women with mechanical prosthetic heart valves: a prospective descriptive population based study using the United Kingdom Obstetric Surveillance System (UKOSS) data collection system. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2016;124(9):1411-1419.
259. Mayer KH, Anderson BL, Cu-Uvin S. Pregnancy and Optimal Care of HIV-Infected Patients. *Clinical Infectious Diseases.* 2009;48(4):449-455.
260. Pieper PG. Use of medication for cardiovascular disease during pregnancy. *Nature Reviews Cardiology.* 2015;12(12):718-729.
261. Department of Health South Africa. *Saving mothers 2008-2010: fifth report on confidential enquiries into maternal deaths in South Africa.* 2012.
262. Naidoo DP, Desai DK, Moodley J. Maternal deaths due to pre-existing cardiac disease. *Cardiovascular journal of South Africa : official journal for Southern Africa Cardiac Society [and] South African Society of Cardiac Practitioners.* 2002;13(1):17-20.
263. Duffy J, Rolph R, Gale C, et al. Core outcome sets in women's and newborn health: a systematic review. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2017;124(10):1481-1489.
264. Schaap T, Bloemenkamp K, Deneux-Tharaux C, et al. Defining definitions: a Delphi study to develop a core outcome set for conditions of severe maternal morbidity. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2019;126(3):394-401.
265. D'Souza R, Silversides C, Sermer M, et al. COSCarP study (Core outcome sets for studies on cardiac disease in pregnancy). 2018; http://www.comet-initiative.org/studies/details/834.
266. Kirkham JJ, Gorst S, Altman DG, et al. Core Outcome Set–STAndards for Reporting: The COS-STAR Statement. *PLOS Medicine.* 2016;13(10):e1002148.
267. Khan K. The CROWN Initiative: journal editors invite researchers to develop core outcomes in women's health. *BJOG: An International Journal of Obstetrics & Gynaecology.* 2014;121(10):1181-1182.
268. Geirsson RT, Eggebø T. Core outcomes for reporting women's health. *Acta Obstetricia et Gynecologica Scandinavica.* 2014;93(9):843-844.
269. Silversides CK, Grewal J, Mason J, et al. Pregnancy Outcomes in Women With Heart Disease: The CARPREG II Study. *J Am Coll Cardiol.* 2018;71(21):2419-2430.
270. Mertens D, Bledsoe K, Sullivan M, Wilson A. Utilization of Mixed Methods for Transformative Purposes. In: Tashakkori A, Teddlie C, eds. *Sage handbook of mixed methods in social & behavioral research*. 2nd ed. Thousand Oaks: SAGE Publications, Inc.; 2010:193-214.
271. Creswell JW, Plano Clark VL. Choosing a mixed methods design. In. *Designing and conducting mixed methods research.* Vol 2. Thousand Oaks: Sage Publications; 2011:53-106.
272. Seers K. What is a qualitative synthesis? *Evidence Based Nursing.* 2012;15(4):101.

273. Vaughan G, Dawson A, Peek M, Carapetis J, Sullivan E. Standardizing clinical care measures of rheumatic heart disease in pregnancy: a qualitative synthesis. *Birth: Issues in Perinatal Care.* 2019;00 (early view):1-14.
274. Bourgeault I, Dingwall R, De Vries R. The SAGE Handbook of Qualitative Methods in Health Research. In: London: SAGE Publications Ltd; 2010: http://methods.sagepub.com/book/sage-hdbk-qualitative-methods-in-health-research. Accessed 2018/01/19.
275. Slater J. Social constructionism. In: Allen M, ed. *The SAGE encyclopedia of communication research methods.* Thousand Oaks: SAGE Publications; 2017.
276. Barber RM, Fullman N, Sorensen RJD, et al. Healthcare access and Quality Index based on mortality from causes amenable to personal health care in 195 countries and territories, 1990-2015: a novel analysis from the Global Burden of Disease Study 2015. *The Lancet.* 2017;390(10091):231-266.
277. Patton MQ. *Qualitative research & evaluation methods: Integrating theory and practice (4th ed.).* California, USA: Sage; 2015.
278. Gentles SJ, Charles C, Ploeg J, McKibbon KA. Sampling in Qualitative Research: Insights from an Overview of the Methods Literature. *The Qualitative Report.* 2015;20(11).
