Support Services for adolescents and young adults with cancer or a blood disorder:

Measurement Properties and Validation of Quality of Life Instruments for Adolescents and Young Adults with Cancer or a Blood Disorder

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Certificate of Authorship/Originality

I certify that the work in this thesis has not previously been submitted for a degree nor has it been submitted as part of requirements for a degree except as fully acknowledged within the text.

I also certify that the thesis has been written by me. Any help that I have received in my research work and the preparation of the thesis itself has been acknowledged. In addition, I certify that all information sources and literature used are indicated in the thesis.

Signature of Candidate

Jane Elizabeth Ewing

Dedication

I dedicate this thesis to my family, David my husband and our beloved children Elizabeth, William and Victoria for their support, love and encouragement. Also, in memory of my Mother, Heather for her patient and self-less love as always and in helping me nurse Elizabeth in her final stage of life; in memory of my Father, Lance who has keenly watched my progress in this neglected field of healthcare; in memory of my cousins Donald, Tracey and Judith whose lives were also cut short by cancer.

In particular I dedicate this thesis to the memory of my elder daughter, Elizabeth, a vivacious, gifted, generous, loving, courageous, community spirited individual with a passion for encouraging cultural harmony. She achieved much and inspired many in her 19 years. Her cancer gave her family an unwanted personal introduction to the huge gap in the health system for adolescents and young adults and the impact that cancer has on them, their siblings, parents, families and friends.

This horrendous tragedy is the driving force behind my work to help reduce the burden of illness for adolescents and young adults living with cancer, and their families. Elizabeth laid the initial challenge before me to help make the system fairer for these young people. I work and live in the hope that her untimely death will not be in vain.

Finally, I dedicate this thesis to my beloved New Zealand, Aotearoa.

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I am indebted to Madeleine King for agreeing to become my Co-supervisor. I am grateful for her understanding and appreciation of my vision, seeing the potential in my ideas, and helping me refashion and strengthen my long-term plan in this area of youth health. My learning curve has been very steep in many areas, such as, critical and precise thinking, building conceptual models, working with clinicians, developing project budgets, writing funding applications, politics, and maintaining focus.

The field-testing of this study was done with the co-operation of the Centre for Children's Cancer and Blood disorders (CCCBD) at Sydney Children's Hospital (SCH), Randwick. I am grateful to Belinda Goodenough, Hospital Scientist for her on-going support and encouragement in helping me through the many hoops in gaining access to the required patients, smoothing the research process at the coal face, and assisting and refining funding applications, presentations and items for publication. Belinda's ability to make things happen in the hospital environment complemented Madeleine and Narelle's expertise. I am grateful to Glen Marshall, Director of the CCCBD, for permitting me to work closely with the staff at his Centre, particularly clinicians: Richard Cohn, Marcus Vowels, and Sue Russell for allowing access to their patients.

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List of Abbreviations

AHS Area Health Service

AIHW Australian Institute for Health and Welfare

AYA Adolescents and young adults

ALL Acute lymphatic leukaemia

CAAH Centre for Advancement of Adolescent Health

CCCBD Centre for Children's Cancer & Blood Disorders at SCH

CHeRP Centre for Health Research & Psycho-oncology

CNC Clinical Nurse Consultant

CRASH Centre for Research into Adolescents' Health

DHB District Health Board

DHN District Health Nurse

EEMF Elizabeth Ewing Memorial Framework

EEMCVAFH Elizabeth Ewing Memorial Conceptual View of AYA Family Health

EFA Exploratory Factor Analysis

GMCT The Greater Metropolitan Clinical Taskforce

GMTT The Greater Metropolitan Transition Taskforce

GP General Practitioner

HRQOL Health-Related Quality of Life

IAAH International Association for Adolescent Health

IPOS International Psycho-Oncology Society

MSAS The Memorial Symptom Assessment Scale

NAAH NSW Association for Adolescent Health

NGO Non-Government Organisation

NHMRC National Health & Medical Research Council

NZAAHD New Zealand Association for Adolescent Health & Development

OT Occupational Therapist

PMCC Peter Macullum Centre for Cancer

POWH The Prince of Wales Hospital, Randwick, NSW, Australia

QOL Quality of Life, in general

RCT Randomised Clinical Trials

RHW Royal Hospital for Women, Randwick, NSW Australia

SCH Sydney Children's Hospital, Randwick, NSW, Australia

SCNS Supportive Care Needs Survey

SEALS South Eastern Area Laboratory Services

SESAHS South Eastern Sydney Area Health Service, NSW, Australia

TCYPCCI Transition Care for Young People with Chronic Childhood Illnesses

UNCROC United Nations Convention on the Rights of the Child

UTS University of Technology, Sydney

WHO World Health Organisation

WINZ Work and Income New Zealand

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Abstract

Health-Related Quality of Life (HRQOL) is an important outcomes measure in cancer and there are specific issues depending on the site, stage, treatment and patient age. Although numerous instruments are available for cancer HRQOL, most are designed for adults, some for children, but none for adolescents and young adults (AYA) who have special age-specific concerns and poor improvement in survival compared with other age groups.

An existing HRQOL instrument was modified to ensure its suitability for AYA, its validity, reliability and sensitivity were tested in Australians aged 16 to 25 years old diagnosed with cancer or a blood disorder. Varni's PedsQL^{TM} Measurement Model (13-18 year olds) was selected, modified then administered to families recruited from haematology/oncology clinics and wards at three Sydney Metropolitan Hospitals in person or by telephone. The Memorial Symptom Assessment Scale was used to categorise participants into groups reflecting sensitivity of symptom severity (slight, moderate and severe).

The instruments demonstrated excellent internal consistency reliability, making them suitable for both group and individual comparisons. Clinical validity, construct validity, and discriminant validity were demonstrated by "known-groups" analysis, exploratory factor analysis and correlations, respectively.

These new versions of the PedsQL Generic Core and Cancer Module are reliable, valid and sensitive measures of HRQOL in patients aged 16-25 years diagnosed with cancer or a blood disorder. The measures will soon be available for use as outcome measures in clinical trials and clinical practice with this age cohort in Australasia and internationally.

C h a p t e r   1

1. Introduction

This thesis forms part of phase one of a two-phase research programme to help facilitate a reduction in the burden of illness for adolescents and young adults with a cancer or a blood disorder diagnosis, and their families.

Phase one is the field-testing and validation in an Australian context, of a set of instruments to measure health related quality of life (HRQOL), patient satisfaction, preferences for support services, the impact of illness and treatment on the patient and their family, and the development of a good recruitment strategy for the second phase. In phase two, the previously validated instruments of phase one will be utilised to carry out the aims of the programme.

Overall the programme aims to document the adequacy of support services currently used in NSW and identify directions for improvement and development of age-appropriate care and services which may better address unmet needs and preferences of this population group. Using questionnaire and interview methods, this programme is very timely as it can inform the process and delivery of services. It fits well with current NSW State and Federal age-specific and general policy and legislation, and can provide baseline data for evaluating and monitoring changes in support services, to help guide improvement, and assist clinicians in forming partnerships with this patient group.

After obtaining ethical approval from the appropriate institutions, the collection, handling, and analysis of data commenced. Consequently, this thesis field-tests and validates the previously validated HRQOL instruments, which were modified for a slightly older patient group. For efficiency in data collection, the satisfaction, preferences and impact report instruments were included in the survey booklets but these data will be analysed after this thesis is completed, and in preparation for phase two of the programme.

Chapter one provides the introduction and establishes the context for the validation of the HRQOL instruments. It briefly defines "adolescents and young adults" (AYA) and outlines the health problems for AYA cancer patients, possible solutions and improvements achieved to date.

Chapter two describes my contribution to helping improve health outcomes for adolescents and young adults with cancer and blood disorders, my current research contribution as presented in this thesis, a description of HRQOL and the instruments used to measure HRQOL, and more specifically the PedsQL™ Measurement Model.

Chapter three describes the methodology of the study including instrument modifications, recruitment, data collection, the scoring of the data and the analysis. The analysis includes investigation of reliability, feasibility, and validity of the modified instrument. Validity incorporates Construct Validity, Convergent and Divergent Validity, and Clinical (Discriminant) Validity.

Chapter four provides the results of the analyses. Chapter five outlines the conclusions, implications and limitations of this validation study; speculation on its use; and suggests further research required in the quest for better health outcomes for adolescents and young adults with cancer and blood disorders.

1.1. Background

1.1.1. Literature Review

For relevant citations I searched via several search engines: Informit for databases Meditext, AMI, APAIS Health and Australian Family and Society; Ovid for the databases Medline, Cinahl, PsycInfo; and the Cochrane Library. The search criteria were "youth or adolescent or young adult", with "support services or illness burden or psychosocial needs", and "cancer or blood disorder or neoplasms" (or chronic illness), then with Health-Related Quality of Life (HRQOL).

1.1.1.1. Definition and terminology for target group

There are many different definitions and terminologies for people aged 16-24 years old (inclusive), with much debate and no consensus. Michelagnoli reports that the term

`adolescent' is less than ideal because it has implications, for many people, that tend to typecast the patient as potentially immature, rebellious and, as far as treatment is concerned, often non-compliant (Hemming, 1960; Michelagnoli et al., 2003, p439).

The World Health Organisation (WHO) and the Youth Advisory Council Act (1989) use the term "young people" to describe people in the age bracket 12-24 years (inclusive). Young adults have also been defined as 12-25 and 15-30 years old. Hospitals on the other hand, tend to define adolescence depending on whether the hospital is primarily an adult hospital where these patients are 'young adults', or a paediatric hospital where adolescents are 10-16, but some variation on a case-by-case basis occurs depending on the patient's age at diagnosis, maturity and the illness. For example, sometimes paediatric hospitals have been known to extend to 20-21 years, while some adult hospitals have taken adolescents of 14 years old into their oncology/haematology unit. These definitions possibly have their roots from a combination of the school leaving age at the beginning of 20th Century, the legal working age at that time, and subsequently tied to other milestones of recognized maturity in terms of our laws. Lack of consensus in definition and terminology is likely the first obstacle to addressing their needs. I define the age group 16 up to 25 years adolescent and young adults.

For the purpose of my study I considered 'young adults' (16-25) to be a subset of the broader category of 'young people' (12-25). (The term 'youth' is also commonly used in both Australian and New Zealand policy for the 12-24 inclusive year olds). Therefore I use the terms, 'young adult', 'adolescent', 'youth', and 'young person/people' interchangeably to mean 16 up to 25 year olds, and for ease I abbreviate the term 'adolescents and young adults' as AYA. Hence, the target group for this study is AYA patients with a cancer or blood disorder diagnosis.

The term 'parent' is used to define the AYA person's nominated carer-proxy. This person could be the young person's biological parent, guardian, spouse or their main caregiver.

In the next sections I outline the health outcome problem for AYA patients, what advances have been made so far, and how my research contributes to improving health outcomes for this population group.

1.1.1.2. Problems for adolescents & young adults in the health system

Mortality statistics indicate 10 to 24 year olds are the only age group not to have had a significant improvement in health status since 1960 (Watson, 2001). One clear indicator of a health outcome problem for AYA cancer and blood disorder patients is their high mortality rate, which has been slow to decline (Mitchell et al., 2004). Of all age groups, young people diagnosed with cancer have the least improvement in survival over the last 25 years (Bleyer, 2002a), and although there has been a modest improvement in survival this has not been commensurate with the impressive technological advances reflected in the survival rates as enjoyed by other patient groups (Figure 1.1), particularly in patients over 50 years old, and in paediatric oncology (Pentheroudakis et al., 2005).

Figure 1-1: **Improvement in Relative Cancer Survivorship by Age**
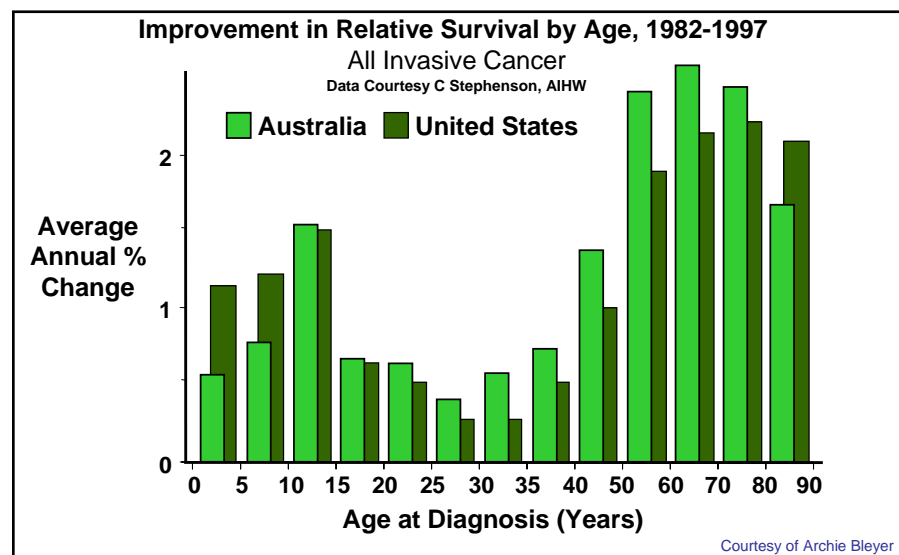
The Governing Committee of the *Cancer Control Network* acknowledges that adolescents with cancer "present a challenge that is not adequately addressed by current systems or models of care in Australia" (White, 2002). The survivors of various childhood diseases are a growing and vulnerable population, which must be "appropriately provided for while they traverse the turbulence of adolescence" (NSW Ministerial Youth Health Taskforce, 1991).

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Incidence of cancer

The WHO predicts "a quantum leap in cancer deaths" world-wide in the next 15 years (World Health Organisation, 2003). For AYA, cancer incidence has increased (Barr, 1999; Giles et al., 1996; Mitchell et al., 2004) by 30% in 10-24 year olds between 1993 and 2001 - an eight year period (Giles et al., 1996), while for all groups in NSW cancer incidence has increased by almost 25% over the twenty year period 1980 to 2000 (NSW Department of Health, 2003). Barr (1999) reports that "since the inception of the Surveillance, Epidemiology, and End Results (SEER) program of the National Cancer Institute in USA in 1973, the increase in cancer incidence has been greater than 30% for 15-19 year olds and less than 10% in any of the younger quintiles with similar incidence rates reported in Canada and Australia" (Barr, 1999). Bleyer (2002) reports cancer incidence for AYA at nearly twice the rate observed in 5-14 year olds (Bleyer, 2002a), and increasing at a faster rate than in the younger paediatric age bracket, i.e. the 0-15 year olds. Cancer control experts anticipate significant increases in cancer incidence in New Zealand over the next 10 years (Minister of Health, 2003). In Australasia about 1200 new cases of cancer in AYA are diagnosed each year (AIHW, 1997-1999; NZHIS, 1996-97).

In NSW, malignant melanoma accounted for a quarter of all cancers at age 15-19 and the incidence was three times that in the USA white population (Stiller, 2002). Furthermore, in New Zealand the "cancer death rate has increased at a faster rate, and is now higher than that of comparable countries" (New Zealand Ministry of Health et al., 2003).

To help counter the projected increase in cancer incidence, Australia and New Zealand, like America, Britain and Canada, have developed cancer control strategies to oppose this trend. The strategy is to target prevention, screening, early diagnosis, treatments, rehabilitation, support and palliative care. Preventative measures for example, include anti-smoking campaigns, skin care and better nutritional awareness, which targets young people. However, health systems and governments often fail (Ewing, 2000) to engage sufficient resources to rectify the gap in care for AYA patients with cancer to ensure improved health outcomes. For example, "the 5-year outcome in 15-19 year olds with leukaemias and sarcomas is not only worse than in younger patients, but also lower

in this population at large than in patients of the same age treated at Children's Cancer Group institutions" (Bleyer, 2002a). Therefore, "the North American experience strongly suggests a specialty of adolescent medicine" (Malus, 1992) to help ensure the concerns of this group are addressed.

1.1.1.2.1. Factors contributing to poor mortality

Several factors may contribute to the poor mortality rate for adolescents and young adult patients, such as the gap between adult care and paediatric care (Ewing, 2000, 2000a; Leonard et al., 1995; Pentheroudakis et al., 2005), lack of specialty medicine for AYA with cancer (Ewing, 1999; Geehan, 2003; Hemming, 1960; Oppong - Odiseng et al., 1997), late diagnosis (Geehan, 2003; Hartley et al., 2001), health professional self-perceived lack of training in adolescent health (Blum et al., 1990; Gans et al., 1991), lack of appropriate models of care (White, 2002), poor inclusion rate in clinical trials (Mitchell et al., 2004), inappropriate data collection, often poor follow-up on 'late effects', the often unmet psychosocial needs of AYA and their carers (Holland, 2003), and lack of research funding in this area (Ewing, 2003). Hence, youth with chronic illness have not realized the health gains experienced by other age groups over the last 30 years (Barr, 1999). These factors could be simplified to two main concerns: cancer incidence and the lack of AYA specialty medicine.

The literature points to:
i. The gap in care between the paediatric service and the adult service; and the need for:
ii. An adolescent and young adult cancer specialty medicine.
"Infants and children have their paediatric advocates, elderly patients have their geriatricians. Both these groups of specialists have advanced primary care medicine with insights that have now become part of standard care" (Malus, 1992). AYA patients are in need of specialised care, skilled nursing care and interaction with physicians (Pentheroudakis et al., 2005). Furthermore the Adolescent Medicine Paediatric Tertiary Services Review reports that the current service which is not cancer specific, is ad hoc, unco-ordinated and fragmented (Health Funding Authority et al., 1998), those with cancer are overlooked. Hemming reports that, we "have been slow to give adolescence the same concentrated care that has gone into the study of the baby and young child … If as a society we are to understand our adolescents instead of being estranged by them, we have to catch up fast on our ignorance of what life means to adolescents today, and

what the world looks like through their eyes." (Hemming, 1960; Oppong - Odiseng et al., 1997). According to the Calman Report the time is right for purchasers to recognise the needs of these AYA patients (Leonard et al., 1995).

An AYA specialty medicine in NSW could service about 20% of the population (about 1.25 million) aged 12 up to 25 years old (NSW Health, 1999), and the Youth Health Policy 1991 reports 10 to 20% of people under 20 years are affected by chronic illness or disability (NSW Ministerial Youth Health Taskforce, 1991). In NZ 15 to 20% of the population is aged between 12 and 24 years. Although, 1991 marked the founding of Australia's first National Academic Centre in Adolescent Health (Melbourne), it was 2005 before the first Chair of Adolescent Health in Australia was established, and Professor Susan Sawyer took this role. Sydney is close to establishing a Chair of Adolescent Health, but New Zealand lags behind.

iii. Facilities

Unfortunately, "the care of adolescent patients is often seen as neither the preserve of adult [services] ... nor the preserve of paediatric [services]" and hence the label: the 'lost tribe' (Michelagnoli et al., 2003) is quite fitting. Also in the hospital system AYA who are first diagnosed at age 16 and some as young as 14 years old are classified as 'adult', but "adult [services] do not extend to the arrangement of ancillary medical, psychological and educational support that are so important to people who are facing dangerous diseases and taxing treatment at a vulnerable time of their lives." Paediatric services on the other hand do provide this important supporting care (Leonard et al., 1995). But AYA prefer to be thought of as young adults rather than older children in a paediatric ward. Barr implores the "urgent need to rise to the challenge of effecting a seamless transition from pediatric to adult care" to bridge the gap in care (Barr, 1999). Transition Care refers to the "purposeful planned movement of adolescents and young adults with chronic physical and medical conditions from child centred to adult orientated health care systems" (Blum et al., 1993).