279. Liamputtong P. *Qualitative Research Methods (4th ed).* Melbourne Vic, Australia: Oxford University Press; 2013.
280. Ayres L. Thematic Coding and Analysis. In: Given LM, ed. *The SAGE Encyclopedia of Qualitative Research Methods*. Thousand Oaks, California: SAGE Publications, Inc.; 2008.
281. Braun V, Clarke V. Using thematic analysis in psychology. *Qualitative Research in Psychology.* 2006;3(2).
282. Padgett DK. *Qualitative and Mixed Methods in Public Health.* Thousand Oaks, USA: Sage; 2012.
283. Shenton AK. Strategies for ensuring trustworthiness in qualitative research projects. *Education for Information.* 2004;22(63).
284. Robson C. Real World Research: a Resource for Social Scientists and Practitioner-Researchers. In. Vol 2nd: New York: John Wiley and Sons; 2002.
285. Grbich C. *New approaches in social research.* Sage; 2004.
286. Abouzeid M, Wyber R, La Vincente S, et al. Time to tackle rheumatic heart disease: Data needed to drive global policy dialogues. *Glob Public Health.* 2018:1-13.
287. AMHMAC. *Aboriginal and Torres Strait Islander Health Performance Framework 2017 Report.* Canberra 2017.
288. Farquhar C, McLintock C, Mahony F, et al. Rheumatic heart disease in pregnancy: a prospective New Zealand cohort: ep13. 44. *BJOG: An International Journal of Obstetrics and Gynaecology.* 2015;122:271.
289. Curtis S, Marsden-Williams J, Sullivan C, et al. Current trends in the management of heart disease in pregnancy. *Int J Cardiol.* 2009;133(1):62-69.
290. Van Hanegem N, Miltenburg AS, Zwart JJ, Bloemenkamp KWM, Van Roosmalen JOS. Severe acute maternal morbidity in asylum seekers: a two-year nationwide cohort study in the Netherlands. *Acta Obstetricia et Gynecologica Scandinavica.* 2011;90(9):1010-1016.
291. Watkins D, Sebitloane M, Engel M, Mayosi B. The burden of antenatal heart disease in South Africa: a systematic review. *BMC Cardiovasc Disor.* 2012;12(1).
292. Baumgartner H, Falk V, Bax JJ, et al. 2017 ESC/EACTS Guidelines for the management of valvular heart disease. *European Heart Journal.* 2017;38(36):2739-2791.

293. Geirsson RT, Khan K. The CROWN Initiative: journal editors invite researchers to develop core outcomes in women's health. *Acta Obstetricia et Gynecologica Scandinavica.* 2014;93(9):845-847.
294. Hsieh H-F, Shannon S. Three approaches to qualitative content analysis. *Qual Health Res.* 2005;15(9):1277-1288.
295. Schardt C, Adams MB, Owens T, Keitz S, Fontelo P. Utilization of the PICO framework to improve searching PubMed for clinical questions. *BMC medical informatics and decision making.* 2007;7(1):16.
296. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLOS Medicine.* 2009;6(7):e1000097.
297. Critical Appraisal Skills Programme (CASP). CASP Appraisal Checklists. 2014; https://casp-uk.net/casp-tools-checklists/.
298. Johanna Briggs Institute. Critical Appraisal Checklists. 2017; http://joannabriggs.org/research/critical-appraisal-tools.html.
299. Wasim T, Amer W, Majrroh A, Siddiq S. Foetomaternal outcome of pregnancy with cardiac disease. *J Pak Med Assn.* 2008;58(4).
300. Carapetis J. The stark reality of rheumatic heart disease. *Eur Heart J.* 2015;36(18):1070-1073.
301. Avila W, Rossi E, Ramires J, et al. Pregnancy in patients with heart disease: Experience with 1,000 cases. *Clin Cardiol.* 2003;26(3):135-142.
302. Barbosa P, Lopes A, Feitosa G, et al. Prognostic factors of rheumatic mitral stenosis during pregnancy and puerperium. *Arq Bras Cardiol.* 2000;75(3):215-224.
303. Chhetri S, Shrestha N, Pilgrim T. Pregnancy complicated by heart disease in Nepal. *Heart Asia.* 2014;6(1):26-29.
304. Chumpathong S, Sirithaweesit C, Pechpaisit N, et al. Predictors for complications in pregnant women with heart disease, a retrospective study. *Journal of the Medical Association of Thailand = Chotmaihet thangphaet.* 2014;97(7):730-735.