Unfortunately, "until transition to adult care is recognised by the adult health care system as requiring a demonstrable change in attitude and resources, little real progress will be possible" (Bennett et al., 2005). This difficulty could be due to the different way in which adult health care and paediatric health care deliver their services. In addition,

since paediatrics are well "versed in organizational aspect and supporting care" (Leonard et al., 1995), it is their systems that could be utilised as a starting point to developing best practice guidelines for an AYA specialty medicine, transition care and facilities.

Another "challenge before both the medical and pediatric health care communities is to assure adolescent patients the benefits of treatment …in age-appropriate settings." (Newburger et al., 2002). Hence, either dedicated centres or "programmes that can deliver the necessary age appropriate multidisciplinary management" are required (Capra et al., 2003).

There are far too few age appropriate facilities for AYA with cancer and what there is compares poorly with the number of paediatric units. For example, a position statement for the Society for Adolescent Medicine reported about 40-60 Adolescent Units in North America, with several units in Europe, Asia, South America and Australia. The numbers of beds ranged between 6 and 35 with most units having 11-20 beds. The age range of patients these units cared for was from 10-13 year olds through to 17-24 year old patients (Fisher et al., 1996).

In NZ over 84,000 young people are treated in hospitals each year (NZHIS, 1996-97), but they are usually scattered throughout each hospital. Pentheroudakis reports that adolescents and young adults not only need specialised care for intensive treatment and interaction with peers, family and physicians but also continuous psychosocial support (Pentheroudakis et al., 2005).

iv. Staff training

Most health professionals caring for adolescents have little or no formal training in adolescent health (Hein et al., 1994) and "many physicians feel that their specialty training did not prepare them with the necessary skills to manage effectively the complex social and emotional problems of adolescents" (Gans et al., 1991). Also, in a national survey of 3066 health professionals in USA covering physicians, nurses, social workers, nutritionists, and psychologists, they perceived themselves to have low levels of competency in meeting the health needs of adolescents (Blum et al., 1990). Gordon

reports that this is "one of the major barriers for adolescents accessing health care" (Gordon, 1996).

Furthermore, from time to time "physicians may vary in their interest in adolescent health problems and their experience in dealing with them…important aspects of adolescent growth and development were overlooked and resulted in diagnostic errors" (Strasburger, 1984). Many AYA patients experience late diagnosis, and yet delays in diagnosis are often crucial to survival (Geehan, 2003). The "WHO describes cancer diagnosis as the first step to cancer management" (New Zealand Ministry of Health et al., 2003). Early recognition and appropriate treatment is the key (Hartley et al., 2001). It may be rare for general practitioners (GPs) to see young people with cancer, but referrals months after initial presentation at the GP surgery as shown in the Table 1.1 are not infrequent and are a significant cause for concern (Whiteson, 2003).

Table 1-1 Time from first presentation with symptoms to diagnosis

Time to diagnosis	3 to 6 months	Greater than 6 months
13–17 year olds	26.3%	15.8%
18+ years	40%	20%

Source: Department of Paediatric and Adolescent Referral Patterns, RVI, Newcastle, cited in Whiteson 2003

Klein-Geltink reports that for Canadian adolescents treated in adult centres the "time between symptom onset and first treatment was longer for these adolescents, primarily due to the time between first health-care contact and assessment by a treating oncologist or surgeon" (Klein-Geltink et al., 2005).

"In the past physicians have believed that health practitioners should restrict their efforts to medical issues, and have placed behavioral and psychosocial concerns outside their purview. Recently there has been some movement towards an expanded role definition in which the comprehensive care of the patient is seen legitimately to include behavioral and psychosocial issues. In the case of adolescents with chronic illness, the psychosocial and medical concerns are so closely intertwined that no other position is tenable" (Hamburg, 1982, p439; World Health Organisation, 2003) Klopfenstein reports that "the psychological impact of a diagnosis of cancer and its life-threatening nature

can be viewed and understood if the developmental tasks of adolescents are considered" (Klopfenstein, 1999).

v. Psychosocial needs

"Psychosocial oncology is defined as a discipline that is concerned with all clinical and scientific attempts to clarify the significance of psychological and social factors in the development and course of cancer. Furthermore it addresses psychological and social factors in the patient's and family's process of coping with the disease, and attempts to apply this knowledge systematically to prevention, early detection, diagnosis, treatment, and rehabilitation of cancer patients." (Mehnert et al., 2005). In 1999 the Canadian Association of Psychosocial Oncology (CAPO) developed the National Psychosocial Oncology Standards for Canada, where they define psychosocial oncology as a professional sub-specialty in oncology which includes the formal study, understanding and treatment of the social, psychological, emotional, spiritual, quality of life and functional aspects of cancer as applied across the cancer trajectory from prevention through bereavement. (Cull et al. 1995, cited in Otfinowski et al., 2003).

It has been found that chronic illness (including cancer) places considerable burdens on the interpersonal relations of patients with their families (Hatchett et al., 1997). For the patient who is in adolescence or young adulthood, it is a time of major change both psychologically and physically, and is a period where they have unique psychosocial needs. Difficulties intrinsic in all of these steps to adult life are magnified by chronic illness, and immediately it becomes harder for young patients to feel accepted by their peer group, plan realistically for the future, and more difficult to become independent (Conway, 1998).

The New Zealand Cancer Control Strategy Action Plan sums it up well:

> "Adolescents with cancer have specific psychosocial needs, which are poorly addressed within the current arrangement of services. The diagnosis of cancer in the adolescent threatens to disrupt many of the maturational tasks desirable to attain adulthood. This results in increased dependence on caregivers; reduced peer contact and acceptance; disturbance of physical maturation and appearance; profound effects on developing sexual identity; and interrupted education and

career plans; Between 20 and 30 percent of survivors of cancer during adolescence develop symptoms of post-traumatic stress disorder" (Cancer Control Taskforce, 2005, p59).

A cancer diagnosis also impacts on the lives of the young person's siblings and parents and may extend beyond families to wider social circles. Furthermore the impact of bereavement on adolescent-aged siblings can be huge with far reaching consequences that can require considerable support for many years. On the release of the Clinical Guidelines on the Psychosocial Needs of Adults with Cancer, (National Breast Cancer Centre et al., 2003) Senator Patterson stated that "Undetected and untreated, emotional disorders have the potential to impact on the patient's family, friends, social networks and employment" (Patterson, 2003).

Otfinowski reports that in a study to determine the availability of psychosocial oncology care beyond the tertiary cancer centres in Alberta, Canada, 95% (144/151) of health care providers who responded to their survey felt it was important for cancer patients to have access to psychosocial care. However, only 18% were satisfied with the support services available in their community" (Otfinowski et al., 2003).

Also, a grounded theory study, aimed to develop a conceptual framework of the life experience of Taiwanese adolescents with cancer resulted in a qualitative study of sixteen adolescents involving an interview with the patient and primary caregiver, observations, medical chart reviews, nurses' notes, and researchers' reflexive journals. This study showed that "an unsettled state of mind emerged as the core category representing the life experience of adolescents with cancer" (Yeh, 2002).

The psychological differences between adolescents and adults are striking and are important to understand in order to provide optimal supportive care for young adults. If the developmental tasks of AYA are considered, then the psychological impact of a diagnosis of cancer and its life-threatening nature can be better understood (Klopfenstein, 1999). "The intense, complex, and enduring demands of treatment for childhood cancer on the family are well established. National and international recommendations for comprehensive care emphasize the importance of psychosocial services" (Kazak et al., 2003). But unfortunately, psychosocial care is sometimes

perceived as expensive and less essential than other evaluations and interventions yet psychosocial difficulties affect medical treatment (Kazak et al., 2003). When young people are consulted about health services, they frequently report that health services are often inaccessible and inappropriate to their needs (Pentheroudakis et al., 2005).

The psychological and social needs of cancer patients and their families are fundamental requirements to their care (International Psycho-Oncology Society, 2006) regardless of patient age. However, often " knowledge of relevant services is not available to the seriously ill patient and their family caregivers at the time of most need" (Burns et al., 2004) and this is regularly the situation for AYA at any stage of their disease. Fawzy and Devine call for a large variety of psychosocial interventions for potential psychological and physical health benefits as demonstrated in studies on psychosocial care for cancer patients where longer survival times were apparent for increasing numbers of patients. (Fawzy et al., 1995). Furthermore, additional funding, improved methodology, and multi-institutional cooperation will aid future paediatric psycho-oncology investigation (Patenaude et al., 2005).

Reduction of illness burden can be brought about through appropriate, well co-ordinated and timely support service provision that addresses the psychosocial needs of patients, their carers and families (Kazak et al., 2003). Spiegel reports that group support for cancer patients can result in significantly enhanced survival times and greater quality of life with less anxiety and depression, and half as much pain (Spiegel et al., 2000).

vi. Lack of Data

Another problem for AYA is the inappropriateness of data collection and the lack of entry for AYA into clinical trials where higher survival is associated with entry to trials or centralised treatment for certain cancers in this age group (Stiller, 2002). Bleyer (2002a) reports that only 5% of 15-25 year olds with cancer in the United States are entered onto clinical trials, in contrast to 60-65% of younger patients (Bleyer, 2002a). The seriousness of the gap in service provision and health outcomes for AYA patients has been disguised for too long. Firstly, defining adolescence, young people and young adults by chronological age or by developmental age poses challenges. Debate is on going and tends to counter progress in targeting this group in a suitable manner due to this lack of consensus.

Secondly, young people are in a state of transition between childhood and adulthood, and are recognised as a particularly vulnerable health demographic that is regularly overlooked. The historical splitting of the AYA population's health data into paediatric 0-14 and adult 15-44 data has contributed to the difficulty in addressing their unique issues and concerns because their data are lost within the paediatric or adult statistic. The collection of appropriate data would enable monitoring, evaluation, comparison and measurement, which in turn would facilitate identification and prioritisation of areas of concern for AYA. For example, there is little data on the psychosocial needs of AYA with cancer or chronic illness and complex needs, or the adequacy of existing support services in meeting their needs. Consequently, the needs of these young adults remain potentially unmet by paediatric and adult cancer services.

Thirdly, given significantly improved patient health outcomes for those included in clinical trials, it is of concern that many adolescents and young adults with cancer are usually excluded from trials (Mitchell et al., 2004). Adolescents aged 10–19 years are more likely to be recruited to a clinical trial if treated at a paediatric hospital. For example, 83% of Victorian (Australia) adolescents aged 10–15 years, and 14% of those aged 16–19 years were treated at paediatric institutions and had access to paediatric clinical trials. But only 4% of young adults aged 20–24 years were treated within clinical trials (Mitchell et al., 2004). More than 70% of older adolescent patients are not treated at institutions representing paediatric co-operative groups and are not enrolled in clinical trials. Failure to refer adolescent cancer patients to specialized paediatric oncology treatment centres also has an impact on their quality of care. Another "challenge before both the medical and pediatric health care communities is to assure adolescent patients the benefits of inclusion in clinical trials..." (Capra et al., 2003; Mitchell et al., 2004; Newburger et al., 2002).

As the number of paediatric cancer survivors increase, psychosocial researchers will be better able to conduct longitudinal studies on the impact of medical treatments and interventions to ameliorate the late effects of treatment (Patenaude et al., 2005).

vii Funding for Research

Finally, the lack of research funding continues to obstruct the constructive progress in this field of endeavour where the psychosocial needs of AYA cancer and blood disorder patients need to be addressed (Ewing, 2003).

1.1.1.3. Possible Solutions

Cancer during adolescence and early adulthood has been relatively neglected and merits enhanced national research programs and resources (Bleyer, 2002b).

i. Staff training

Training in AYA health (needs and issues) for primary, secondary and tertiary health care providers would help ensure "timely diagnosis for those with cancer and timely access to high-quality care throughout their experience of cancer" (New Zealand Ministry of Health et al., 2003).

"Adolescent specialists need continually to bring their clinical experience and research findings back to the primary care doctors…" (Malus, 1992). Spiro reports that doctors and other health professionals need to have a better insight into the spiritual aspects of life and death possibly by encouraging premedical students and qualified doctors to take courses in anthropology, religion or philosophy, and history to help them learn how different people deal with life and death (Spiro, 1996). This would highlight the need for staff training and sensitivity to the psychosocial needs of all patients and particularly of AYA.

ii. Clinical Practice Guidelines in Cancer

In training staff to assist in better patient outcomes, evidence-based practice guidelines are being developed in some countries in the field of cancer. For example, Australia's *Clinical Practice Guidelines for the Psychosocial Care of Adults with Cancer* 2003, which underscores the need for psychosocial intervention to complement all cancer treatment (National Breast Cancer Centre et al., 2003). It is expected to help improve outcomes and quality of life for Australian cancer patients and their families through the provision of comprehensive, evidence-based information about preventing, managing and treating the social and psychological consequences of cancer (National Breast

Cancer Centre et al., 2003). The Minister for Health and Ageing, Senator Kay Patterson also said,

> "It's time we all recognised that treating cancer patients is not just about managing the physical aspects of the disease. The psychological impact of cancer is equally important, with up to 50,000 Australians experiencing anxiety or depression each year following a diagnosis with cancer." (Patterson, 2003).

This is good progress and it may be helpful to measure the extent to which these guidelines are utilised by health professionals to direct patient care. But unfortunately, the psychosocial dimensions are often overlooked for AYA, and the above NSW guidelines acknowledge AYA cancer sufferers in only one short paragraph on p118. The NSW Cancer Plan, developed by the Cancer Institute established in 2003, also overlooks the needs of AYA as well as paediatric cancer patients (The Cancer Institute NSW, 2004). Perhaps these guidelines could be used as a base for developing practice guidelines for the AYA patient and family. New Zealand has, in draft form, a Service Specification for an AYA Oncology/Haematology Service. It is heartening that health care professionals are encouraged to translate psychosocial research findings into practice guidelines. For example, the brief for Hinds' team's research was to focus on hope research related to adolescents with cancer (Hinds et al., 2003), but more is required.

In addition to best practice guidelines, a national policy is required to provide a plan for the co-ordination of transition care between paediatric and adult services and the role of transition co-ordinators (Bennett et al., 2005). We still have a long way to go to achieve developmentally appropriate care, including facilities that meet the transitional needs of AYA.

iii. AYA specific Units
"The Calman Report [also states] ... the time is right for purchasers …to develop a national strategy for adolescent … units linked to major …centres" (Leonard et al., 1995). Also, Fisher and Kaufman (1996) write that, "It is imperative that appropriate care of hospitalized adolescents be included in the planning of health care services at

local, regional, and national levels." Furthermore, "The Society for Adolescent Medicine advocates the continuation and establishment of adolescent medicine inpatient units in both pediatric and general hospitals as an optimal approach to the delivery of developmentally appropriate health care to hospitalized adolescents. Such units should be geared to meeting the psychosocial needs of adolescents and the training needs of health professional students…" (Fisher et al., 1996).

In Britain, The Teenage Cancer Trust (TCT) felt that the advances enjoyed by paediatric patients identified by the Calman-Hine report, should also be available to teenagers and young adults (13 to 25 years) with malignant disease. Hence there are eight Teenage Cancer Units around the UK, with another 12 Units planned or being developed to provide a `user-friendly' physical environment and a concentration of expertise. These Units encourage a philosophy and practice of management, which enhances both `life chances' and quality of life for patients (Senate Community Affairs References Committee Secretariat, 2005, p114-115; Whiteson, 2003). Geehan, a survivor of childhood cancer reflects that AYA find it "immensely valuable to be surrounded by others of a similar age, all fighting the same thing" and not having "to try and explain how you feel all the time is quite a relief." The reciprocal inspiration and support … "procures a real sense of family, both amongst the patients themselves and also amongst the patients' families who can draw on the same special reserves of understanding from the parents of other patients on the ward" (Geehan, 2003; Senate Community Affairs References Committee Secretariat, 2005).

In addition, adolescent hospital units provide significant benefit to the advancement of knowledge, and provide appropriate research milieux for research students studying adolescents. These units are also beneficial to the staff because of the job satisfaction enjoyed, while the adolescents are happier and more comfortable in a developmentally appropriate environment (Tebbi et al., 1983). Grouping AYA in this way would also make it easier to study and monitor these young people more easily.

iv. Clinical Trials and Protocols

Pentheroudakis (2005) reports that "enrollment in clinical research trials and close follow-up via the development of a co-operative infrastructure are imperative for the optimisation of management and avoidance of late effects". Similar to geriatric and

paediatric oncology, Pentheroudakis et al. call for the intensification of treatment, support and research multidisciplinary efforts in order to better fulfil the pressing demands of the adolescent and young adult patient group (Pentheroudakis et al., 2005). To achieve inclusion of AYA into clinical trials, Mitchell suggests the development of a cancer resource network that can provide easily accessible information on current clinical studies for paediatric and adult oncologists, other specialists, and AYA and their families (Mitchell et al., 2004). Since the medical and psychosocial issues for AYA are intertwined, (Hamburg, 1982, p439) it is important that the formula for care include the psychosocial needs of the patient and their carer/family.

v. Psychosocial Support for families and caregivers

Psycho-oncology has developed since the mid-1970s and is one of the youngest subspecialties of oncology. It is one of the most clearly defined subspecialties of consultation-liaison psychiatry, and is a broad multidisciplinary application of the behavioural and social sciences. Psycho-oncology history has produced a model where the psychological domain has been integrated, as a subspecialty, into the disease-specific specialty of oncology and contributes to the clinical care of patients and families, to the training of staff in psychological management, and to collaborative research that ranges from the behavioural issues in cancer prevention to the management of psychiatric disorders and the psychosocial problems during the continuum of the cancer illness, including end-of-life care (Holland, 2003).

Geehan recommends that, "when focusing on care for a defined age group, a far more comprehensive level of care can be offered that attempts to address and maintain, as far as possible, all aspects of a 'normal' teenager's life, whilst simultaneously fighting an illness in a highly supportive and positive environment" (Geehan, 2003).

Rapid advances in medical technology have resulted in increased survival of younger children with chronic illness. As a result, growing numbers of adolescents and their families need psychosocial services and this calls for further efforts by health professionals and social workers to address the needs of chronically ill adolescents (Ell et al., 1990). Stiller reports in 2002 that "there have been no studies of outcome in relation to patterns of organization of care exclusively for adolescents with cancer..." (Stiller, 2002). Perhaps it is time that such studies were conducted.

vi. Consumer involvement

As consumers, AYA patients and their families need to be involved in the planning of how services and support are to be delivered. Young people have clear views regarding the nature of services they would like to see provided, and their preferences for care must be taken into account in developing future services (Ministry of Health, 2002; Ministry of Youth Affairs, 2002; Nebrig et al., 2004; Oppong - Odiseng et al., 1997).

In summary, until the gap in care has been bridged for AYA with cancer or blood disorders through the development of a specialty medicine that can address the above concerns, the health system is likely to continue failing them by denying them inclusion in clinical trials and in the provision of timely, well co-ordinated and developmentally appropriate care, facilities and psychosocial support by an appropriately trained multi-disciplinary team.

Similar to geriatric and paediatric oncology, Pentheroudakis et al. calls for the intensification of treatment support and research multidisciplinary efforts in order to better fulfil the pressing needs of cancer patients (Pentheroudakis et al., 2005).

1.1.1.4. Advances in adolescent and young adult research

Since America declared "war on cancer" in 1971 with an "unprecedented expansion of the US National Cancer Institute" (Elwood et al., 2002) and the WHO defined Cancer Control as "a systematic approach to the reduction of the burden of cancer" (World Health Organization, 1995), many countries have followed suit in developing their own cancer control strategies. For example, Australia's National Cancer Control Initiative (National Cancer Control Initiative et al., 1997) and the New Zealand Cancer Control Strategy (New Zealand Ministry of Health et al., 2003) provide substantial foundations for reducing the cancer burden. Furthermore, the relatively newly established NSW Cancer Institute has since developed the NSW Cancer Plan "to provide optimal cancer management for <u>all patients requiring care</u>" (Cancer Institute NSW, 2004), but unfortunately this crucial document does not recognise AYA with cancer or the paediatric population of cancer sufferers. However, the Senate enquiry on cancer (Senate Community Affairs References Committee Secretariat, 2005, p114) has since made recommendations on cancer care for adolescents. For example, the committee recommends that:

- "Cancer Australia consider the development of appropriate referral pathways that take account of the particular difficulties confronted by adolescents with cancer" (Recommendation 31);
- "State and Territory Governments recognise the difficulties experienced by adolescent cancer patients being placed with inappropriate age groups and examine the feasibility of establishing specialised adolescent cancer care units in public hospitals" (Recommendation 32); and
- In regard to improved data collection: "the committee recommends that Cancer Australia, in consultation with State and Territory Governments and the Australian Institute of Health and Welfare, take a leadership role in coordinating the development of a national approach to the collection of cancer staging data" (Recommendation 33).

In NSW, the Transition Care for Young People with Chronic Childhood Illnesses (TCYPCCI) group (Greater Metropolitan Clinical Taskforce, 2004) was established in December 2002 to identify the issues arising during the transfer of care from the paediatric setting to adult health care services, and develop a state-wide strategy to address transition (Bennett et al., 2005). It was evident that a co-ordinated Sydney-wide network of centres interested in the management of thalassaemia (a blood disorder) patients in particular, needed to be developed due to the complex problems experienced by adolescents from a range of specialty and special needs groups. Hence, the Greater Metropolitan Clinical Taskforce GMCT, part of the Greater Metropolitan Transitional Taskforce (GMTT) which implements health policy, has since developed guiding principles for the transition of AYA from paediatric to adult services, a generic framework and toolkit for essentially all illness groups, appointed young people as consumer participants and completed an extensive search into different models for providing transition services. To oversee this initiative GMCT appointed a Program Manager (May 2004), and three Transition Coordinators (October/Nov 2004) based at Royal Prince Alfred, John Hunter and Westmead Hospitals to work closely with the tertiary paediatric units within the three state-wide paediatric networks.

The initial focus is on identifying gaps in transition services and on working closely with clinicians, young people and their families to determine what is needed. Furthermore, they have appointed young people as consumer participants, and

performed an extensive search into different models for providing transition services (Greater Metropolitan Clinical Taskforce, 2004). In addition to this coordinating role and framework, a minimum data set (MDS) is being developed to help enable the monitoring of this patient group.

Another development is the establishment of a database of available clinical trials to make it easier for doctors to include their AYA patients in trials. This initiative (established in December 2003) is through the Peter MacCallum Cancer Centre (PMCC) in close collaboration with cancer services at other metropolitan hospitals and regional centres throughout Victoria and Tasmania. The PMCC is also developing best practice guidelines that will have a national impact on care for patients aged 15 to 30. This is Australia's first co-ordinated and integrated cancer program for AYA, which focuses on treatment and protocols.

In addition to the PMCC, an Australian NHMRC funded project is underway to find out about the needs of teenagers with cancer and the needs of their parents/carers. The aim is to develop a reliable and valid measure of patient perceived needs to assist in the development and implementation of an appropriate intervention strategy to improve future health care provided to teenagers with cancer. The project asks teenagers and parents/carers for their ideas about what they would most like help with during and after cancer treatment. The researchers involved are from the University of NSW (Dr Anthony Shakeshaft), the University of Newcastle (Professor Rob Sanson-Fisher) and the Cancer Council NSW (Associate Professor Afaf Girgis).

However, while the above milestones indicate a reasonable starting point, AYA with cancer continue to present "an underestimated challenge that merits specific resources, solutions, and an international focus" (Bleyer, 2002a). Resources should also be devoted to "educating the public, health professionals, insurers, and legislators about the special needs of adolescent and young adult patients with cancer" (Bleyer, 2002b). Collaboration with the Education Department and employers' organisations would also be essential. Furthermore, opportunities for clinical research and improvements in outcome require a focused approach and questions specific to this AYA demographic (Barr, 1999; Bleyer, 2002b; Newburger et al., 2002). Hence, it is from this need to reduce the burden of illness that my research developed.

C h a p t e r   2

2. My contribution to improve health outcomes for AYA

The purpose of my research is to inform policy development and services for AYA and increase the research and knowledge base about New Zealand (Ministry of Health, 2002; Ministry of Youth Affairs, 2002, p35; Sarzin, 2003) and Australian AYA with the aim of achieving comparable survival outcomes to those of other age-groups, as well as improved health and development outcomes for AYA living with cancer. It is similarly envisaged to help globally.

My research dovetails well with the work mentioned in Chapter 1, e.g. the GMCT Transition Care, the PMCC and Shakeshaft, and provides validated HRQOL instruments, as well as AYA and proxy versions of measures for satisfaction, preferences for different types of services, and an impact report on the impact of illness and treatment on AYA goals and relationships, and that of their nominated proxy.

Since AYA who are ill have many complex issues and needs that are different to other age groups I devised the Elizabeth Ewing Memorial Framework (Figure 2-1) to provide a global view of what needed to be done and the dynamics involved. The instruments form some of the important elements of the Support Services component of the Framework. In addition to the Framework I developed a Conceptual Diagram (the Elizabeth Ewing Memorial Conceptual View of an AYA & Family Centred Care Model EEMCV/AFCCM, see Figure 2-2) to assist in how I could best help achieve my objective. The Framework and Conceptual View (detailed below) helped me identify essential stakeholders for collaboration and provided direction for achieving my goal.

The Elizabeth Ewing Memorial Framework (EEMF), figure 2.1, is a 15-year plan that provides a structure for implementing my strategy for improving health outcomes primarily for AYA with cancer and blood disorders, but with a view that it could be applied to other illness groups. I am (currently at year 8) working systematically through this framework to target the root of the problem underlying the poor health status and outcomes of AYA patients i.e. limited acknowledgement and recognition in each of the seven (7) components of the EEMF. To date gaining recognition, policy

development, writing submissions and continuous advocacy have been my main focus prior to the planning of this thesis that was devised to develop age-appropriate measures and data as indicated in the EEM Conceptual View. The adolescent psyche and issues of AYA is an area of specialization globally that is only awakening. I felt that through encouraging and promoting research into the psychosocial needs of AYA firstly through the Elizabeth Ewing Memorial Scholarship in Youth Health at the University of Auckland School of Medicine Foundation, and secondly through my research, that health professionals would develop a better understanding of AYA patients as the number of health professionals trained in youth health increased, and this would equate to more health professionals advocating on behalf of AYA. It is expected that researchers of AYA with chronic illness and complex needs, would firstly need to understand the issues and concerns characteristic of the usual turbulent phase of adolescence before trying to overlay the impact of the additional difficulties, concerns and uncertainty associated with for example a cancer diagnosis.

Figure 2-1 Elizabeth Ewing Memorial Framework for improved AYA Health
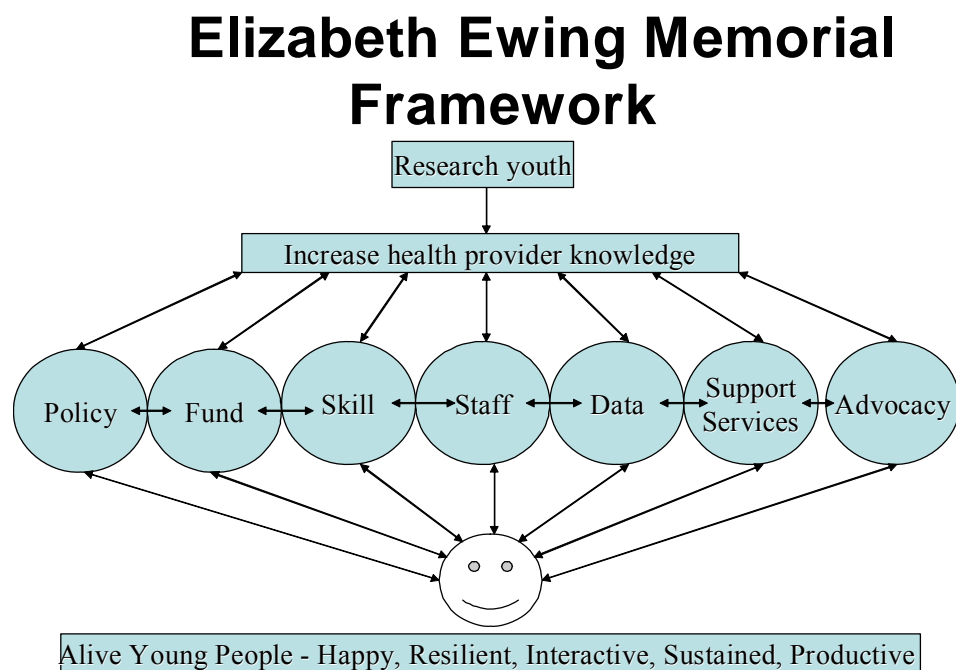
Auckland District Health Board considered the Elizabeth Ewing Memorial Fund as:

> "a long term solution to raise the standard of care for adolescents and young adults. We know very well that the health needs of adolescents and young people are sorely neglected. We also know from experience in other fields that when health professionals enter the work place with specialist knowledge of a

particular consumer group things begin to change inexorably for the better. At the other end of the age spectrum, for example, we are seeing a real change in the care of the elderly as the number of trained geriatricians increases. Specialist training in adolescent health will make a difference..."

[Halbert et al personal communication, 1999]

To achieve better AYA health and well-being outcomes, a co-ordinated and collaborative approach is required between each of the seven (7) components:

1. **Legislation & policy.** Government Departments, local and international organisations and NGOs need to work collaboratively and harmoniously to rectify current breaches of the UNCROC (both Australia and NZ are signatories). In NZ, for example, numerous AYA patients are denied their right to Sickness Benefits and Allowances because they are not informed of their eligibility, and the Social Security Act provides no discretionary power to permit payments retrospectively.

2. **Funding.** AYA need to be visible in the Health Funding Formula, and research funding must be more readily available for investigating the needs of AYA with cancer and other chronic illnesses, and in providing infrastructure for an AYA medicine specialty, staff training, suitable facilities, and data collection.

3. **Knowledge and Skills.** The knowledge base about the psychosocial needs of AYA patients and their families requires constant updates and on-going development.

4. **Advocacy & dissemination of research.** The argument for the AYA cause is on going. It must be supported by more AYA research and needs to be strategically positioned in the public domain to attract suitable funding.

5. **Data Collection** needs to be appropriate (e.g. 12-25years) to ensure ease of identification of the needs for this age-group and for monitoring progress.

6. **Staff.** Active collaboration between multidisciplinary health professionals from adult health care services, paediatric health care services and specialised adolescent health professionals is required to ensure AYA needs are better catered. Health professionals specialising in AYA care are usually suitably equipped and able to relate and cater to the unique needs of AYA, and enjoy caring for them. Their communication skills need to be very good.

7. **Support Services** need to be accessible, informative, appropriate, timely and well co-ordinated to help AYA patients and their families cope.

Accountability and responsibility for appropriateness of information and service delivery is essential. If these components mesh together well in the short-term, the development of an AYA health specialty with appropriate facilities and specially trained staff could become a reality a little sooner for New Zealand and Australia.

My research (commenced in 2002) complements that of Mitchell, Shakeshaft and GMCT as it focuses on the provision of appropriate support services through research that develops age-appropriate measures for: HRQOL, preferences for support services, satisfaction, needs assessments and impact of illness and treatment, through parallel AYA and proxy-carer surveys. My research was formulated initially: while writing the Elizabeth Ewing Report 1998 to Auckland District Health Board (ADHB); during my involvement with the NZ Cancer Control Strategy Workshop 1999 and subsequent stages of development of the NZ Cancer Control Strategy; when writing a presentation to the Health Services Development Plan (HSDP) Manager, Ian Wolstencroft to gain recognition of adolescents as a separate special needs group within ADHB, then helping to develop the HSDP's policy on Adolescent Issues 2000; as a member of the External Reference Group for the NZ Ministry of Health and NZ Ministry of Youth Affairs to develop the policy on youth health i.e. *Youth Health: A Guide to Action 2002* (Ministry of Health, 2002) and in my submission for the Youth Development Strategy Aotearoa (Ministry of Youth Affairs, 2002); during the writing of my thesis literature review; and finally through my preparation of various publications e.g. on awareness of the adolescent patient group (Ewing, 2000), and asking whether we are really doing enough for young people with chronic illness and complex needs (Ewing, 2003), and conference presentations in both NZ and Australia.

My research supports the NSW Youth Health Policy, *Young People's Health: Our Future*; NSW Health's *A Clinical Service Framework for Optimising Cancer Care in NSW*, 2003 (NSW Department of Health, 2003); the NZ Ministry of Health's *Youth Health: A Guide to Action*; the NZ Ministry of Youth Affairs' *Youth Development Strategy*; Sporle's Pilot *Survey of Auckland Adolescents' Perception of their Health Needs* (1993), and various reports and reviews e.g. *Through the eyes of the child*, the

NZ Paediatric Services Review, the NZ Cancer Services Review, and NSW Access Study for Young People.

2.1. My Research

My research supports the notion of a specialised AYA patient centred care model and develops instruments (as depicted in the conceptual view Figure 2.2) for use in a two-phase programme. This thesis represents the first step, where I describe the development, field-testing and validation of AYA-specific HRQOL instruments and develop a feasible recruitment strategy. As mentioned data were also collected for measuring Preferences for Support Services, Patient Satisfaction, and an Impact Report, but the analysis of these measures are not included in this thesis. However, once all of these instruments have been validated, the proposed second phase of research can commence.

The proposed second phase of research is to help facilitate the development and potential delivery of developmentally appropriate quality health care services and better health outcomes for AYA by:

  i. Documenting support services currently used

 ii. Measuring how well support services meet their needs

iii. Identifying ways that services could be improved to meet unmet needs

iv. Establishing baseline data for monitoring and evaluating the impact of changes in the design and delivery of services; and

 v. Providing an information platform to assist clinicians in forming partnerships with this patient group.

To fulfil the above aims I was permitted by the author to modify and use the currently existing PedsQL Measurement Model, and to use the Memorial Symptom Assessment Scale. Author permission was also gained to develop an AYA Patient Satisfaction questionnaire based on a parent-reported version. The instruments for measuring AYA and proxy Preferences for Support Services, and Impact of illness and treatment were developed specifically for this study, and will be analysed and described in later publications. All instruments are described in detail in Chapter 3.

A conceptual view from the perspective of the AYA patient and their families is demonstrated in Figure 2.2. It indicates several rings (or layers) requiring collaboration for achieving developmentally appropriate care for AYA and the instruments required for measuring and monitoring these key elements.

Figure 2-2 Elizabeth Ewing Memorial Conceptual View of AYA & Proxy Health

The EEM Conceptual View from the perspective of the AYA with complex needs
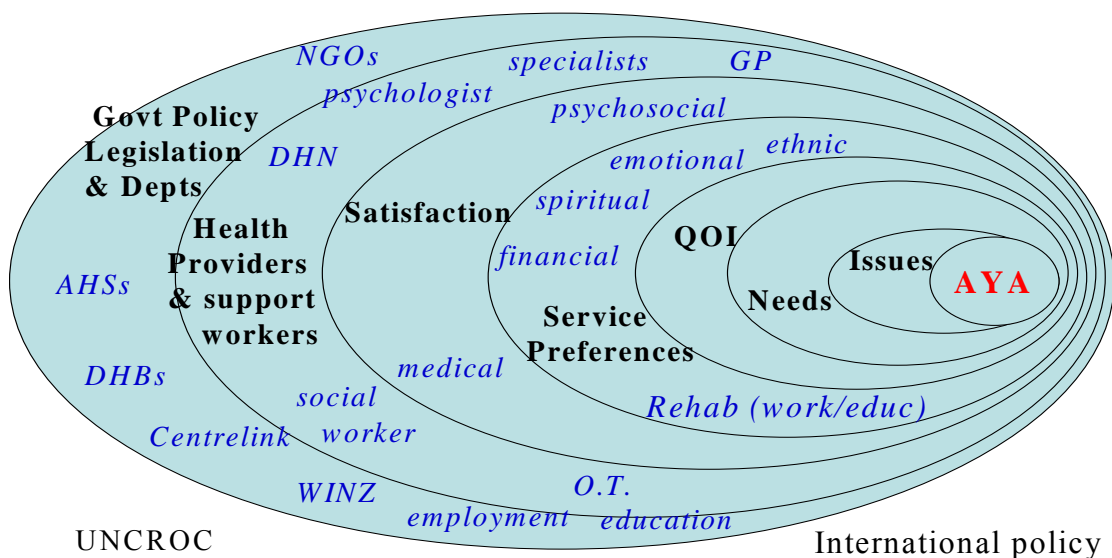
Central to the EEM Conceptual View is the AYA (and Proxy or family) centred care and the degree to which the AYA patient feels connected to their family, peers, school/work colleagues and community (Ministry of Youth Affairs, 2002).

The first layer enveloping the AYA contains the issues and concerns peculiar to AYA, while the second layer is a measure of their needs, and those of their nominated proxy. The third layer surrounding the AYA is a measure of their HRQOL and encompasses their physical functioning, emotional functioning, social functioning, role functioning (i.e. study/work), and more specifically for cancer patients it also measures pain, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance, sexuality, fertility and communication. HRQOL is the main focus of this research.

The fourth layer constitutes the preferences for support services that the patient and their proxy may have for different types of support service i.e. ethnic/cultural, emotional, spiritual, financial, rehabilitative support (back to study of work) and psychological support services.

The fifth layer consists of the AYA and proxy's measure of satisfaction i.e. the level of family inclusion, emotional needs, communication, information, and technical skills (which will be the subject of a future paper).

The sixth layer indicates a measure from the perspective of the health providers and support workers of AYA service delivery, since their views are likely to have influenced the AYA/proxy responses in the earlier layers and the patient and carer health and well-being outcomes.