305. Fu Q, Lin J. Risk factors for heart failure during pregnancy among Chinese women with cardiac disease. *Int J Gynaecol Obstet.* 2015;130(3):266-269.
306. Michaelson-Cohen R, Elstein D, Ioscovich A, et al. Severe heart disease complicating pregnancy does not preclude a favourable pregnancy outcome: 15 years' experience in a single centre. *J Obstet Gynaecol.* 2011;31(7):597-602.
307. Puri S, Bharti A, Mohan B, Bindal V, Verma S. Maternal heart disease and pregnancy outcomes. *JK Science.* 2013;15(1):7-10.
308. Soma-Pillay P, MacDonald A, Mathivha T, Bakker J, Mackintosh M. Cardiac disease in pregnancy: a 4-year audit at Pretoria Academic Hospital. *S Afr Med J.* 2008;98(7):553-556.
309. van Hagen I, Thorne S, Taha N, et al. Pregnancy outcomes in women with rheumatic mitral valve disease. *Circulation.* 2018;137(8).
310. Asghar F, Kokab H. Evaluation and outcome of pregnancy complicated by heart disease. *J Pak Med Assn.* 2005;55(10):416-419.
311. Konar H, Chudhuri S. Pregnancy complicated by maternal heart disease: A review of 281 women. *J Obstet Gynecol India.* 2012;62(3):301-306.
312. Malhotra M, Sharma J, Tripathii R, Arora P, Arora R. Maternal and fetal outcome in valvular heart disease. *Int J Gynecol Obstet.* 2004;84(0020-7292 (Print)).
313. Ahmed N, Kausar H, Ali L, Rakhshinda. Fetomaternal outcome of pregnancy with mitral stenosis. *Pak J Med Sci.* 2015;31(3):643-647.
314. Faiz S, Al-Meshari A, Sporrong B. Pregnancy and valvular heart disease. *Saudi Med J.* 2003;24(10).

315. Schoon M, Bam R, Wolmarans L. Cardiac disease during pregnancy - a Free State perspective on maternal morbidity and mortality. *Cardiovasc J Afr.* 1997;87(1):C19 - C22.
316. Iung B. Pregnancy-related cardiac complications: A consequence of the burden of rheumatic heart disease in sub-Saharan Africa. *Archives of Cardiovascular Diseases.* 2011;104(6):367-369.
317. Pratibha D, Kiranmai D, Vemuri U, Rani V, Vani N. Pregnancy outcome in chronic rheumatic heart disease. *J Obstet Gynecol India.* 2009;59(1):41-46.
318. Martins L, Freire C, Capurucu C, Nunes M, Rezende C. Risk prediction of cardiovascular complications in pregnant women with heart disease. *Arq Bras Cardiol.* 2016;106(4):289-296.
319. Sawhney H, Aggarwal N, Suri V, Vasishta K, Sharma Y, Grover A. Maternal and perinatal outcome in rheumatic heart disease. *Int J Gynecol Obstet.* 2003;80(1):9-14.
320. Abdel-Hady E, El-Shamy M, El-Rifai A, Goda H, Abdel-Samad A, Moussa S. Maternal and perinatal outcome of pregnancies complicated by cardiac disease. *Int J Gynaecol Obstet.* 2005;90(1):21 - 25.
321. Kovavisarach E, Nualplot P. Outcome of pregnancy among parturients complicated with heart disease in Rajavithi Hospital. *Journal of the Medical Association of Thailand = Chotmaihet thangphaet.* 2007;90(11):2253-2259.
322. Rahman J, Rahman M, Al-Suleiman S, Al-Jama F. Pregnancy complicated by maternal cardiac disease: a review of 274 patients. *J Obstet Gynaecol.* 2000;20(3):242-245.
323. Bhutta S, Aziz S, Korejo R. Pregnancy following cardiac surgery. *J Pak Med Assn.* 2003;53(9):407-413.
324. Kaluarachchi A, Seneviratne H. Heart disease in pregnancy - Evaluation of disease pattern and outcome in Sri Lanka. *J Obstet Gynaecol.* 1995;15(1):9-14.
325. Kanwar R, Sharma M, Marwah S, Mittal P, Kumari N. Heart disease in pregnancy-evaluation of spectrum, association of predictors with obstetric outcome and need for comprehensive medical care. *J Clin Diagn Res.* 2018;12(1):QC20-QC24.