The seventh layer comprises the AYA specific government policy, legislation and departmental goals and objectives at local, state and national level that dictate the emphasis of health system delivery through budgetary allocations.

Finally, encompassing all inner layers is the influence of international policy in relation to AYA e.g. the United Nations Convention on the Rights of the Child (UNCROC), though this policy is effective for 16-18 year olds only, of the target group.

Although my validation study covers NSW, the proposed substantive study is planned to include NZ, where the population is similar and there is good potential for collaboration.

Since the focus of this thesis is on HRQOL, I now focus on the tools for measuring this aspect of care. The methods are outlined in Chapter 3, the results of the analysis in Chapter 4, and the conclusions in Chapter 5, which also discusses, W*here to next in the bigger AYA picture?*

2.2. Quality of Life

2.2.1. What is quality of life?

There are a number of definitions of quality of life (QOL) and of health, with some linking the two, but no universally accepted definition has been agreed upon. The World Health Organisation (WHO, 1948) defines health to be "a state of complete physical, mental and social well-being, and not merely the absence of disease" (Fayers et al., 2000 p3). QOL has different meanings to different people and in different contexts, and different meanings to the same people at different times and in different situations.

The term "health-related quality of life" (HRQOL) is usually used to differentiate between the general sense of quality of life and that which refers to a more medical or health-related sense. Since a more formal agreement on a definition has not been reached, investigators usually provide a definition for HRQOL peculiar to their research question, which is then reflected in the emphasis of the questionnaire items. However, according to Fayers and Machin, it is generally agreed that HRQOL studies can include general health, physical functioning, physical symptoms & toxicity, emotional functioning, cognitive functioning, role functioning, social well-being and functioning, sexual functioning, and existential issues (Fayers et al., 2000 p3). HRQOL measurement has become a major research field e.g. in 1973 only five HRQOL articles were cited in Medline (Mahler, 2000) but now there are well over 1500 articles.

Some proposed conceptualisations of HRQOL are: the Expectations model (Calman 1984) which defines QOL as the difference between an individual's hopes and expectations and their present experience (Calman, 1984); and the Reintegration to Normal Living (RNL) Index connects the patient's ability to do as they should or want to do though they may not be free of disease or symptoms (Wood-Dauphinee et al., 1988). Other models emphasising the Meaning in Life (Scale) as called for by specialists as this aspect of life is not adequately measured by HRQOL instruments (Warner et al., 1987), Satisfaction with Life (SWL) as a whole, through the SWL Survey (Pavot et al., 1993), while patient needs can potentially clarify the types of interventions needed to address specific areas of dysfunction (Coyle et al., 1996), the inclusion of spiritual well-being in a bio-psychosocial-spiritual model for QOL measurement in oncology, (Brady et al., 1999), the financial burden of illness (Cohn et

al., 2003), and finally the power differential between the patient/family and the health professionals who inherently carry the weight of the medical establishment (Chesler et al., 1987). For people who are sick, improved health may be the means to better quality of life (Fayers et al., 2000), and for those with incurable and or chronic illness, co-ordinated, appropriate and timely services and support may be a means to better quality of life. While some HRQOL instruments may investigate only one aspect, usually HRQOL instruments are considered to be a multidimensional construct and include at least some items that target physical, emotional and social functioning.

HRQOL assessment is subjective and therefore it is preferable that patients complete self-reports on their own HRQOL (McColl et al., 2005, p139). The judgment of proxies in reporting on a patient's HRQOL, whether it is their doctor, other healthcare staff or other people (relatives or friends of the patient), is often substantially different to that of the patient's self-assessment. However, in some circumstances a proxy may be required to make an assessment of patient-HRQOL in cases where patients are too ill, too young, or mentally disabled (Fayers et al., 2000, p11). Although this is not an ideal situation, proxy-reports have been found to give a reasonable indication of patient HRQOL if necessary (Fayers et al., 2000 p4).

2.2.2. Why measure HRQOL?

HRQOL has become an important outcomes measure in cancer and there are specific issues depending on the site, stage, treatment and patient age. Essentially HRQOL is used to differentiate between people who have a better health status from those with a worse health status for the purposes of health care decision-making (Fayers et al., 2000; Mahler, 2000; McHorney, 1997) in the treatment and care of patients. While medicine has traditionally concentrated on mortality, morbidity and symptom relief as an outcome measure, in more recent times other issues that may impact on the patient's life and be of equal or more importance to them are considered (Fayers et al., 2000, p7). Consequently, standardized instruments have been developed to provide measurement of health status and levels of impairment compared in individuals and in groups of patients to test the efficacy of therapeutic intervention (Mahler, 2000). For example, instruments are used to detect how much HRQOL has changed in response to therapy or compare treatments in the hope of improving patient HRQOL and reducing negative changes such as toxicity and side-effects (Fayers et al., 2000, p7).

Accordingly, HRQOL generic and disease specific instruments continue to be developed and refined. Generic HRQOL instruments permit comparisons across different patient populations for detecting previously unrecognised adverse effects. The disease-specific instrument is more relevant to the individual patient's medical problem and different stages of disease (Mahler, 2000). HRQOL has become recognised as an important aspect of Randomised Clinical Trials (RCT). The type of HRQOL instrument utilised, depends on the type of RCT and the aspects of HRQOL being examined. For example, the focus of a RCT might be treatment with a curative or palliative intent, or for improving symptom relief, care or rehabilitation, or perhaps researchers may wish to 'establish information about the range of issues and concerns that affect patients'. This is organised to help future patients to 'anticipate and understand the consequences of their illness and treatment' (Fayers et al., 2000 p11). Furthermore, self-reported HRQOL of long-term cancer survivors may be overlooked because the survivors, contrary to expectation, may have continuing problems (both physically and psychologically) long after completion of treatment, called 'late effects'. Consequently, investigators choose the most appropriate tool in conjunction with their research goals, from the variety of HRQOL instruments now available. But unfortunately, at the time of writing there were no HRQOL instruments available with a specific AYA focus on cancer.

HRQOL measures can be useful in medical decision-making as a predictor of treatment success and predictive of survival in clinical trials with a curative or palliative intent; improving symptom relief, care or rehabilitation; facilitation of communication with patients; and 'late-effects' (Fayers et al., 2000, p7-13). HRQOL scores may reflect an early patient perception of disease progression and therefore would be predictive of outcome. Or, HRQOL status may influence the course of disease in some way, in which case interventions to improve HRQOL could become an active form of therapy to enhance patient outcome. Preoperative HRQOL can be predictive of recovery and changes in HRQOL during treatment can have prognostic value (Fayers et al., 2000, p14).

Although numerous instruments are available for cancer HRQOL, most are designed for adults, some for children, but none for adolescents and young adults (AYA) who may

have special concerns and have poor improvements in survival compared with other age groups. Thus, HRQOL instruments peculiar to the special needs of AYA need further study and development.

Accordingly, my thesis establishes the choice of instrument (section 2.4 and 3.1) and the reliability, validation and sensitivity (sections 4.2, 4.4 and 4.5, respectively) of a modified version of the PedsQL™ Measurement Model for use with AYA in the area of cancer and blood disorders and establishes baseline data for this patient group.

2.3. Cancer and Blood Disorders and their effect on AYA

Cancer

The NSW Cancer Council defines cancer as a disease of the cells, which are the body's basic building blocks. "Our bodies constantly make new cells to enable us to grow, to replace worn-out cells, or to heal damaged cells after an injury. Certain genes control this process. Normally, cells grow and multiply in an orderly way. However, damaged genes can cause them to behave abnormally and they may grow into a lump (tumour)" (Cancer Council NSW, 2005).

Tumours can be benign (not cancerous) or malignant (cancerous). Benign tumours do not spread outside their normal boundary to other parts of the body. A malignant tumour is made up of cancer cells. When it first develops, this malignant tumour may be confined to its original site. This is known as a cancer in situ (or carcinoma in situ). If these cells are not treated, they may spread beyond their normal boundaries and into surrounding tissues, becoming invasive cancer" (Cancer Council NSW, 2005).

There are a number of different types of cancer affecting AYA, for example, acute lymphatic leukaemia (ALL) is a type of cancer of the blood, involving a malignant proliferation of the white blood cells. ALL represents 35% of paediatric oncological disease, and is the commonest cancer in children. Eighty-five percent (85%) of leukaemias in children are ALL; the remainder is mainly acute myeloid leukaemia (AML). ALL is slightly more common in males than in females and is rare in the first year of life and peaks at 3 years (Cancer Council NSW, 2005; GE Healthcare, 2005).

Ewing Sarcoma also known as Peripheral Neuroectodermal Tumour (PNET) is a class of disease that arises from very primitive cells in the body. Although this is usually thought of as a bone tumour, Ewing Sarcoma is increasingly recognized to arise in soft tissues in the body as well. Osteosarcoma (also known as osteogenic sarcoma) is a tumour of cells, which forms bone. (Virtual Cancer Centre, 2004)

My study does not differentiate between the many different types of cancer, but the side effects of treatment are fairly similar e.g. nausea, so I will not elaborate on the different forms of cancer but keep the discussion general.

Blood disorders

A blood disorder is a disturbance of the normal working of the blood. Like cancer there are many different types of blood disorder. The level of severity, and degree to which the patient and their HRQOL are affected can vary considerably from patient to patient.

Thalassaemia is one example of the many different types of blood disorder. It was singled out on request of Dr Robert Lindeman of POWH. Thalassaemia is the name of a group of genetic blood disorders where the red blood cells do not form properly and cannot carry sufficient oxygen. Haemoglobin, the oxygen-carrying component of the red blood cells consists of two different proteins, alpha and beta proteins. If the body doesn't produce enough of either of these two proteins, the result is anaemia, which begins in early childhood and lasts throughout life. There are various types of Thalassaemia, which affect the human body in similar ways. In the most severe form of beta Thalassaemia, *Thalassaemia Major* or *Cooley's Anaemia*, complete lack of beta protein causes life-threatening anaemia that requires regular blood transfusions and extensive ongoing medical care. The resultant iron-overload in turn requires chelation therapy to prevent early death from organ failure (Cooley's Anemia Foundation, 2001).

2.3.1. How cancer and blood disorders affect the HRQOL of young people.

In addition to the common risk factors associated with AYA, it is important that AYA are helped to maintain or develop their sense of connectedness with their peers, families, school/workplace and community as this determines their resilience (Ministry of Youth Affairs, 2002). AYA with chronic health problems, such as cancer and blood disorders often experience delays in the usual developmental tasks of adolescence, such

as identity and autonomy as they may need to revert to dependence on parents for care and support at a time when they are pushing their boundaries and asserting their independence. Instead they may find they are unable to keep up with their peers and do the kinds of things other young people do such as study, work and going out, due to fatigue (Edwards et al., 2003), lack of stamina, forgetfulness and concentration. They do not wish to be seen as different, but treatments causing such things as nausea, vomiting, pain, alopecia and swelling can damage their self-confidence through changes in appearance and energy loss causing them further anxiety, worry and opting out of many activities when it simply becomes too difficult, and potentially becoming non-compliant (Bleyer, 2001; Ellis, 1991).

Furthermore, another crucial aspect of psychosocial need for AYA with cancer is understanding the implications that cancer treatment has on sexuality and fertility, both areas of which also remain poorly researched across the cancer field with most work focussing on females with breast cancer (Katz, 2005).

2.4. Instruments for measuring HRQOL in young people

2.4.1. HRQOL instruments

The HRQOL instruments potentially suitable for the age range of my study (see Table 2-1) were the Child Health and Illness Profile –Adolescent Edition (CHIP-AE), Dartmouth COOP Picture Charts (COOP), Health Utilities Index (HUI2), PedsQL Measurement Model, SF-36 Health Survey and the Quality of Life Profile Adolescent Version (QOLP-AV). All except the PedsQL Measurement Model either did not include items covering the issues as mentioned in the last section, or were too long and cumbersome, or did not have a proxy version which is also important particularly when a patient is unable to self-assess their HRQOL. For example, concerning the inclusion of issues, the SF-36 (Ware et al., 1992) used world-wide on populations from as young as 14years old, provides a wide lens on some of the major areas of life, but overlooks the importance of, for example, an AYA's connectedness to peers and rehabilitation back to work and/or study.

However, the flaw in the PedsQL Measurement Model is that it has overlooked for adolescents the issues of sexuality and fertility, which are very relevant to the

assessment of HRQOL in AYA, and arguably so in my sample which included some AYA patients who were married. This was one of the challenges in using a paediatric instrument, but nevertheless to use an adult focussed tool would have meant losing the other aspects inherent in AYA. It was therefore decided that additional items to address these issues could be included and tested in the substantive study.

Table 2-1 Paediatric Health-Related Quality of Life Instruments

Instrument	Age group	Respondent	Recall period	Response options	Completion time (min)
CHIP-AE	11-17	Self & parent	Past 4 weeks	Uses 3-5 mixed points	20-30
COOP	12-21	Self	Past month	5 points only	10-15
HUI 2-18	2-18	Self & clinician/ parent proxy	Present	Uses 3-5 statements	10-15
PedsQL	2-18	Self & parent	Past month	5 points only	8
SF-36	14 +	Self	Past 4 weeks	Up to 6 points	5-10
QOLP-AV	14-20	Self	Unstated	5 points only	10-15

Source: Peter Fayers, Ron Hays Assessing quality of life in clinical trials 2005

In addition to the above instruments the Nottingham Health Profile, Sickness Impact Profile, the Medical Outcomes Study, and the Sickness Impact Report were also considered.

However, it was apparent that the PedsQL Measurement Model was the most appropriate for use with my target age-group, the treatment side effects were covered adequately, and it was short, easily understood and needed the least modification.

The important features of the PedsQL Measurement Model were its domain in the Generic Core about role functioning that focused on school, due to education being of importance for AYA aged patients; and the inclusion in the disease-specific module of domains such as perceived physical appearance, and problems with communication. Also the disease-specific module complemented the generic core without repetition (see the next section for more detail, and the appendix for the modified instruments).

In addition, Varni and his team from San Diego have been researching, developing and refining HRQOL for children and young people for more than 15 years. Over this period the PedsQL measurement model has been rigorously tested for validity, reliability, responsiveness and sensitivity as a modular approach to measuring HRQOL in children and adolescents (Laffel et al., 2003; Meeske et al., 2004; Sawyer et al., 2004; Seid et al., 2000; Skarr et al., 2002; Uzark et al., 2003; Varni, Burwinkle, Jacobs et al., 2003; Varni, Burwinkle, Seid et al., 2003; Varni et al., 2004; Varni et al., 2002a; Varni, Seid, Knight, Burwinkle et al., 2002; Varni, Seid, Knight, Uzark et al., 2002; Varni et al., 2001; Varni et al., 1999).

2.4.2. The PedsQL$^{\text{TM}}$ 4.0 Measurement Model

PedsQL$^{\text{TM}}$ 4.0 Measurement Model for HRQOL was designed to measure the core dimensions of health, as defined by the World Health Organisation (WHO, 1948) i.e. Physical, Emotional, and Social Functioning, as well as Role (school) functioning for HRQOL in children and adolescents. The PedsQL$^{\text{TM}}$ condition-specific modules complement the generic core scales for use in designated clinical populations integrating seamlessly both generic core scales and disease-specific modules into the one measurement system. Its purpose is to measure HRQOL in healthy children and adolescents, and those with acute and chronic health conditions. It is brief, practical, flexible and relevant to children and young people. It has structured and fixed questionnaires with questions of a multidimensional nature, and fixed response scales (Varni et al., 2002a; Varni, Seid, Knight, Uzark et al., 2002; Varni et al., 2001).

The PedsQLT$^{\text{M}}$ Measurement Model contains several different instruments that are sensitive to the cognitive development of children and young people aged 2-4, 5-7, 8-12 and 13-18 with self-report beginning at 5 years old.

The items included in the instruments for 13 to 18 year olds were chosen by Varni and his team to emphasise the adolescent patient's perceptions of HRQOL while the items of the proxy-report were constructed to directly parallel the adolescent self-report items to measure the proxy's perception of their child's HRQOL. Hence, the instruments comprise parallel self-report and proxy-report forms for both the generic and disease-specific measures differing only in first or third person tense (Varni et al., 2002a; Varni,

Seid, Knight, Uzark et al., 2002; Varni et al., 2001). The PedsQL™ condition-specific modules complement the generic core scales for use in designated clinical populations.

In addition, Varni's instruments have been translated into different languages that have also undergone rigorous testing for validation. Furthermore, Varni and his associates have also developed disease-specific modules (apart from cancer), which complement the generic core, for use with children with heart disease (Uzark et al., 2003), Diabetes type I (Laffel et al., 2003) and Diabetes type I & II (Varni, Burwinkle, Jacobs et al., 2003), paediatric rheumatology (Varni, Seid, Knight, Burwinkle et al., 2002), to name a few.

Varni and his associates developed these instruments at the Centre for Child Health Outcomes, Children's Hospital and Health Center, San Diego. Licence agreements are required for their use, and are available from the MAPI Research Institute, France.

The aim of this thesis was to find an existing HRQOL instrument suitable for modification for AYA, to modify it for them and test its validity, reliability and sensitivity in Australians aged 16-25 years old with a cancer or a blood disorder diagnosis. This thesis describes the modification, field-testing and validation of the PedsQL Measurement Model for AYA patients and their nominated proxies. However, as detailed in the section on the survey pack below, in addition to the HRQOL, I collected data about AYA satisfaction, preferences for services and impact of illness and treatment.

Chapter 3

3. Methods

This chapter describes the instrument modifications for a slightly older group, and the methods I used to field-test and validate the PedsQL Generic Scale and the PedsQL Cancer Module instruments in a sample of 16 to 25 year old Australians diagnosed with cancer or a blood disorder.

3.1. Description of the instruments utilised

3.1.1. PedsQL^{TM} 4.0 Measurement Model

As mentioned in section 2.4, this measurement model combines the PedsQL^{TM} 4.0 Generic Core and the PedsQL^{TM} 3.0 Cancer Module, which complement each other for greater measurement sensitivity of the target group. The treatment side effects are covered adequately, and without repetition. It is a brief, practical, easily understood, valid, reliable, responsive and sensitive modular approach to measuring HRQOL in children and adolescents, with parallel proxy-report versions. Minimal modification was required and is described below.

The PedsQL^{TM} 4.0 Generic Core Scale

The Core instrument comprises 23 items, in 4 domain-specific scales of functioning: (1) Physical, (2) Emotional, (3) Social, and (4) School. In addition, it also generated a Total Scale Score, a Physical Health Summary Score and a Psychosocial Health Summary Score (Varni, Burwinkle, Seid et al., 2003; Varni et al., 2002a cited in Fayers 2000 p107; Varni, Seid, Knight, Uzark et al., 2002; Varni et al., 2001; Varni et al., 1999).

The PedsQL^{TM} 3.0 Cancer Module

I used the *PedsQL^{TM} Cancer Module*, version 3.0 (Varni et al., 2002a) in addition to the generic score scale instrument (PedsQL^{TM} 4.0). The Cancer module comprises 27 items, on the following domain-specific scales: (1) Pain and hurt, (2) Nausea, (3) Procedural Anxiety, (4) Treatment Anxiety, (5) Worry, (6) Cognitive problems, (7) Perceived Physical Appearance, and (8) Communication (Varni et al., 2002a; Varni et al., 1999).

3.1.1.1. PedsQL Instrument modification

Author permission was granted to make minor adjustments (see Table 3-1 below) to the instrument's wording (Varni, personal communications 2002, 2003). Nine of the 23 Generic Core items and one of the 27 Cancer Module items were altered for relevance to the slightly older age bracket i.e. AYA 16 - 25 years old as opposed to the original version for adolescents 13 -18 year olds.

Further to the usual domains of any QOL instrument, namely, physical, emotional and social functioning, the issues of adolescents and young adults lead to the need for a further domain about school functioning. Varni's instruments provided this additional domain and I was permitted to adjust it to "work/school functioning" in order to capture the appropriate measure for the slightly older age group who could be engaged in any combination of study and work or neither studying nor working. Under the social domain – "(Problems with…) How I get along with other teens", I was permitted to use the term 'young people'.

Table 3-1 Alterations to wording in PedsQL as approved by author

Original terms	Replacement terms	Domain & item numbers	Approval dates
Ages 13-18	Ages 16-24 (in the title)	N/A	10 Feb 2003
Teen or teens	Young person or young people	Social Functioning: items 1 to 5	29 Oct 2002
	or young adult(s)		10 Feb 2003
School	Study/Work or Study	Work/Study Fn: item 4 & 5. Cognitive Problems: item 3	10 Feb 2003
Class	Class/at work	Work/Study Functioning: item 1	10 Feb 2003
Schoolwork	Study/Work duties	Work/Study Functioning: item 3	10 Feb 2003
Child	Child/charge (in covering question)	N/A	10 Feb 2003

3.1.2. Memorial Symptom Assessment Scale (MSAS)

The MSAS is a validated 30-item patient-rated instrument providing multidimensional information about the symptoms experienced in the past week, by people with cancer. It was introduced to classify patients into "known groups" of slight, moderate and severe symptom experience, and had previously been tested for validity and reliability in the

Australian context (Collins et al., 2000; Collins et al., 2002). (See Appendix A for the instrument, and Section 3.4 for more details).

Since the focus of this thesis is on the validation of the PedsQL measure, the instruments additional to this objective (satisfaction, preferences and impact report) are detailed under the Survey Pack (Section 3.3.3).

3.1.3. Socio-demographic and clinical information

The socio-demographic information requested included: ethnic grouping; gender; marital status; level of education; relationship between the patient-proxy dyad (pair); and postcode. In addition, young adult patients were asked whether they were studying, working, or engaged in both study and work.

The clinical information included: the medical condition (whether the condition was on-going, in remission, relapsed, or cured); the treatment received over the past month; and their expected treatment in the following month (see Table 3-2). This information was requested from the young adult patient only, and formed a modification that was introduced in the second version of the survey during field-testing (after the Ethics Committee had approved the amendment) because a better understanding of the patient's health status was required in terms of their illness and treatment.

Table 3-2 Clinical Information about the adolescent & young adult patient

About your Condition and Treatment (Please tick as many as applies to you)				
My condition or disease is …..		**Treatment over past month included:**	**Treatment expected over the next month**	
	On-going	Chemotherapy	Chemotherapy	
		Radiation Therapy	Radiation Therapy	
	In remission	Surgery	Surgery	
		Transplant	Transplant	
	In relapse	Transfusion	Transfusion	
		Chelation	Chelation	
	Cured	No treatment	No treatment	

Patients were asked about their health service utilisation in the last 12 months in terms of the number of overnight hospital visits and emergency admissions, and in the past 30 days: the number of days missed from study/work, fun day activities missed, and days when a carer was required.

Proxies were asked about their: age bracket (18-19; 20-29; 30-39; 40-49; 50-59; 60-69; and 70+); occupation; how the young adult's health had affected their own (work attendance, daily routine, and their ability to concentrate); how many of their workdays were missed due to the patient's physical or mental health.

3.2. Participants and recruitment

The aims of this research required a heterogeneous sample of young adult patients representing a wide range of QOL experiences, and their nominated proxy (see Section 3.6.1). The original plan was to study young people who had been diagnosed with cancer or a blood disorder while in their youth i.e. aged 12 to 24 inclusive. However, I was advised that this would be difficult to recruit to, as most young people with a blood disorder would have been diagnosed long before this age. [B. Caveletto, private communication 2002]

As the number of young adults affected by cancer or a blood disorder is relatively small this study was multi-centred to enable a sufficiently large group to be accrued. Patients were recruited from Sydney Children's Hospital (SCH), the Prince of Wales Hospital (POWH) and the Royal Hospital for Women (RHW). Located on neighbouring campuses, each hospital provides specialist services to Metropolitan Sydney and a wide area of New South Wales and other parts of Australia, as well as the South Pacific region. The Centre for Children's Cancer and Blood Disorders (CCCBD) is a specialist paediatric haematology and oncology unit within the Sydney Children's Hospital. The CCCBD accepts approximately 200 new referrals for childhood cancer and blood disorders each year, with 58% of patients from regions outside the Sydney metropolitan area. Most adolescents with cancer or a blood disorder are referred to services in South Eastern Sydney Area Health Service (SESAHS), and are initially treated by the CCCBD at Sydney Children's Hospital.

After SCH ran security checks on my background, they issued me an identity card and security number for entry to the CCCBD work area. Dr Goodenough introduced me to the SCH clinic and ward staff. A letter of introduction and photograph were required for the staff notice boards (see Appendix D) to enable staff to identify me easily while on the wards or in the clinics. SCH requested that I provide potential SCH participants with a UTS business card and that of Dr Goodenough. Further to this I prepared an article for publication in the quarterly CCCBD Family Newsletter September 2003 to inform patients and their parents about the study (see Appendix D).

3.2.1. Inclusion Criteria

Eligible participants were aged 16 to 25 years old, had either a cancer or a blood disorder diagnosis, could read and write English, and were at least 3 months post diagnosis but not requiring end-of-life care. I delayed approaching recently diagnosed patients until at least 3 month post-diagnosis to give these potential recruits an opportunity to experience available services, and develop an awareness of the kinds of service or support that might be helpful.

3.2.2. Exclusion Criteria

Young adults with co-morbidity such as Down's Syndrome were excluded due to the potential confounding effects from additional conditions not under investigation. Such young adults may also have had difficulty in completing the questionnaires without caregiver assistance and (possibly) influence. Patients receiving end-of-life care were excluded due to their additional complexities and needs, which were beyond the aims of this validation study. Long-term survivors were channeled into the concurrent "Long-term effects" study.

3.2.3. Ethical Approval

I gained approval to recruit patients from the South Eastern Sydney Area Health Service (SESAHS) Human Research Ethics Committee (HREC #03/061) and the University of Technology, Sydney Human Research Ethics Committee (UTS HREC #02/125). Scientific assessment and approval were received from the Scientific Review Committees of SESAHS and Royal Hospital for Women (RHW) separately. Informed assent and consent were gained before recruitment, and participants could withdraw at any time.

3.2.4. Recruitment Procedure

Each hospital required slightly different recruitment procedures and sometimes it was necessary to develop or tailor procedures peculiar to the needs of individual clinicians. Eligible patients at SCH were identified by a member of the medical team in liaison with the Hospital Research Scientist (SCH-HRS). At the RHW, the data manager in liaison with the Departmental Head identified eligible patients on the basis of age. At the POWH there was no composite database so eligible patients were identified by various database managers and/or Clinical Nurse Consultants (CNC) from either small databases or patient records. Permission was then sought from the treating clinician to approach the identified patients to explain the study.

Prior to recruitment at each of the hospitals patients were pre-advised of the study, e.g. at SCH by their treating physician or via publication of the study's commencement in the CCCBD Family Newsletter, while at the POWH and the RHW the treating physician authorised a letter of introduction to be mailed to potential recruits (see Appendix D).

Participants i.e. patient (and proxy dyad) were recruited consecutively by one of two recruitment strategies. If the eligible patient was in a ward or due at clinic, they were recruited at the hospital (method i). If they were not expected to be at the hospital during the recruitment period, they were recruited by telephone (method ii):

i. Clinic/hospital recruitment: Recruitment began at SCH, where I attended relevant oncology and haematology clinics and wards at suitable times to make the first contact with potential participants

After securing their assent, the project was explained, the information sheet provided, any questions were answered, signed consent obtained and a survey booklet (self-report and proxy-report) was provided for them to complete without collaboration. If the patient's nominated proxy was not present, I left the proxy-report information with the patient and visited on another occasion when the proxy was present. Participants who wished to complete the questionnaire booklets at home were asked to include a contact phone number and note convenient times for a reminder or follow-up phone call, on the consent form if required.

ii Telephone recruitment: On telephoning the patient, I identified myself and if the patient was happy for me to continue I proceeded to explain the project as above.

Potential recruits who gave verbal assent were sent the survey pack (see section 3.3.3) by traditional post for their consideration to potentially complete and return. Permission was requested to call again in a couple of weeks to ensure they had received the pack and answer any further questions if required.

Recruitment commenced at SCH in July 2003, and at the RHW and POWH in November 2003, and finished on 31 May 2004.

3.3. Data collection

3.3.1. Information from hospital databases

The information provided by the clinic database manager or CNC about each eligible recruit included the patient's name, their date of birth, illness, contact details (address and phone number) and the name of the doctor responsible for their care. The SCH and RHW were also able to provide with ease, date and age at diagnosis, and date of last follow-up appointment.

3.3.2. Administration of questionnaires

Varni's Guidelines for administration of the QOL questionnaires were followed. For example, in clinics and wards I was able to ensure the patient-proxy dyad did not collaborate on their answers prior to completion. For participants recruited by phone, I could only stress the importance of patients and proxies completing the booklets independently. Varni also specified, that participants recruited at clinic should complete the PedsQL questionnaires prior to their seeing the doctor if at all possible to reduce the possibility of participant responses being influenced by the meeting. Usually these participants completed their questionnaires while waiting in clinic. Inpatients were able to complete their questionnaires at their convenience during their hospital stay, and I would return to collect them.

When two caregivers jointly filled in the proxy-report booklet, the proxy demographic information used was that of the official proxy as specified on the signed consent form.

I prepared the questionnaire booklets and packs for all participants, organised the data entry, entered and checked all data against the original questionnaires, and made follow-up phone calls to maximise recipient response rate and minimise missing data.

Sometimes I needed to mail a second questionnaire pack or an additional booklet, consent form or envelope when participants mislaid these.

For ease, the coding of questionnaire administration and method of recruitment for the proxy was based on that of the AYA patient. For example, if a patient was recruited in clinic and took the proxy materials home for the caregiver to complete and return, both were coded as recruited at clinic at the particular hospital.

3.3.3. Survey Pack including the additional instruments

The survey pack included: a gold questionnaire booklet for the young adult patient; a pale green booklet for the proxy; participant information sheet; consent form; and self-addressed reply-paid envelope. A brief hand written note (on UTS complimentary note paper) was attached to the booklet and as suggested by the SCH-HRS, I included my business card. For potential participants from SCH, the business card of the SCH-HRS was also included.

The questionnaire booklets contained parallel versions of the AYA self-report and proxy-report questionnaire booklets, colour coded gold and pale green respectively. Each booklet contained the self-report or proxy version of the following instruments: the PedsQL Generic Core, PedsQL Cancer Module, Satisfaction Survey, Preferences for Support Services, the Memorial Symptom Assessment Scale (for the patient only), the Impact Report, clinical information (for the patient only) and socio-demographic information (in that order). The instruments not previously described (in section 2.4.2 and 3.1) are detailed in the following subsections.

The satisfaction survey led on from the HRQOL instruments, and was an opportunity for respondents to consider their cumulative experience of the health service in relation to the AYA patient's care. The instrument on Preferences was placed third because it was anticipated that the HRQOL and Satisfaction instruments could assist in triggering respondents' thoughts about the kinds of services that they might find helpful. The ordering allowed participants to complete the questionnaires requiring their perspective first and the fixed information socio-demographic information (the easy parts) last when they were likely to be tired. Also, the socio-demographic and clinical information could be completed at any time or by proxy if need be.

Copies of both the self-report and proxy-report questionnaire booklets are in Appendix A. I was able to include a small gift of appreciation for participants, such as pens, CanTeen tattoos, miniature stapler (courtesy of Ampere), burger and Time-out vouchers (courtesy of CanTeen for younger participants) until supplies ran out.

3.3.3.1. Satisfaction Survey

The Pediatric Hematology/Oncology Parent Satisfaction Survey, designed to measure parent satisfaction with medical care provided to their children aged 13 to 18 years was used for proxies in this study. Since Varni and his team had not developed a patient self-report form he gave permission for me to modify the wording to a self-report scale appropriate for AYA (Varni, personal communication 2003).

The original instrument has six (6) domains: general satisfaction, information, inclusion of family, communication, technical skill, and emotional needs of the patient, and of the parent. There is evidence for the validity and reliability of this instrument (Varni et al., 2000).

To improve the face validity of the instrument a further modification was made to both AYA and proxy versions, with the addition of 13 items. These items had been developed in previous qualitative research with a sample of the AYA cancer population in New Zealand (Ewing, 2001; Health Services Development Plan, 2000). While eleven of these items fit under the existing six domains, the remaining two items (financial needs and current state of well-being) may be additional domains that will be tested and published at a later date. These additional items are appended to the end of Varni's questionnaire to minimise the potential effect on the integrity of the original instrument.

3.3.3.2. Preference for Support Services instrument

In collaboration with Dr Madeleine King, and Ms Patsy Kenny also from the Centre for Health Economics Research and Evaluation (CHERE, UTS) and an expert in the developing preference surveys, I developed an instrument to measure preferences for support services.

The data is arranged according to an associated "Discrete Choice Experiment" constructed using established principles of experimental design. This allows estimation of parameters in a Discrete Choice Model, which reflect respondents' "stated preferences" for, or relative valuation of the attributes of the support services. This instrument is considered highly innovative, and at the time of development was a first in the field of preferences for health services.

The Preferences for Support Service instrument for this study has 3 elements:

1) An information sheet, which describes the study, defines the aspects of the "experiment" in terms of the "attributes" of a hypothetical mix of support services, and describes the choice task for the respondent. (There were 6 types of support service i.e. cultural, spiritual, financial, rehabilitative, and emotional support for the young adult patient, and emotional support for the proxy and family);

2) An example of a choice task (where participants are required to choose between a pair of hypothetical scenarios, labelled Mix A and Mix B, which offer a different combination of support services); and

3) Sixteen (16) pairs of scenarios. These 16 scenario-pairs are a fractional factorial subset of all possible scenario-pairs, identified (by A/Prof Deborah Street, Department of Mathematical Sciences at UTS) as 97% efficient for the estimation of the main effects associated with each of the 6 attributes.

Since the analysis of the preferences instrument does not form part of this thesis, it will be published at a later time.

3.3.3.3. Impact Report

The impact of illness on patient and family during and beyond treatment, and cure (or bereavement) can have a hugely detrimental and long-term impact on patients and their families in different ways (Bleyer, 2001; Cohn et al., 2003; Edwards et al., 2003; Hatchett et al., 1997; Huizinga et al., 2005; Klein-Geltink et al., 2005; Langeveld et al., 2002; Weber et al., 2005; Wolfe et al., 2000; Zebrack et al., 2004). Also, since their individual response mechanisms may also vary in relation to the continuum of the illness it was important to add this further dimension to the survey booklets.

Accordingly, in collaboration with Dr Belinda Goodenough (SCH-HRS of the CCCDB) and current literature I constructed the Impact Report. It was not feasible to conduct interviews and focus groups at that time. This instrument consisted of a series of open-

ended questions and provided an opportunity for respondents to comment on the extent, and in what way the AYA's illness and treatment had (i) disrupted (and/or enhanced) their ability to pursue their goals, and earn income (or meet expenses) (Cohn et al., 2003); and in what way it had (ii) interfered with (or strengthened) family and/or personal relationships (Giammona et al., 2002; Hatchett et al., 1997; Lavee et al., 2003).

The Impact Report, also requested information from the AYA about the number of overnight hospital visits and emergency room visits experienced in the last 12 months, and finally the number of occasions in the last month where activities were missed or a carer needed. This survey form is one-page and is positioned on the penultimate page of the booklets.

3.4. Scoring the data

3.4.1. PedsQL™ Measurement Model

The instructions for both the young adult version and the parent proxy version of the PedsQL instrument ask 'how much of a problem has…' each situation been in the past one month. The same five-point Likert response scale is used for all items of the PedsQL Generic Core Scale and the PedsQL Cancer Module for both the young adult and proxy versions of the instruments. This is shown in Table 3-3. The items were then reverse scored and linearly transformed and standardised to a scale range (0-100) so that higher PedsQL scores represented a better quality of life, while lower scores mean a poorer quality of life, as described by Varni's method of scoring (Varni et al., 2002a). This is demonstrated in the next two sections.

Table 3-3 The Likert Scale for Scoring the PedsQL Measurement Model

Response Choices	Never	Almost never	Sometimes	Often	Almost always
Raw Scores	0	1	2	3	4
0-100 Scale	100	75	50	25	0

3.4.1.1. PedsQL™ 4.0 Generic Core

The Core instrument (as introduced in Chapter 2) has 23 items in 4 domains shown in Table 3-4: Physical Functioning (Ph); Emotional Functioning (Em); Social Functioning (Soc), and Study/Work Functioning (SW).

The Generic Core generates six composite scales (see Table 3-4 and Table 3-5): one for each of the four domains, a Total Scale Score (Tot) and an additional Psychosocial Health Summary Score (*Psy*, which aggregates the three psychosocial domains).