326. Rezk M, Elkilani O, Shaheen A, Gamal A, Badr H. Maternal hemodynamic changes and predictors of poor obstetric outcome in women with rheumatic heart disease: a five-year observational study. *The Journal of Maternal-Fetal & Neonatal Medicine.* 2018;31(12):1542-1547.
327. Thanajiraprapa T, Phupong V. Pregnancy complications in women with heart disease. *The Journal of Maternal-Fetal & Neonatal Medicine.* 2010;23(10):1200-1204.
328. Nobuyoshi M, Arita T, Shirai S-i, et al. Percutaneous balloon mitral valvuloplasty: a review. *Circulation.* 2009;119(8):e211-e219.
329. Watkins D, Johnson C, Colquhoun S, et al. Global, regional, and national burden of rheumatic heart disease, 1990–2015. *New Engl J Med.* 2017;377(8):713-722.
330. Mirabel M, Tafflet M, Noel B, et al. Newly diagnosed rheumatic heart disease among indigenous populations in the Pacific. *Heart (British Cardiac Society).* 2015;101(23):1901-1906.
331. Colquhoun S. *Global epidemiology, prevention and control of rheumatic heart disease with a focus on the Pacific Islands region* Darwin, Australia: Global Health Division, Menzies School of Health Research, Charles Darwin University; 2015.
332. Sullivan E, Vaughan G, Jackson Pulver L, et al. Perinatal outcomes of women with rheumatic heart disease. *J Paediatr Child Health.* 2016;52(Vol 52 Supplement S2):1–131.

333. Saxena A. Rheumatic heart disease screening by “point-of-care” echocardiography: an acceptable alternative in resource limited settings? *Translational Pediatrics.* 2015;4(3):210-213.
334. Beaton A, Okello E, Destigter K, Scheel A, Perlman L, Sable C. Impact of rheumatic heart disease on maternal outcomes in pregnancy: Leveraging existing infrastructure to address a critical knowledge gap. *Global Heart.* 2016;11(2 SUPPL. 1):e75.
335. Steinberg ZL, Dominguez-Islas CP, Otto CM, Stout KK, Krieger EV. Maternal and fetal outcomes of anticoagulation in pregnant women with mechanical heart valves. *Journal of the American College of Cardiology.* 2017;69(22):2681-2691.
336. Forcillo J, Watkins DA, Brooks A, et al. Making cardiac surgery feasible in African countries: Experience from Namibia, Uganda, and Zambia. *The Journal of Thoracic and Cardiovascular Surgery.* 2019.
337. Zilla P, Bolman RM, Yacoub MH, et al. The Cape Town Declaration on Access to Cardiac Surgery in the Developing World. *Cardiovasc J Afr.* 2018;29(4):256-259.
338. Zilla P, Yacoub M, Zühlke L, et al. Global unmet needs in Cardiac Surgery. *Global heart.* 2018.
339. Yacoub M, ElGuindy A, Afifi A, Yacoub L, Wright G. Taking cardiac surgery to the people. *Journal of Cardiovascular Translational Research.* 2014;7(9):797-802.
340. Yacoub M, Mayosi B, ElGuindy A, Carpentier A, Yusuf S. Eliminating acute rheumatic fever and rheumatic heart disease. *The Lancet.* 2017;390(10091):212-213.
341. Fauveau V, Sherratt D, de Bernis L. Human resources for maternal health: multi-purpose or specialists? *Hum Resour Health.* 2008;6(1):21.
342. Lee I, Purbrick B, Barzi F, et al. Cohort profile: The Pregnancy and Neonatal Diabetes Outcomes in Remote Australia (PANDORA) Study. *Int J Epidemiol.* 2018.
343. Sliwa K, Azibani F, Baard J, et al. Reducing late maternal death due to cardiovascular disease - A pragmatic pilot study. *Int J Cardiol.* 2018.
344. Kennedy HP, Cheyney M, Dahlen HG, et al. Asking different questions: A call to action for research to improve the quality of care for every woman, every child. *Birth.* 2018;45(3):222-231.
345. Powell C. The Delphi technique: myths and realities. *J Adv Nurs.* 2003;41(4):376-382.
346. Boulkedid R, Abdoul H, Loustau M, Sibony O, Alberti C. Using and reporting the Delphi method for selecting healthcare quality indicators: a systematic review. *PLOS ONE.* 2011;6(6):e20476.