Table 3-4 The PedsQL 4.0 Generic Core domain-specific items

Physical Functioning (Ph) $(ph_{i=1\ to\ 8})$	Emotional Functioning (Em) $(em_{i=1\ to\ 5})$	Social Functioning (SOC) $(soc_{i=1\ to\ 5})$	Study/Work Functioning (SW) $(sw_{i=1\ to\ 5})$
1. It is hard for me to walk more than one block	1. I feel afraid or scared	1. I have trouble getting along with other young people	1. It is hard to pay attention in class/at work
2. It is hard for me to run	2. I feel sad or blue	2. Other young people do not want to be my friend	2. I forget things
3. It is hard for me to do sports activity or exercise	3. I feel angry	3. Other young people tease me	3. I have trouble keeping up with my study/work duties
4. It is hard for me to lift something heavy	4. I have trouble sleeping	4. I cannot do things that other young people my age do	4. I miss study/work because of not feeling well
5. It is hard for me to take a bath or shower by myself	5. I worry about what will happen to me	5. It is hard to keep up with my peers	5. I miss study/work to go to the doctor or hospital
6. It is hard for me to do chores around the house			
7. I hurt or ache			
8. I have low energy			

The Total Scale Score (Total$_j$), equation (i), is the mean of all (23) items within the PedsQL Generic Core for participant j.

$$\text{Total}_{(j)} = \frac{\sum_{i=1}^{8} ph_{ji} + \sum_{i=1}^{5} em_{ji} + \sum_{i=1}^{5} soc_{ji} + \sum_{i=1}^{5} sw_{ji}}{23},$$

where, the items are aggregated within the domains as follows:

$$\sum_{i=1}^{8} ph_{ji} = ph_{j1} + ph_{j2} + \ldots + ph_{j8}$$

$$\sum_{i=1}^{5} em_{ji} = em_{j1} + em_{j2} + \ldots + em_{j5}$$

$$\sum_{i-1}^{5} soc_{ji} = soc_{j1} + soc_{j2} + \ldots + soc_{j5,} \text{ and}$$

$$\sum_{i=1}^{5} sw_{ji} = sw_{j1} + sw_{j2} + \ldots + sw_{j5}$$

Table 3-5 The PedsQL Generic Core Summary Scales

Total Scale Score	Physical Health Summary Score	Psychosocial Functioning
All 23 items	8 items from physical functioning	15 items from emotional, social and study/work functioning
Tot_j	ph_j	psy_j

The final step in scoring all these scales was to reverse score, linearly transform and standardise the summary scale scores i.e. Total Health Summary Score (Tot_j), the Physical Health Summary Score *(ph$_j$) and* Psychosocial Functioning (*psy$_j$*) to a scale range (0-100), to do this I carried out the following calculations, as for Varni's original scoring algorithm:

$$Tot_j = \frac{100n - 25(\sum_{i=1}^{8} ph_{ji} + \sum_{i=1}^{5} em_{ji} + \sum_{i=1}^{5} soc_{ji} + \sum_{i=1}^{5} sw_{ji})}{n}, \text{ where } n = 23 \qquad \text{(ii)}$$

$$ph_j = \frac{100n - 25\sum_{i=1}^{8} ph_{ji}}{n}, \text{ where } n = 8 \qquad \text{(iii)}$$

$$psy_j = \frac{100n - 25(\sum_{i=1}^{5} em_{ji} + \sum_{i=1}^{5} soc_{ji} + \sum_{i=1}^{5} sw_{ji})}{n}, \text{ where } n = 15 \qquad \text{(iv)}$$

In addition to an overall psychosocial functioning scale, the three component domains are kept as separate scales. The scale scores for these domains were:

$$em_j = \frac{100n - 25\sum_{i=1}^{5} em_{ji}}{n}, \text{ where } n = 5 \qquad \text{(v)}$$

$$soc_j = \frac{100n - 25\sum_{i=1}^{5} soc_{ji}}{n}, \text{ where } n = 5 \qquad \text{(vi)}$$

$$sw_j = \frac{100n - 25\sum\limits_{i=1}^{5} \text{sw}_{ji}}{n}, \quad \text{where } n = 5 \qquad (vii)$$

Missing items and scale scores:

If less than 50% of a scale was missing, the mean of the items completed within that scale was calculated as the scale score. This method is considered the least biased procedure for missing data, but it may artificially reduce variability (Fairclough et al., 1996 cited in King, 2001, p123) and (Varni et al., 2001). In instances where half or more of the item responses were missing for a scale, the scale score was recorded as missing (Fayers et al., 1999 cited in King, 2001, p123; Ware et al., 1993 cited in King, 2001, p123).

3.4.1.2. PedsQL™ 3.0 Cancer Module

The PedsQL™ Cancer Module (see Table 3-6) has 27 items, grouped into 8 domain-specific scales as follows: Pain and Hurt (*P*); Nausea (*N*); Procedural Anxiety (*PA*); Treatment Anxiety (*TA*); Worry (*W*); Cognitive Problems (*CP*); Perceived Physical Appearance (*A*); and Communication (*C*). The response choices were the same as those for the PedsQL Generic Core (in Table 3-3).

The Cancer Module contained one item that was not applicable to participants with blood disorders, such as Thalassaemia. However, for the substantive study, approval will be sought from the author to alter this question from, "*I worry that my cancer will return*" to read, '*I worry that my illness will get worse*'.

Scaling of the PedsQL Cancer Module was similar to the Generic Core, i.e. scale scores were the mean of items within each domain. Unlike the Generic Core, there was no overall total scale score for the Cancer Module. The domain scale scores were calculated in the following way for each participant:

The Pain & Hurt (*P_j*) scale score, equation (viii), is the average of the two (2) items within the *P* domain of the Cancer Module for each participant*j*:

$$P_j = \frac{\sum\limits_{i=1}^{n} P_{ji}}{n}, \quad \text{where } n = 2 \qquad (viii)$$

The Nausea Scale Score (N_j), equation (ix), is the mean of the five (5) items forming the N domain:

$$N_j = \frac{\sum_{i=1}^{n} N_{ji}}{n}, \quad \text{where } n = 5 \tag{ix}$$

Table 3-6 PedsQL Cancer Module domain-specific items

Cancer Module Domains	No of items	Items
Pain & Hurt (P)	2	1. I ache or hurt in my joints and/or muscles
		2. I hurt a lot
Nausea (N)	5	1. I become sick to my stomach when I have medical treatments
		2. Food does not taste very good to me
		3. I become sick to my stomach when I think about medical treatments
		4. I feel too sick to my stomach to eat
		5. Some foods and smells make me sick to my stomach
Procedural Anxiety(PA)	3	1. Needle Sticks (i.e. injections, blood tests, IV's) hurt
		2. I get scared when I have to have blood tests
		3. I get scared about having needle sticks (i.e. injections, blood tests, IV's)
Treatment Anxiety (TA)	3	1. I get scared when waiting to see the Doctor
		2. I get scared when I have to go to the doctor
		3. I get scared when I have to go to the hospital
Worry (W)	3	1. I worry about side effects from the medical treatments
		2. I worry about whether or not my medical treatments are working
		3. I worry that my cancer will come back or relapse
Cognitive Problems (CP)	5	1. It is hard for me to figure out what to do when something bothers me
		2. I have trouble solving math problems
		3. I have trouble writing study papers or reports
		4. It is hard for me to pay attention to things
		5. It is hard for me to remember what I read
Perceived Physical Appearance (A)	3	1. I feel I am not good looking
		2. I don't like other people to see my scars
		3. I am embarrassed when others see my body
Communication (C)	3	1. It is hard for me to tell the doctors and nurses how I feel
		2. It is hard for me to ask the doctors and nurses questions
		3. It is hard for me to explain my illness to other people

Procedural Anxiety (PA_j), equation (x), is the mean of the three (3) items forming the *PA* domain:

$$PA_j = \frac{\sum_{i=1}^{n} PA_{ji}}{n}, \quad \text{where } n = 3 \qquad \text{(x)}$$

Treatment Anxiety (TA_j), equation (xi), is the mean of the three (3) items forming the *TA* domain:

$$TA_j = \frac{\sum_{i=1}^{n} TA_{ji}}{n}, \quad \text{where } n = 3 \qquad \text{(xi)}$$

Worry (W_j), equation (xii), is the mean of the three (3) items forming the *W* domain:

$$W_j = \frac{\sum_{i=1}^{n} W_{ji}}{n}, \quad \text{where } n = 3 \qquad \text{(xii)}$$

Cognitive Problems (CP_j), equation (xiii), is the mean of the five (5) items forming the *CP* domain:

$$CP_j = \frac{\sum_{i=1}^{n} CP_{ji}}{n}, \text{ where } n = 5 \qquad \text{(xiii)}$$

Perceived Physical Appearance (A_j), equation (xiv), is the mean of the three (3) items forming the A_j domain:

$$A_j = \frac{\sum_{i=1}^{n} A_{ji}}{n}, \text{ where } n = 3 \qquad \text{(xiv)}$$

Communication (C_j), equation (viii), is the mean of the three (3) items forming the C_j domain:

$$C_j = \frac{\sum_{i=1}^{n} C_{ji}}{n}, \text{ where } n = 3 \qquad \text{(xv)}$$

3.4.2. Memorial Symptom Assessment Scale (MSAS) scoring

As mentioned in Section 3.1.2 the 30-item MSAS was used to classify participants into "known groups" of slight, moderate and severe symptom experience. Symptom prevalence is not necessarily related to its reported frequency or severity or distress. But the "measurement of these characteristics yields clinical insights that cannot be captured by symptom checklist alone" (Collins et al., 2000). For the MSAS, higher scores reflected greater symptom severity.

The first twenty-two (22) symptom items consist of three "dimensions" (3-D), i.e. frequency, severity, and distress. Participants were asked to rate these symptoms in the following way: How often they experienced the symptom; its severity; and how much it distressed them. The last eight (8) items were longer lasting symptoms, such as hair loss and mouth sores. Frequency ratings were not appropriate to these symptoms. Hence, these symptoms were two dimensional (2-D), with assessments of severity and distress only.

The calculation of symptom scores:

Each domain or symptom score was calculated as the mean of the component items according to the standard methods prescribed by the instrument authors (Collins et al., 2000). For 3-D symptoms the domain score was calculated as the average of the item values reported for 'severity', 'frequency' and 'distress'. The domain score for 2-D symptoms was the mean of the item values for 'severity' and 'distress' only.

Table 3-7 provides the scores that could be allocated to the symptom frequency (*Freq*), severity (*Sev*) and distress (*Dis*) as assessed in the self-report.

For example, if a patient reported that they were experiencing a 'moderate' lack of energy 'almost always' which bothered them 'very much', the scoring for this lack of energy, En_j for participant j, for this 3-D domain scenario was as follows:

$$En_j = \frac{Freq + Sev + Dis}{3} \qquad \text{(xvi)}$$

$$= \frac{4+2+4}{3}, \qquad \text{where } Freq = \text{'almost always'} \rightarrow 4$$

$$Sev = \text{'moderate'} \rightarrow 2$$

$$Dis = \text{'very much'} \rightarrow 4$$

$$= 3.33$$

This patient, for example, also reported that they 'sometimes' had pain in the last week, which was 'moderate' and bothered them 'quite a bit'. The scoring for this Pain (P_j) 3-D symptom scenario was as follows:

$$P_j = \frac{\sum (freq('sometimes' = 2), Sev('mod' = 2), Dis('quite-a-bit' = 3))}{3} \quad \text{(xvii)}$$

$$= \frac{2+2+3}{3}$$

$$= 2.33$$

Table 3-7 The MSAS 3-Dimensional Symptom Scoring

In the past week did you have any:....? Yes/No						
How often did you have it?	Frequency	-	Almost never	Sometimes	A lot	Almost always
How severe was it usually?	Severity	-	Slight	Moderate	Severe	Very severe
How much did it bother or distress you?	Distress	Not at all	A little bit	Somewhat	Quite a bit	Very much
Scoring:		**0**	**1**	**2**	**3**	**4**

The scoring of a patient reporting a 2-D symptom such as 'very severe' hair loss (hl_j), which bothered them 'quite a bit', was as follows:

$$hl_j = \frac{\sum Sev('very-severe'), Dis('quite-a-bit')}{2} \quad \text{(xviii)}$$

$$= \frac{4+3}{2}$$

$$= 3.5$$

The calculation of summary scores:

While the Total-MSAS is the mean of all 30 symptom scores, the subscale scores for: Physical (PHYS- MSAS), Psychological (PSYCH- MSAS), and the Global Distress Index (GDI-MSAS) are a little more complex as described below.

Table 3-8 provides a list of the symptoms making up the Total-MSAS and each of the subscale summary scores.

For example, the *Total-MSAS* score is calculated by finding the mean of all 30-domain (symptom) scores, that is:

Total-MSAS = Σ(energy, pain, drowsy, nausea, appetite, constipation, dry mouth, vomiting, taste, weight loss, dizzy, sad, worry, irritability, nervous, insomnia, concentration, cough, itching, hair loss, numbness, sweating, diarrhoea, skin change, dyspnea, looks, mouth sores, swallowing, swelling, urination) ÷ 30

For example, consider the resulting *Total-MSAS* symptom score for a patient reporting a lack of energy, pain and hair loss scores above (xvi, xvii, xviii in calculating symptom scores), a nausea domain score = 2, irritability domain score = 1, a worry domain score = 1, and all other symptoms were = zero. Hence:

$$Total\text{-}MSAS \text{ score} = \frac{\sum(\text{energy, pain, hair loss, nausea, irritability, worry})}{30}$$

$$= \Sigma(3.3, 2.3, 3.5, 2, 1, 1) \div 30$$
$$= 13.1 \div 30$$
$$= 0.44$$

The *PHYS-MSAS* subscale score is the mean of 11, 3-D physical symptom scores i.e. lack of energy, pain, feeling drowsy, nausea, lack of appetite, constipation, dry mouth, vomiting, change in food taste, weight loss, and dizziness.

For example, consider the resulting *PHYS-MSAS* symptom score for a patient reporting in the last week a 'moderate' (value = 2) lack of energy affecting them 'a lot' (value = 3), which bothered them 'very much' (value = 4). The resulting Lack of Energy (*En*) domain score resulted in a mean value of 3. This patient also reported 'severe' (value = 3) weight loss, which bothered or distressed them 'quite a bit' (also value = 3). The average of the weight loss domain gives a score of 3. This patient does not experience any of the remaining nine symptoms in this scale. The resulting *PHYS-MSAS* for this patient is calculated as follows:

PHYS-MSAS = Σ(pain-0, energy-3, dry mouth-0, nausea-0, drowsy-0, vomiting-0, appetite-0, dizziness-0, taste-0, weight loss-3, constipation-0) ÷ 11

$$= \frac{\sum(0,\ 3,\ 0,\ 0,\ 0,\ 0,\ 0,\ 0,\ 0,\ 3,\ 0)}{11}$$

$$= \frac{6}{11}$$

$$= 0.55$$

Table 3-8 The MSAS Subscale Scores

Total-MSAS 3-D		GDI-MSAS[2] (mean 1-dimension)		PSYCH-MSAS 3-D[3]	PHYS-MSAS 3-D[4]
Mean of all 30 symptom scores		**Mean of 4 frequency values + 6 distress values:**		**Mean of 6 symptom scores:**	**Mean of 11 symptom scores:**
Energy	Insomnia	**Frequency values for:**	**Distress values for:**	Concentration	Energy
Pain	Concentration			Nervous	Pain
Drowsy	Cough	Sadness	Appetite	Insomnia	Drowsy
Nausea	Itching	Worry	Energy	Sadness	Nausea
Appetite	Hair loss	Irritability	Pain	Worry	Appetite
Constipation	Numbness	Nervous	Drowsy	Irritability	Constipation
Dry mouth	Sweating		Constipation		Dry mouth
Vomiting	Diarrhoea		Dry mouth		Vomiting
Taste	Skin change				Taste
Weight loss	Dyspnea				Weight loss
Dizziness	Looks				Dizziness
Sadness	Mouth sores				
Worry	Swallowing				
Irritability	Swelling				
Nervous	Urination				

Notes:
1. Each symptom or domain score is the mean of the symptom component items
2. The GDI utilises one dimension of each symptom, i.e. frequency item of 4 symptoms plus the distress items of 6 symptoms
3. PSYCH score = the mean of 6 symptom 3-Dimensional scores
4. PHYS score = the mean of 11symptom 3-Dimensional scores (frequency, severity, distress)

The *PSYCH-MSAS* subscale score is the mean of the six 3-D psychological symptoms, i.e. difficulty concentrating, feeling nervous, difficulty sleeping, feeling sad, worrying, and feeling irritable. For example, consider the resulting PSYCH-*MSAS* symptom score for a patient reporting they 'sometimes' (value = 2) had difficulty sleeping in the last week, the severity was 'slight' (value = 1) but it did not bother ('distress' value = 0) the patient. The patient was not affected by the other symptoms included in this scale. The insomnia domain score resulted in a mean value of unity (1).

PSYCH-MSAS = Σ(concentration-0, nervous-0, insomnia-1, sadness-0,

irritability-0, worry-0) ÷ 6

$$= \frac{\sum(0,\ 0,\ 1,\ 0,\ 0,\ 0)}{6}$$

$$= 0.165$$

The *Global Distress Index* (*GDI-MSAS*) subscale score is different to the other scales as it focuses on merely one dimension (1-D) from each of a selected set of symptoms. It is the mean of 10 item values made up of 4 symptom frequency values and 6 symptom distress values. The frequency values are for sadness, worry, irritability and nervousness. The distress values are for lack of appetite, lack of energy, pain, drowsy, constipation and dry mouth. For example the GDI-MSAS scale score for participant coded as 135 was:

$GDI\text{-}MSAS_{135} = Freq$(sadness-1, worry-1, irritability-0, nervousness-0) +

        Distress (lack of appetite-0, lack of energy-3, pain-0, drowsy-0,

        constipation-0, dry mouth-0)] ÷ 10

$$= \frac{\sum[(1,1,0,0)+(0,3,0,0,0,0)]}{10}$$

$$= 0.5$$

3.4.3. Imputation of missing values

Imputation of missing PedsQL data was addressed in section 3.4.1.1 above, while missing data for the MSAS, socio-demographic and illness information are detailed below:

For the MSAS, missing data were dealt with in a similar way to that of the PedsQL for 3-D symptom domain scores where only one of the three-symptom/item dimensions of each domain was missing. But for the few 2-D domain scores where one of the two items of the domain was missing, and for the GDI-MSAS where only one dimension of certain symptoms was averaged, a different way of scoring was required.

If one dimension was missing for a reported symptom, the subscale for Tot-MSAS, PSYCH, PHYS, or GDI were calculated as the sum of the symptom dimension values divided by the number of dimensions reported. The symptom could not merely be listed as missing, because in cases where it was clear that the patient was experiencing numerous symptoms but consistently reported only one symptom dimension, this would result in NIL symptoms, subsequently placing the patient into the lowest symptom group.

For example, if the 3-D symptom, dry mouth was missing frequency and severity, and yet the distress was assessed as 'somewhat' distressing (code 2), the Physical (*PHY*) subscale was calculated as:

PHYS = [Σ(pain-(0,0,0), energy-(0,0,0), dry mouth-(0,0,2), nausea-(0,0,0), drowsy-(0,0,0), vomiting-(0,0,0), appetite-(0,0,0), dizziness-(0,0,0), taste-(0,0), weight loss-(0,0), constipation-(0,0)] ÷ 30 items

$$= \frac{2}{30}$$

$$= 0.067$$

Missing socio-demographic & illness information:

Ethnic group:

If the ethnicity of a member of the dyad pair was missing, the relationship between the pair was considered. If the dyad pair included the biological mother and son, for instance, and ethnicity was the only missing information, then to minimize calls to participants, the ethnic group was assumed to be the same. However, if several items of information needed to be checked then ethnicity was confirmed or corrected at the same time.

Days absent:

For each dyad, the patient and proxy responses were compared visually immediately after data collection for consistency in relation to the number of days that the patient was absent from work or study and the number of days that the proxy missed work (in the past 30 days) due to the patient's physical or mental health. If proxies had not reported interruption to their daily routine then I coded the missing "days absent" as a red zero, '0'. Also, when the patient's or proxy's data for this item was their only missing data, the response from the other dyad pair was sufficient to save troubling participants for this piece of information alone.

3.5. Analysis

The purpose of this thesis was to check the validity, sensitivity and reliability of the modified HRQOL instruments for use in the Australian context for 16 to 25 year old patients with a cancer or blood disorder diagnosis. I have therefore attempted to measure the unobservable latent variable HRQOL and can "only infer that the instrument is valid in so far as it correlates with other observable behaviour" (Fayers et al., 2000, p45).

Hence, I collected data to test whether the instruments represent the intended constructs and produce useful measurements that reflect patient HRQOL.

3.5.1. Reliability

Internal consistency reliability gives a measure of the extent to which the items are interrelated. The research question was: Does the modified PedsQL show internal consistency for the targeted 16-25 year olds, and their proxies?

Cronbach's Alpha coefficient was utilised to provide this measure of the Internal Reliability Consistency of participant responses for each of the PedsQL domain scales (Core and Module).

$$\alpha_{Cronbach} = \frac{m}{m-1}(1 - \frac{\sum Var(x_i)}{Var(S)}),$$

where m = the number of items, $Var(x_i)$ is the variance of the ith item in the scale, calculated from the sample of patients who completed the QOL assessment, and $S = \Sigma x_i$. The minimum standard for group comparisons is an alpha value of 0.7, though values above 0.8 are considered good and a value of 0.9 excellent. For analysing individual participant scores the recommended reliability criterion is 0.9. (Cronbach, 1951; Nunnally et al., 1994 cited in Varni 2002). The expectation was that the alpha values generated by my sample would be comparable to those of Varni's sample.

3.5.2. Feasibility

Varni determined feasibility and practicality through the percentage of missing values for each item (McHorney et al., 1994; Varni et al., 2002a; Varni, Seid, Knight, Uzark et al., 2002), and item response distributions (Varni et al., 2001). These were also checked across the full-scale range and the floor and ceiling effects.

3.5.2.1. Missing PedsQL Measurement Model data

Although Varni used the percentage of missing data as a criterion of study feasibility, the amount of missing data in my sample was minimised as mentioned earlier due to the follow-up phone call option as approved by the ethics committee. This option, which many participants permitted me to use, was administered with great sensitivity to participants due to the potentially serious nature of their illness. Hence the amount of

missing data in my sample was not comparable with that of Varni. Also, resulting from these follow-up phone calls some participants who had mislaid the Survey pack were able to request a replacement, which was duly dispatched, while other participants found it more convenient to complete their questionnaires over the phone.

The window of opportunity to complete missing responses or clarify ambiguous responses depended on the type of data, e.g. family information was unlikely to change over the data collection period, so these data could be completed at any time. However, HRQOL responses and the MSAS symptom levels needed to be followed up within a week, to ensure accuracy and consistency with the information already provided and the specific completion timeframe of their questionnaire.

For the demographic and clinical information any contradictory information between the young adult and their proxy was also checked for confirmation leaving very few missing data. Having participant permission for follow-up a phone call (mentioned above) enabled this timely feedback for their queries and the subsequent reduction in the potential loss of data.

Non-applicable items:

Non-applicable items were coded as 'zero' as they were 'Never' a problem for patients. For example, the item, "I worry that my cancer will come back or relapse" and the domain about Nausea (Cancer Module) were not applicable to blood disorder patients as these items reflected a side effect of cancer treatment.

3.5.2.2. Floor and Ceiling Effects of the PedsQL Measurement Model

The floor and ceiling effects give an indication of the HRQOL data being grouped at the maximum or minimum extreme value. The floor effect refers to the percentage of minimum scores, and the ceiling effect refers to the percentage of maximum scores.

3.5.3. Validity

The validation process checks that the instruments measure what they intend to measure and whether they are useful for their intended purpose (Fayers et al., 2000, p45). As previously mentioned, I replicated Varni's procedures and compared my results with his

to ascertain the validity of the PedsQL Generic Core and the PedsQL Cancer Module. I also used additional analyses.

There are various types of validity, and I used various analyses and parts of the data to assess these:

- Construct validity was assessed by item to item correlations derived using factor analysis on both patient and proxy versions of the PedsQL Generic Core and the PedsQL Cancer Module;

- Convergent and Divergent validity were assessed by
    - Correlations among the domains within both instruments for patient and proxy responses separately; and by
    - Correlations between PedsQL Generic Core and PedsQL Cancer Module for patient and proxy responses separately;

- Proxy validity was assessed by patient-proxy correlations and mean differences for Core and Module domain scales.

- Clinical validity was assessed with Known-groups methodology to investigate the patterns of HRQOL in groups known to differ by other clinically relevant criteria.

3.5.3.1. Construct Validity

An Exploratory Factor Analysis (EFA) provides item-to-item correlations that then automatically group items into a few main factors to test the underlying dimensions. Varni used oblique rotation, however this was no longer available in the SPSS software as Varimax rotation had superseded it as a better choice for QOL data (Fayers et al., 2000). Therefore, I used the Principal Components Factor Analysis with Varimax rotation and the standard Eigenvalue cut-off set at 1. Hence, the pattern of how well items grouped together was important rather than comparing my factor weights with Varni's directly. My results were compared with Varni's results to answer the following questions:

a. Is the factor structure of the modified PedsQL Generic Core, observed in young Australians (16-25 years old), the same as that of the original instrument observed in young Americans (13-18 years old)?

b. Is the factor structure of the modified PedsQL Generic Core, observed in the proxies of young Australians (16-25 years old) the same as that of the original instrument observed by the proxies of young Americans (13-18 years old)?

c. Are the factors generated from the PedsQL Cancer Module (self- and proxy-reports) consistent with Varni's a priori hypothesized factor structure?

Varni and colleagues did not conduct a factor analysis on the PedsQL disease-specific modules. His methodology for the PedsQL Cancer Module was instead driven by focus groups, cognitive interviews, conceptual framework and subsequent internal consistency reliability coefficients of the sample (Varni et al., 2004; Varni et al., 2002a) [J.W. Varni personal communication, May 2004]. However, I also conducted an EFA to confirm whether the items of the PedsQL Cancer Module would fall roughly into similar a priori domains.

Factor Analyses were carried out on the items of the:
    a. PedsQL Generic Core for both the self- and proxy-report; and
    b. PedsQL Cancer Module for both the self- and proxy-report.
I examined the correlations between the multi-item domain scales of both the PedsQL Generic Core and the PedsQL Cancer Module to determine whether the scores correlated in the manner expected amongst each other within and between each instrument. The scale scores of both PedsQL instruments for self-report and proxy-report were included in a large correlation matrix.

Assumption of Normality

"The standard factor analysis model makes no special assumption about data being continuous and Normally distributed" (Fayers et al., 2000, p107). However, underlying estimation procedures (ML and 'least squares") for factor analysis assume continuous data from a Normal Distribution. But, many forms of QOL data violate this assumption by being highly asymmetrical and categorical (Fayers et al., 2000, p107). For example, the five categories of the Likert Scale in the PedsQL instruments were: 'Never', 'Almost Never', 'Sometimes', 'Almost Always' and 'Always' which are not likely to yield equal-interval scales for patient responses, and depending on the disease and treatment many of the items are likely to take extreme values i.e. ceiling effects in the scaled data. Since

QOL items frequently have highly skewed non-Normal distributions (Fayers et al., 2000, p108) I endeavoured to include as wide a range of disease severity (slight to severe) as possible. Also I increased the sample size by including two additional hospitals into the study i.e. POWH and RHW, and extending the recruitment period by about 6 months.

Although mathematical theory for factor analysis of categorical data has been developed, its effectiveness for estimating the structure, and large sample size required remains unclear for stable and consistent estimation of factors (Bartholomew cited in Fayers et al., 2000, p107). Attempts to develop factor analysis, which makes no assumptions about the distribution of data, suggest that very large samples may be necessary (Bartholomew as cited in Fayers et al., 2000, p110). It is estimated that the sample size should be around five times the number of items in the questionnaire. In the case of the Generic Core the sample size would need to be (23 items*5=) 115 participants, while the Cancer Module would need to have (27 items*5 =) 135 participants.

While the effect of these violations of assumptions is mainly unknown, empirical results as well as simulation studies suggest that the techniques may be relatively robust, and the sample size should be increased to compensate (Fayers et al., 2000Bartholomew).

3.5.3.2. Convergent and Divergent Validity

Highly correlated items or domains demonstrate convergent validity, while those that are relatively uncorrelated are considered to indicate divergent validity. For example a performance-based measure of walking should be positively correlated with walking a block (Fayers et al., 2000, p45; Hays et al., 2005) hence convergent validity, but divergent validity would be demonstrated in a weak correlation with say perceived physical appearance.

The following intercorrelations were estimated to assess convergent validity:

(a) PedsQL Generic Core

Between domains within the Generic Core for:

- Young adult patients, and
- Proxies

(b) PedsQL Cancer Module

    Between domains within Cancer Module for:

- Young adult patients, and
- Proxies

(c) Between the PedsQL Generic Core and PedsQL Cancer Module for:

- Young adult patients, and
- Proxies

Correlation coefficients are a measure of the degree of association between continuous variables. I used Pearson's Correlation (*r*) and with *n* subjects and two variables x_i and y_i (where *i* ranges from 1 to *n*),

$$r = \frac{\sum (x_i - \overline{x})(y_i - \overline{y})}{\sqrt{\sum (x_i - \overline{x})^2 \sum (y_i - \overline{y})^2}},$$

where $\overline{x}$ and $\overline{y}$ are the mean values of the two variables *x* and *y*.

Pearson's Correlation measures the scatter of the observations around a trend line. The greater the scatter, the lower the correlation. Values of r lie between +1 and –1, and for r = 0 no association is indicated between x and y.

It was expected that: the inter-correlations between the Generic Core and Cancer Module would demonstrate medium to large effects; items in the external domain i.e. Physical Functioning, would correlate more highly with each other than with internal domains, i.e. Psychosocial Functioning, because issues about physical concerns are external concepts which are very different and easier to assess than those of an internal nature, such as emotional, social or work/study functioning. It was also expected to see high correlations between the Physical HSS (Generic Core) and Pain & hurt (Cancer Module); and high correlations between the Psycho-social HSS (Generic Core) with Procedural Anxiety, Treatment Anxiety, Worry, and Cognitive Problems (Cancer Module). No association was expected between Social Functioning and Procedural or Treatment Anxiety.

3.5.3.3. Patient-Proxy Concordance

Are other people able to validly assess patient HRQOL? The level of agreement between the patient and their nominated proxy can be provided through cross-informant variance. This was examined in two ways:

(i) Patient-proxy correlations, and (ii) Mean differences for each of the scales of the Generic Core and the Cancer Module, providing particular insights and permitting answers to the following questions:

1. How good are proxies at observing and reporting the health of young adult patients?
2. How closely correlated are the young adult patient and proxy reports of HRQOL?

In addition to investigating the correlations above, the differences between the self- and proxy-reports were correlated and compared with those of Varni's sample. The difference d, between the self and proxy-report is:

$$d = \bar{x}_I - \bar{x}_{II},$$

where, $\bar{x}_I, \bar{x}_{II}$ are the mean domain scores for self-report and for proxy-report respectively. The standard error of this difference is:

$$SE(d) = \sqrt{\frac{SD_I^2}{n_I} + \frac{SD_{II}^2}{n_{II}}}, \text{ where } n_I = \text{self-report sample size}$$
$$n_{II} = \text{proxy sample size}$$

I used Cohen's rule for correlation effect sizes: small (0.10 - 0.29), medium (0.30 – 0.49), and large ($\geq$0.50) as I was replicating Varni's methods. Medium to large effect sizes were expected for patient versus proxy concordance for all corresponding subscales but not to the extent that patient reports would be unnecessary.

It is possible to have perfect correlation ($p<0.001$) between two sets of paired scores (young adult patient and proxy) but the mean values could be quite different by a large amount. Hence, I also investigated the differences between the mean (SD) of each domain of the patient-proxy dyad to test for any bias, which is reflected in a mean difference greater than zero.

3.5.4. Sensitivity - Clinical Validity ("Known-groups")

'Known groups' comparisons were used to test whether the HRQOL instruments were sensitive to differences between groups of patients and proxies that were known to be different by clinically relevant criteria. These analyses were used to assess the clinical validity of the HRQOL measures (Osoba et al., 2005).

Comparisons between degrees of health in my sample were based on a valid measure of symptom severity, i.e. the Memorial Symptom Assessment Scale (MSAS) as reported by the patient, while Varni's good-health versus poor-health was based on whether the patient was On-treatment or Off-treatment for $\leq$ 12months, or Off-treatment for >12months. Comparisons between known groups were evaluated using the difference of two means (t-Test), Analysis of Variance (ANOVA), the Kruskal-Wallace non-parametric test and the Mann-Whitney U test.

The null hypothesis was that there was no significant difference between the HRQOL means of the two groups, e.g. young adult patients needing a carer in the past 30 days compared with those who did not require a carer.

$$H_o: \; \mu_x - \mu_y = 0$$

$$H_1: \; \mu_x - \mu_y \neq 0$$

The alternative hypothesis (H_1) was that a significant difference existed between the means of the two groups, at the 5% level of significance. The standard error of the difference between the means is calculated using the standard error of the difference:

$$SE(\overline{X} - \overline{Y}) = \sqrt{SE(\overline{X})^2 + SE(\overline{Y})^2} \, ,$$

where $SE(\overline{X}) = \dfrac{S_X}{\sqrt{n_X}}$, is the standard error of the mean of sample X, and

n_x is the number of observations in X, and

$SE(\overline{Y}) = \dfrac{S_Y}{\sqrt{n_Y}}$, is the standard error of the mean of sample Y, and

n_Y is the number of observations in Y.

The 95% Confidence Interval for the difference between these two samples is:

$$95\% \text{ CI} = \bar{X} - \bar{Y} \pm 1.96(\text{SE})$$

3.5.4.1. Comparing Symptom Severity Groups

As mentioned previously (in Section 3.4.2), the MSAS instrument was used to divide patients into three 'known-groups' of symptom experience, i.e. those with slight, moderate and severe symptoms, to test the hypothesis that HRQOL is better (higher) for those with slight symptoms, and lower for those with severe symptoms.

The HRQOL scales were expected to follow the health gradient described by the symptom severity groups 1 to 3 (slight, moderate and severe) where increasing symptoms translate to decreasing QOL (King, 2001). The strength of correlations in my sample was expected to be comparable to the corresponding correlations in Varni's sample. It was hypothesized that patients off-treatment would report better (higher) HRQOL scores than those on-treatment. Those in good health, as ascertained through the Memorial Symptom Assessment Scale (MSAS) were hypothesized to have higher QOL scores. Also, patients with a cancer diagnosis and on-treatment tend to have more problems with nausea than those with a blood disorder. Therefore the correlation between blood disorder patients and nausea was expected to be low.

These symptom severity groupings were identified initially through scatter-plots of the scale TOT-MSAS against the subscales PHYS-MSAS, PSYCH-MSAS and GDI-MSAS. However, PHYS-MSAS against the TOT-MSAS scores presented the clearest results for splitting the patients into three separate groups of similar size. The cut-points (i.e. 0.2 and 1) for the three symptom groups (0-0.2; 0.2-1; 1+) were extracted and utilised (see Appendix B for the plot of these scores).

The Normality and Homogeneity of variance testing on the PedsQL$^{\text{TM}}$ Generic Core and the PedsQL$^{\text{TM}}$ Cancer Module indicated that although the data had a small ceiling effect (common with QOL data) it was reasonably normally distributed and that the three symptom severity groups (means and standard deviations) could be tested for significant differences between the symptom severity groups using a one-way ANOVA with Tukey's

Honestly Significant Difference (HSD) measure. Table 3-9 shows the various components of the ANOVA for the three (3) symptom severity groups.

The PedsQL Cancer Module data, on the other hand, was more strongly skewed and hence violated the assumption of homogeneity of variance. Therefore a non-parametric test was required to test the sensitivity of this instrument (see section 4.3.1). The Kruskal-Wallis (KW) test, being the rank-based analogue to the F test, was deemed the most suitable for indicating a significant difference between symptom severity groups (see the formula below, and section 4.4), and hence the sensitivity of this instrument. The Mann-Whitney U Test provided the detail of the significance between pairs of groups within each domain.

For the PedsQL Cancer Module the Kruskal-Wallis (*KW*) non-parametric test was used where individual values of QOL were replaced by their ranked values.

$$KW_j = \frac{12 \sum n_i (\overline{R}_i - \frac{(N+1)}{2})^2}{N(N+1)},$$

where, N = Total number of observations, and

$\overline{R}_i$ = mean of the ranks in the *i*th group.

The Ranks were then summed separately for the *g* different groups.

3.6. Software

Statistical Package for Social Scientists (SPSS), version 11.0 for Windows PC, Excel 2000, and Microsoft Office 2000 Professional were used for all analyses and word processing.

Table 3-9 Analysis of Variance (ANOVA) for comparing the means of the three Symptom Severity Groups

Source of variation	Sums of squares	Degrees of freedom (df)	Mean Squares	F
Between treatments	$S_{Between} = \dfrac{T_I^2}{n_I} + \dfrac{T_{II}^2}{n_{II}} + \dfrac{T_{III}^2}{n_{III}} - \dfrac{T^2}{n}$	g-1	$MS_{between} = \dfrac{S_{Between}}{g-1}$	$F = \dfrac{MS_{Between}}{MS_{Within}}$
Within treatments	$S_{Within} = S_{Total} - S_{Between}$	$(n_I - 1) + (n_{II} - 1) + (n_{III} - 1)$	$MS_{Within} = \dfrac{S_{Within}}{(n_I - 1) + (n_{II} - 1) + (n_{III} - 1)}$	
Totals	$S_{Total} = \sum x_i^2 - \dfrac{T^2}{N}$	N - 1		

Notes:

$T = T_I + T_{II} + T_{III}$ sum of all N observations

T_I = sum of all n_I observations of treatment I

T_{II} = sum of all n_{II} observations of treatment II

T_{III} = sum of all n_{III} observations of treatment III

$N = n_I + n_{II} + n_{III}$

g = number of different groups

x_i = *the individual observation*

C h a p t e r   4

4. Results

4.1. Participants

Of 210 potential participants approached, 190 (90%) consented (99 AYA, 91 proxies), and 167 (80%) returned completed questionnaires, of which 88 were AYA patients and 79 were proxies. The good response rate is most likely due to the support of the treating physicians. Of the AYA who participated 65 (74%) were diagnosed with cancer and 23 (26%) had a blood disorder. Most AYA participants were female (58%), and most proxies were the mother (64, 73%) of the AYA, with 8% fathers and 6% spouses. The mean age of all AYA patients (16 to 25) was 19.7(SD = 3.4) years old. Participants were recruited through the CCCBD at SCH (46), POWH (29), and RHW (13). The gender imbalance (51, 58% females and 37, 42% males) was due to the inclusion of the RHW in the closing stages of the study. The gender proportions of Varni's sample (2002) were the exact opposite with 58% (196) male and 42% (143) female.

4.1.1. Mode of Administration of Surveys to Participants

Participants were recruited at in-patient clinics, outpatient clinics or by telephone as shown in Table 4.1. Most participants (78%) were recruited by telephone. Some proxies were recruited at the same time as the patient while others were phoned later. Thus the majority of participants received a postal survey pack and completed their questionnaires at home.