347. McDonnell LA, Turek M, Coutinho T, et al. Women's heart health: knowledge, beliefs, and practices of Canadian physicians. *Journal of Women's Health.* 2018;27(1):72-82.
348. Mosca L, Linfante AH, Benjamin EJ, et al. National study of physician awareness and adherence to cardiovascular disease prevention guidelines. *Circulation.* 2005;111(4):499-510.
349. Brown S, Weetra, D, Glover, K, Buckskin, M, Ah Kit, J, Leane, C, Mitchell, A, Stuart-Butler, D, Turner, M, Gartland, D, Yelland, J. Improving Aboriginal women’s experiences of antenatal care: findings from the Aboriginal families study in South Australia. *Birth.* 2015;42(1):27-37.
350. Kildea S, H S, Murphy R, NB L, Gibbons K. The Murri clinic: a comparative retrospective study of an antenatal clinic developed for Aboriginal and Torres Strait Islander women. *BMC Pregnancy and Childbirth.* 2012;12(159):1471-2393 (Electronic).

351. Bertilone CM, McEvoy SP, Gower D, Naylor N, Doyle J, Swift-Otero V. Elements of cultural competence in an Australian Aboriginal maternity program. *Women and Birth.* 2017;30(2):121-128.
352. NSW Health. *NSW Aboriginal Maternal and Infant Health Strategy Evaluation.* North Sydney 2005.
353. Reibel T, Morrison L, Griffin D, Chapman L, Woods H. Young Aboriginal women's voices on pregnancy care: Factors encouraging antenatal engagement. *Women and Birth.* 2015;28(1):47-53.
354. Clarke M, Boyle J. Antenatal care for Aboriginal and Torres Strait Islander women. *Australian Family Physician.* 2014;43(1/2):20-24.
355. Elliott C, Sliwa K, Anthony J. Perinatal outcome in pregnant women with heart disease attending a combined obstetric and cardiology clinic in a resource limited country. *Int J of Gynecology, Obstetrics and Neonatal Care.* 2015;2(2).
356. Campanharo FF, Cecatti JG, Haddad SM, et al. The impact of cardiac diseases during pregnancy on severe maternal morbidity and mortality in Brazil. *PLoS ONE.* 2015;10(12):e0144385.
357. RHD Action. *RHD Global Status Report 2015-2017.* Geneva: World Heart Federation; 2016.
358. Watkins DA, Zühlke LJ, Narula J. Moving forward the RHD agenda at global and national levels. *Global Heart.* 2017;12(1):1-2.
359. Ruddock P. *Aboriginal Health: report from the House of Representatives Standing Committee on Aboriginal Affairs.* Canberra: The Parliament of the Commonwealth of Australia; 1979.
360. Health Policy Analysis. *Evaluation of the Commonwealth Rheumatic Fever Strategy – Final report.* Canberra: Primary Healthcare Branch, Commonwealth Department of Health; 2017.
361. Australian Bureau of Statistics (ABS). Table 1.1 Underlying cause of death, All causes, Australia (All-female age-standardised) 2015. In: ABS, ed. Canberra ACT2016.
362. Katzenellenbogen J, Kruger D, Nedkoff L, de Klerk N, Hung J. Over-counting rheumatic heart disease in hospital administrative data: does it matter and what can be done? *Heart, Lung and Circulation.* 2017;26:S332-S333.
363. Lancellotti P, Badano L, Lang R, et al. Normal reference ranges for echocardiography: rationale, study design, and methodology (NORRE Study). *European Heart Journal – Cardiovascular Imaging.* 2013;14(4):303-308.
364. World Health Organisation (WHO). *Integrating equity, gender, human rights and social determinants into the work of WHO: Roadmap for Action (2014-2019).* Geneva: WHO; 2015.
365. Knowles R, Smith A, Lynn R, Rahi J, on behalf of the British Paediatric Surveillance Unit (BPSU). Using multiple sources to improve and measure case ascertainment in surveillance studies: 20 years of the British Paediatric Surveillance Unit. *Journal of Public Health.* 2006;28(2):157-165.
366. Frenk J, Gómez-Dantés O. False dichotomies in global health: the need for integrative thinking. *The Lancet.* 2017;389(10069):667-670.
367. Soma-Pillay P, Seabe J, Sliwa K. The importance of cardiovascular pathology contributing to maternal death: Confidential Enquiry into Maternal Deaths in South Africa, 2011–2013. *S Afr Med J.* 2016;27(2):60-65.
368. DiGiacomo M, Davidson P, Taylor K, et al. Health information system linkage and coordination are critical for increasing access to secondary prevention in Aboriginal health: a qualitative study. *Quality in Primary Care.* 2010;18:17-26.

369. Kelly J, Ramage M, Perry D, et al. *Managing Two Worlds Together. Stage 3: Improving Aboriginal Patient Journeys - Cardiac Case Studies.* Melbourne: The Lowitja Institute; 2015.
370. World Health Organisation (WHO). *Rheumatic heart disease: Draft resolution proposed by Australia, Brazil, Canada, Cook Islands, Ecuador, Fiji, Japan, Namibia, New Zealand, Pakistan, Samoa, Tonga and Tuvalu.* Geneva, Switzerland: WHO; 2017.
371. World Health Organisation (WHO). *The World Health Report 2013: Research for Universal Health Coverage.* Geneva 2013.
372. World Health Organisation (WHO). About the Sustainable Development Goals. 2019; https://www.un.org/sustainabledevelopment/sustainable-development-goals/, 2019.
373. Nilsson M, Chisholm E, Griggs D, et al. Mapping interactions between the sustainable development goals: lessons learned and ways forward. *Sustainability science.* 2018;13(6):1489-1503.
374. Morton S, Pencheon D, Squires N. Sustainable Development Goals (SDGs), and their implementation: A national global framework for health, development and equity needs a systems approach at every level. *British Medical Bulletin.* 2017;124(1):81-90.
375. Pholeros P, Lea T, Rainow S, Sowerbutts T, Torzillo PJ. Improving the state of health hardware in Australian Indigenous housing: building more houses is not the only answer. *International journal of circumpolar health.* 2013;72(1):21181.
376. Say L, Chou D, Gemmill A, et al. Global causes of maternal death: a WHO systematic analysis. *The Lancet Global Health.* 2014;2(6):e323-e333.
377. Alkema L, Chou D, Hogan D, et al. Global, regional, and national levels and trends in maternal mortality between 1990 and 2015, with scenario-based projections to 2030: a systematic analysis by the UN Maternal Mortality Estimation Inter-Agency Group. *The Lancet.* 2016;387(10017):462-474.
378. Damian DJ, Njau B, Lisasi E, Msuya SE, Boulle A. Trends in maternal and neonatal mortality in South Africa: a systematic review. *Systematic reviews.* 2019;8(1):76-76.
379. Pillay Y, Barron P. On the path to reach the SDG targets: Decreasing maternal and child mortality in South Africa. *South African Medical Journal.* 2018;108(3):2-3.
380. Moodley J, Fawcus S, Pattinson R. Improvements in maternal mortality in South Africa. *South African Medical Journal.* 2018;108(3):4-8.
381. Kassebaum NJ, Bertozzi-Villa A, Coggeshall MS, et al. Global, regional, and national levels and causes of maternal mortality during 1990–2013: a systematic analysis for the Global Burden of Disease Study 2013. *The Lancet.* 2014;384(9947):980-1004.
382. Kassebaum NJ, Lopez AD, Murray CJL, Lozano R. A comparison of maternal mortality estimates from GBD 2013 and WHO. *The Lancet.* 2014;384(9961):2209-2210.
383. Hansen CH, Schellenberg JR. Modest global achievements in maternal survival: more focus on sub-Saharan Africa is needed. *Lancet.* 2016;387(10017):410-411.
384. World Health Organisation (WHO). *Global Strategy for Women's, Children's and Adolescents' Health.* Geneva: WHO; 2016.
385. Campbell OMR, Graham WJ. Strategies for reducing maternal mortality: getting on with what works. *The Lancet.* 2006;368(9543):1284-1299.
386. Creanga AA, Syverson C, Seed K, Callaghan WM. Pregnancy-related mortality in the United States, 2011–2013. *Obstetrics and gynecology.* 2017;130(2):366.
387. MacDorman MF, Declercq E, Thoma ME. Trends in maternal mortality by socio-demographic characteristics and cause of death in 27 states and the District of Columbia. *Obstetrics and gynecology.* 2017;129(5):811.

388. Li L, O'Neil L. *Mothers and Babies 2015: Northern Territory Midwives' Collection.* Darwin: Department of Health; 2018.
389. Ilton M. Personal communication. In:2019.
390. Marangou J, Ferguson D, Ilton M, Remenyi B. Echocardiography in Pregnant Indigenous Australians: An Under-Utilised Resource? *Heart, Lung and Circulation.* 2019;28:S48.
391. Remote Primary Health Care Manuals. Minymaku Kutju Tjukurpa Women's Business Manual. 2017; 6th edition:Standard Treatment Manual for Women's Business in remote and Indigenous health services in central and northern Australia. Available at: https://rphcm.allette.com.au/publication/wbm.html.
392. World Health Organization (WHO). *Selected practice recommendations for contraceptive use (3rd Edition).* WHO; 2016.
393. Boyle J, Eades S. Closing the gap in Aboriginal women's reproductive health: some progress, but still a long way to go. *Australian and New Zealand Journal of Obstetrics and Gynaecology.* 2016;56(3):223-224.
394. World Health Organisation (WHO). *Meeting to develop a global consensus on preconception care to reduce maternal and childhood mortality and morbidity.* Geneva: World Health Organization Headquarters; 2013.
395. Roos-Hesselink J, Cornette J, Sliwa K, Pieper P, Veldtman GR, Johnson M. Contraception and cardiovascular disease. *Eur Heart J.* 2015;36(27):1728-1734.
396. Yankah C, Fynn-Thompson F, Antunes M, et al. Cardiac surgery capacity in sub-saharan Africa: quo vadis? *Thorac Cardiovasc Surg.* 2014;62(5):393-401.
397. Department of Health. Media release 25/2/2019: $35 Million for Vaccine to End Rheumatic Heart Disease. 2019; http://www.health.gov.au/internet/ministers/publishing.nsf/Content/health-mediarel-yr2019-wyatt031.htm. Accessed 27/2/2019.
398. Park S, Abrams R. Alma-Ata 40th birthday celebrations and the Astana Declaration on Primary Health Care 2018. In: British Journal of General Practice; 2019.
399. Dawson A, Gray N. *Human Resources for Health in Maternal, Neonatal and Reproductive Health at the Community Level: A Synthesis of the Literature with a focus on the Asia Pacific Region.* Sydney Australia: UNSW and Burnet Institute; 2010.
400. World Health Organisation (WHO). *Strengthening quality midwifery education for Universal Health Coverage 2030: framework for action.* Geneva: WHO; 2019.
401. Schoon MG, Motlolometsi MWA. Poor maternal outcomes: A factor of poor professional systems design. *S Afr Med J.* 2012;102(10).
402. Horton R, Astudillo O. The power of midwifery. *The Lancet.* 2014;384(9948):1075-1076.
403. Homer CSE, Friberg IK, Dias MAB, et al. The projected effect of scaling up midwifery. *The Lancet.* 2014;384(9948):1146-1157.
404. Kruk ME GA, Arsenault C, Jordan K, Leslie HH, Roder-DeWan S, et al.,. High-quality health systems in the Sustainable Development Goals era: time for a revolution. *Lancet Glob Health.* 2018;6(11):e1196-e1252.
405. Dawson AJ, Nkowane AM, Whelan A. Approaches to improving the contribution of the nursing and midwifery workforce to increasing universal access to primary health care for vulnerable populations: a systematic review. *Human resources for health.* 2015;13(1):97.
406. World Health Organisation (WHO). *Prevention and control of noncommunicable diseases: guidelines for primary health care in low resource settings.* World Health Organization; 2012.

407. Castro Lopes S, Nove A, Ten Hoope-Bender P, et al. A descriptive analysis of midwifery education, regulation and association in 73 countries: the baseline for a post-2015 pathway. *Human Resources for Health.* 2016;14(37).
408. United Nations Population Fund. *The State of the World's Midwifery: Analysis of the Sexual, Reproductive, Maternal, Newborn and Adolescent Health Workforce in East and Southern Africa.* 2017.
409. Filby A, McConville F, Portela A. What Prevents Quality Midwifery Care? A Systematic Mapping of Barriers in Low and Middle Income Countries from the Provider Perspective. *PloS one.* 2016;11(5):e0153391-e0153391.
410. Renfrew MJ, Homer CSE, Downe S, et al. Midwifery: an executive summary for The Lancet's series. *Lancet.* 2014;384(1):8.
411. Adegoke A, Utz B, Msuya SE, van den Broek N. Skilled Birth Attendants: Who is Who? A Descriptive Study of Definitions and Roles from Nine Sub Saharan African Countries. *PLOS ONE.* 2012;7(7):e40220.
412. World Health Organisation (WHO). *WHO recommendations: optimizing health worker roles to improve access to key maternal and newborn health interventions through task shifting.* Geneva 2012.
413. Francis J, Fairhurst H, Kaethner A, et al. Handheld PLAX View echocardiography by healthworkers for detection of RHD. *Heart, Lung and Circulation.* 2019;28:S51-S52.
414. Pholeros P, Torzillo P, Rainow S. *Housing for health: towards a healthy living environment for Aboriginal Australia.* Healthabitat; 1993.
415. Healthabitat. Healthabitat: a brief history. The 'Uwankara Palyanku Kanyintjaku' model. 2011; http://www.healthabitat.com/about-page/organisation-background, 2019.
416. Allen + Clarke. *Healthy Homes Initiative Evaluation: Final Report.* Auckland: Ministry of Health New Zealand,; 2018.
417. NSW Health. *Closing the gap: 10 Years of Housing for Health in NSW - An evaluation of a healthy housing intervention.* Sydney NSW 2010.
418. World Health Organisation (WHO) Europe. *Environmental burden of disease associated with inadequate housing: a method guide to the quantification of health effects of selected housing risks in the WHO European Region.* Denmark: WHO; 2011.
419. World Health Organisation (WHO). *WHO Housing and Health Guidelines.* Geneva: WHO CTI; 2018.
420. Howden-Chapman P, Roebbel N, Chisholm E. Setting Housing Standards to Improve Global Health. *Int J Environ Res Public Health.* 2017;14(12):1542.
421. Lea T. Housing for Health in Indigenous Australia: driving change when research and policy are part of the problem. *Human Organization.* 2008:77-85.
422. de Wet T, Plagerson S, Harpham T, Mathee A. Poor housing, good health: a comparison of formal and informal housing in Johannesburg, South Africa. *Int J Public Health.* 2011;56(6):625-633.
423. United Nations General Assembly. *68/261. Fundamental principles of official statistics.* Geneva 2014.
424. Wagner EH, Austin BT, Davis C, Hindmarsh M, Schaefer J, Bonomi A. Improving chronic illness care: translating evidence into action. *Health Affairs.* 2001;20(6):64-78.
425. Davy C, Bleasel J, Liu H, Tchan M, Ponniah S, Brown A. Effectiveness of chronic care models: opportunities for improving healthcare practice and health outcomes: a systematic review. *BMC Health Serv Res.* 2015;15(1):194.
426. Glasgow RE, Orleans TC, Wagner EH, Curry SJ, Solberg LI. Does the chronic care model serve also as a template for improving prevention? *Milbank Q.* 2001;4.

427. Bodenheimer T, Wagner EH, Grumbach K. Improving primary care for patients with chronic illness: the Chronic Care Model, part 2. *JAMA.* 2002;288.
428. Barr V, Robinson S, Marin-Link B, et al. The expanded chronic care model. *Hosp Q.* 2003;7(1):73-82.
429. Davy C, Bleasel J, Liu H, Tchan M, Ponniah S, Brown A. Factors influencing the implementation of chronic care models: A systematic literature review. *BMC Family Practice.* 2015;16(1):102.
430. Bauer AM, Thielke SM, Katon W, Unützer J, Areán P. Aligning health information technologies with effective service delivery models to improve chronic disease care. *Preventive Medicine.* 2014;66:167-172.
431. George AS, LeFevre AE, Schleiff M, Mancuso A, Sacks E, Sarriot E. Hubris, humility and humanity: expanding evidence approaches for improving and sustaining community health programmes. *BMJ Global Health.* 2018;3(3):e000811.
432. Hollows F. *Fred Hollows: an autobiography with Peter Corris.* Richmond, Melbourne: John Kerr; 1991.
433. Australian Institute of Health and Welfare (AIHW). National Rheumatic Heart Disease Data Collection (NRHDC). 2019; https://www.aihw.gov.au/about-our-data/our-data-collections/national-rheumatic-heart-disease-data-collection. Accessed 26/4/2019.
434. Roberts R, Ketchell A. Clinical assessment of women with cardiovascular abnormalities. *British Journal of Midwifery.* 2012;20(4):246-250.
435. Siu S, Colman JM. Cardiovascular problems and pregnancy: an approach to management. *Cleveland Clinic Journal of Medicine.* 2004;71(12):977-985.