4.1.2. Socio-demographic information

The socio-demographic information about participants is shown in Table 4-2. Of the 88 AYA patients more than half (48, 54%) were aged 16 to 19 years old (19 male, 29 female) and 40 (46%) were in the older age group (20-25 years old). The patients considered themselves as mainly of European descent (68, 77%), and were single (71, 81%) though 10% were married and 8% were in de facto relationships; 54% had not completed 12th Grade of school, while 29% had completed a Tertiary Certificate course (15%) or a Degree (14%). One third (29, 33%) reported they were studying only, and about a third (26, 30%) reported studying and working, while a quarter (22, 25%) of the AYA patients were working only. As expected there were more tertiary qualified young

adults in the older age group with most of these people both working and studying, however, 8% were not engaged in work or study.

AYA-nominated proxies were mainly in the 40-49 age-bracket (46, 52%), most were married (63, 72%), considered themselves of European descent (64, 81%), and the AYA's mother (64, 73%). The educational level of proxies was reasonably evenly spread from year 9 through to Graduate/Professional Degree status, with 44% (38) reporting that they had some tertiary qualification.

Table 4-1 Mode of Survey Administration to participants

	Frequency	Percent (approx %)
SCH		
In-patient	4	5
Out-patient	14	16
Telephone	28	32
POWH		
In-patient	0	0
Out-patient	1	1
Telephone	28	32
RHW		
In-patient	0	0
Out-patient	1	1
Telephone	12	14
Total	**88**	**100**

Note: Variation in the percentage value is due to rounding.

4.1.3. Clinical information

Clinical information is contained in Table 4-3 and shows that about three-quarters of the patients (65, 74%) had been treated for cancer. Two-thirds of them were off-treatment (60, 68%) at the time of participation and reported their illness to be in remission or cured. Of the 23 young adults with a blood disorder, 13 had Thalassaemia and 10 had other blood disorders such as Sickle Cell Anaemia, Von Willebrand's Disease, Anaemia, and Haemophilia. Cancer diagnoses included Leukaemia, Hodgkin's Lymphoma, Non-Hodgkin's Lymphoma, Ewing's Sarcoma, Wilm's Tumour, Neuroblastoma, Carcinoma, and Osteosarcoma. The patients had no co-morbid disease or major developmental disorders as a result of the exclusion criteria, and none were within 3 months of diagnosis or requiring end-of-life care. On-treatment status (28, 32%) was defined as patients who were receiving treatment to induce remission (of cancer) or as on-going maintenance (Thalassaemia). Off-treatment status (60, 68%) was defined as patients who were in

remission or cured, had not received treatment last month as they had completed their treatment, and were not expecting treatment over the following month. Cancer survivors who had achieved the 5-year post-treatment milestone were pre-allocated to a 'Late Effects' study running concurrently, hence the relatively small number of patients in remission or cured in this study.

Table 4-2 Socio-Demographic Information about Participants

Table 4.2a Demographics and Medical characteristics of AYA Patients

Adolescents/Young adults		No.	%
Age brackets:	16 - 19 years	48	54%
	20 - 25 years	40	46%
Ethnic grouping:	European descent	68	77%
	Other	20	23%
Gender:	Males	37	42%
	Females	51	58%
	Total	88	100%
Marital Status:	Single	71	81%
	Married	9	10%
	Separated	0	0%
	Divorced	1	1%
	Defacto	7	8%
Educ. Status:	Missing	2	2%
	7th - 9th Grade	3	3%
	9th - 12th Grade	43	49%
	High School Certificate	15	17%
	Tertiary Cert. course	13	15%
	Graduate or Prof Degree	12	14%
Relationship to Nominated Proxy			
	Daughter	45	51%
	Son	31	35%
	Spouse/Partner	6	7%
	Grandson	1	1%
	Brother	1	1%
	Nephew	1	1%
	Missing	3	3
Employment:	Not Studying or Working	7	8%
	Studying	29	33%
	Working	22	25%
	Study & Work	26	30%
	Missing	4	4%

Table 4.2b Demographics Proxy

Proxy		No.	%
Age brackets:	20 – 29 years	4	5%
	30 - 39	9	10%
	40 – 49	46	52%
	50 – 59	18	21%
	60 – 69	1	1%
	70+	1	1%
Ethnic group:	European descent	64	81%
	Other	15	19%
Gender	Males	12	14%
	Females	67	76%
	Missing	9	10%
Marital Status:	Single	2	2%
	Married	63	72%
	Separated	2	2%
	Divorced	6	7%
	Defacto	5	6%
	Widowed	1	1%
Educ. Status:	6th Grade or less	2	2%
	7th - 9th Grade	7	8%
	9th - 12th Grade	18	21%
	High School Certificate	14	16%
	Tertiary Cert. course	20	23%
	Graduate or Prof Degree	18	21%
Relationship to Patient			
	Mother	64	73%
	Father	7	8%
	Spouse	5	6%
	Grand-mother	1	1%
	Brother	1	1%
	Aunt	1	1%
	Missing	9	10%

Table 4-3 Clinical Information about the AYA patient

		Number	%
Diagnosis:	Cancer	65	74%
	Blood Disorder (non-Thalassaemia)	10	11%
	Thalassaemia	13	15%
Treatment:	Off - Treatment	60	68%
	On - Treatment	28	32%
Treatment Groups/Condition/Disease:			
	On-going treatment	27	31%
	Remission	42	48%
	Relapse	1	1%
	Cured/long term follow-up	18	20%
	Total	**88**	**100%**

4.2. Reliability

Table 4-4 provides the Internal Reliability coefficients (Cronbach's alpha) for the Generic Core and Cancer Module for patient and proxy with Varni's corresponding results for comparison.

The modified PedsQL Generic Core and PedsQL Cancer Module showed internal consistency for the patient and proxy groups. The Cronbach Internal Reliability alpha values generated by my sample demonstrated strong reliability and very good consistency that compared well with those generated by Varni's sample. The Generic Core alpha coefficients in my sample ranged from 0.81 to 0.95, and 0.85 to 0.96 for self- and proxy-reports respectively, compared to Varni's greater range of 0.75 to 0.92, and 0.79 to 0.94.

Both samples exceeded the standard minimum reliability of 0.70 for group comparisons for both patient and proxy. My sample approached or exceeded the alpha of 0.9 recommended for individual patient analysis in all domains of the Generic Core except for Social Functioning self-report (0.81) which was comparable to Varni's values (0.81 and 0.80 respectively) for self and proxy. Varni's alpha values for Study/Work Functioning (0.75 and 0.79 respectively) were also lower which was possibly due to the missing data for this domain because recruitment coincided with the school holidays.

Table 4-4 Internal Reliability Alpha Scores: Generic Core & Cancer Module

Generic Core (23 items)	N of Items	Self-Report E	Self-Report V	Proxy-Report E	Proxy-Report V
Total Score	23	**.95**	.92	**.96**	**.94**
Physical Health	8	**.93**	.88	**.93**	**.90**
Psychosocial Health	15	**.92**	.88	**.94**	**.92**
Emotional Functioning	5	.86	.75	**.91**	.88
Social Functioning	5	.81	.81	.85	.80
Study/Work Functioning	5	.88	.75	.89	.79
Cancer Module (27 items)					
Pain & Hurt	2	.75	.70	.85	**.91**
Nausea	5	.89	.89	**.95**	**.90**
Procedural Anxiety	3	.85	.84	**.98**	**.91**
Treatment Anxiety	3	.83	.88	**.94**	**.92**
Worry	3	.76	.80	**.91**	**.92**
Cognitive Problems	5	**.90**	.78	**.93**	.89
Perceived Appearance	3	.77	.70	.83	.89
Communication	3	.76	.77	.89	.79

Notes: 1. Cronbach's Alpha internal reliability scores
   1. V = Varni results (2002), E = Ewing results
   3. Minimum standard for group comparisons ≥ 0.7
   4. Recommended alpha score for individual analysis ≥ 0.9 (bolded)

The Cancer Module also compared well with Varni. The proxies consistently scored higher alpha values than the patient in each domain. The alpha coefficients in my sample ranged from 0.75 to 0.90, and 0.83 to 0.98 for self- and proxy-reports respectively, compared to Varni's greater range of 0.70 to 0.89, and 0.79 to 0.92.

4.3. Feasibility

4.3.1. Missing Data

HRQOL Questionnaire booklets:

Almost all eligible patients and proxies agreed to participate. If the first potential proxy was unavailable, the patient was usually able to nominate an alternative proxy. When only one booklet of the dyad pair was returned it still provided useful information, but could not be used for paired comparisons. This occurred in six cases where three patients and three proxies of different dyad pairs did not wish to participate. Only two complete patient-proxy dyads did not wish to take part.

Missing items:

There were no missing data for the 88 self-report Generic Core instruments, and few missing data for the Cancer Module. The 79 proxy-reports had few missing data in either

instrument. This was due to my strategy of follow-up phone calls (as described in Chapter 3) to check ambiguous or missing responses. Consequently, the overall percentage of missing data for the Generic instrument for young adults and proxy-reports was 0% and 0.1% respectively, compared with 0.4% and 0.3% for Varni's oncology sample.

Since few items were missing, the domain scales were calculated in most cases from completed items. For the Generic Core proxy-report, one respondent missed one physical item. In the Worry domain of the Cancer Module self-report, 12 respondents missed the same item and 6 in the proxy version missed the corresponding item: 'worry that the cancer will return or relapse' or wrote 'N/A'. These patients had a non-cancerous blood disorder, which made this item irrelevant to them. One patient missed the item about 'perceived physical appearance' and another proxy missed the item about difficulty solving mathematical problems.

Missing scale scores:

One proxy only omitted more than half (>50%) of the five data measurements required for calculating the Study/Work scale score of the Generic instrument. The resulting percentage of missing data for this Work/Study domain was equivalent to 1.1% and was calculated as follows:

$$1 \text{ case} \div 88 \text{ cases} * 100\% = 1.1\%$$

In contrast, for the same Work/Study domain, Varni reported 15% and 38% missing responses for child- and proxy-report respectively, due to administration of the survey during the summer vacation for one of the sites. The minimal missing data in my sample meant that participants could answer the questions and hence the study was feasible.

4.3.2. Missing socio-demographic and illness information

Ethnic group:

A number of participants confused ethnicity with nationality. Many selected 'Other' and wrote 'Australian' or 'British'. In cases where I had not met the patient at clinic or in hospital, I enquired of their ethnicity by telephone. When either of the patient/proxy dyad had stated 'European descent' and was the son/daughter or parent, I coded the other member of the dyad as being of the same ethnic group.

Respondents particularly those with Thalassaemia and Sickle Cell blood disorders gave numerous 'other' ethnic groupings e.g. Egyptian, Middle Eastern, Indian, Maltese. These I coded as European descent. This is not ideal, but I plan to include categories such as Mediterranean and African in the substantive study.

Illness Status:

The late introduction of Illness Status during field-testing explains why nine of the earlier questionnaires have missing values for this section.

Days absent:

Four patients had data missing for missed work/study, however, these could be estimated through other information that was given. For example, when one participant reported needing a carer for 30 days it was reasonable to assume that this patient had missed study/work during that period.

There were four proxies of different patient-proxy pairs from those above, whose days absent from work were estimated as zero because the patient reported that they did not miss any days of study/work due to their physical or mental health.

If participants responded to "days absent" by giving the number of weeks they were able to work in the year, this was converted to days absent on a Pro Rata basis. For example, if they reported being able to work only 4 weeks of whole year, then the missing data (days absent in the past 30 days) was calculated as follows:

$$\text{Days absent} = \frac{48 \text{ weeks (missed work)}}{52 \text{ weeks}} \text{ x } 30 \text{ days}$$
$$= 28 \text{ days missed work}$$

4.3.3. Floor/Ceiling Effects

The range and distribution of responses for each scale was examined. Table 4-5 shows the Generic Core Scale Score mean and standard deviation (SD) for groups with slight, moderate and severe symptoms, and percentage of scores at the extremities of the scaling range (i.e. floor and ceiling effects) for each domain of the PedsQL Generic

Core instrument. The mean and standard deviation of Varni's validation samples of 2001 and 2002 for his ill and healthy groups are included for comparison.

The average QOL scores for the slight, moderate and severe symptom groups decrease as severity of symptoms increases. This is explored further in Section 4.5 on 'known groups'.

The distribution of scores is important for decisions about the appropriateness of further analysis. For example, if no AYA patient had a problem with "taking a bath or shower by him or herself", the data would be bunched at one end of the distribution rather than spread across the full scale, and would violate the assumption of normality for factor analysis and influence estimates of correlation for any such item. When domain scores have large ceiling and floor effects they are rendered insensitive to differences and changes for patients at the floor and ceiling. The distribution of data can sometimes be so highly skewed and even bunched entirely at one end of the scale that analyses using medians may not help. In such cases either the item may need to be reworded but more probably the instrument for the target group may need to be reconsidered. Although the cohort excluded newly diagnosed patients (within 3 months post-diagnosis) and those requiring end-of-life care, the cross-sectional data indicated a wide range of health states within the sample frame.

The domain means of my sample were comparable with Varni's, and apart from Social Functioning in the self-report, my means fell between those of Varni's 2001 and 2002 samples. Varni's samples also contained participants who were healthy (control group) while my sample consisted of young adults who had been diagnosed with cancer or a blood disorder and were in varying states of wellness.

Varni reported virtually no floor effects, but ceiling effects ranged from moderate (e.g. 7% and 10% of healthy respondents respectively for self- and proxy-report Total Scale Score) to considerable (e.g. 47% and 58% of healthy respondents respectively for the self- and proxy-report Social Functioning Subscale). Similarly, my sample has no floor effects, but moderate to considerable ceiling effects were evident in both self- and proxy responses for the healthiest group (slight symptoms), and to lesser extent for the group with moderate symptoms. Interestingly, ceiling effects were more pronounced in the

proxy-responses than the self-responses for particularly the moderate symptom group indicating that proxies under-reported patient problems.

These results confirm that the ceiling effects were in the expected direction i.e. that young adults with minimal symptoms and their proxies reported a better QOL than those with severe symptoms (poorer state of health). Also, apart from young adults with severe symptoms in my sample, ceiling effects were consistently strong in the Social Functioning domain for all samples, but not enough to affect further analysis.

Table 4-5 Generic Core - Known Groups Descriptives: Self- and Proxy-Report

				Ewing Sample								Varni Sample				
Self-Report		Overall n = 88	Slight Symptoms n = 35			Moderate Symptoms n = 34			Severe Symptoms n = 10				Varni 2001				Varni 2002
	No. of items	Mean (SD)	Mean (SD)	%Floor	%Ceiling	Mean (SD)	%Floor	%Ceiling	Mean (SD)	%Floor	%Ceiling		Mean (SD) n=960	Ill %Floor	Ill %Ceiling	Healthy %Floor	Healthy %Ceiling	Mean (SD) N=219
Total Score	23	76 (19)	89 (13)	0	11	72 (15)	0	0	51 (9)	0	0		80 (15)	0	2	0	7	72 (16)
Physical Health	8	75 (25)	88 (16)	0	41	73 (24)	0	10	42 (15)	0	0		80 (19)	0	13	0	26	72 (22)
Psychosocial Health	15	76 (17)	89 (12)	0	14	71 (13)	0	0	55 (10)	0	0		79 (16)	0	5	0	12	73 (16)
Emotional Functioning	5	73 (20)	87 (15)	0	32	65 (15)	0	0	53 (19)	0	0		78 (21)	0	22	1	30	72 (21)
Social Functioning	5	88 (15)	93 (12)	0	68	86 (15)	0	31	74 (15)	0	8		84 (19)	0	33	0	47	77 (20)
Study/Work Functioning	5	69 (26)	87 (17)	0	27	61 (22)	0	0	38 (15)	0	0		76 (20)	0	13	1	23	69 (20) n = 191
Proxy-Report		n = 79	Slight Symptoms n = 35			Moderate Symptoms n = 34			Severe Symptoms n = 10				n =1622					n = 336
Total Score	23	74 (21)	84 (16)	0	23	73 (20)	0	3	47 (18)	0	0		81 (17)	0	4	0	10	70 (19)
Physical Health	8	75 (26)	83 (20)	0	37	74 (26)	0	15	48 (31)	0	0		81 (23)	2	19	0	40	69 (25)
Psychosocial Health	15	74 (21)	84 (17)	0	29	72 (20)	0	3	47 (14)	0	0		81 (17)	0	6	0	14	70 (18)
Emotional Functioning	5	69 (26)	80 (24)	0	40	66 (23)	0	12	41 (16)	0	0		78 (21)	1	20	0	30	68 (20)
Social Functioning	5	85 (18)	89 (16)	0	60	86 (18)	0	41	65 (17)	0	10		85 (19)	1	34	0	58	76 (21)
Study/Work Functioning	5	69 (27)	84 (18)	0	40	63 (26)	0	12	36 (16)	0	0		78 (22)	2	16	0	35	66 (23) n = 250

Notes: 1. % Floor / Ceiling = the percentage of scores at the extremes of the scaling range.

2. Varni 2001 and Varni 2002; note sample sizes for domains vary slightly due to missing data.

3. Slight, Moderate, and Severe Symptom groups as determined by MSAS-PHYS cut-off points at 0.2 and 1.

4. The reduced sample size for Study/Work in Varni 2002 is due to a clash between recruitment and school holidays.

4.4. Validity

Validity of the instruments was demonstrated through Exploratory Factor Analysis (EFA), correlations and known-groups methodology.

4.4.1. Factor Analysis

Construct validity was assessed by item-to-item correlations derived using factor analysis on both the patient and proxy versions of the PedsQL Generic Core and the PedsQL Cancer Module.

The factor structure generated from the modified PedsQL Generic Core and PedsQL Cancer Module, administered to young Australian patients (16-25 years old) and their nominated proxies was similar to that of the original instrument, administered to young Americans (13-18 years old) and their parent-proxies.

Since it is the pattern of the factors rather than the factor weights themselves that are important in the process of validating these instruments it was interesting that Varni's factor pattern for the Cancer Module, generated through focus groups and interviews alone, compared very well with my pattern of factors generated by an EFA.

4.4.1.1. PedsQL Generic Core

Table 4.6 contains factor weights for the Generic Core for patient and proxy scores with Varni's factor weights in the adjacent column for comparison. The underlying factor structure for both the self-report and proxy-report was similar to Varni's five-factor solution, except that for the proxy-report my data produced a four-factor solution. Both self- and proxy-reports accounted for 74% and 75% of the variance as compared with Varni's 52% and 62%, respectively.

To confirm the way that items grouped themselves in the EFA, I not only compared across the factor weightings for each item, to find on which factor the item was most strongly loaded, but also highlighted all factor weights greater than 0.6 and found similar loadings. Both methods led to domains similar to those of Varni for the PedsQL

Generic Core instrument. The following features of the patterns in the AYA patient and their proxy's responses are worth noting:

Self-report

In my sample, items of physical and emotional content load onto factors one and two respectively, but the order is reversed in Varni's sample. Apart from this the factors and absolute factor weights are consistent with Varni's and the a priori hypothesized factor structure, with a few exceptions as follows:

There was no apparent cross-loading in the Generic Core self-report in Varni's sample where one item was shared by two factors while in my sample minimal cross-loading occurred between items in Social and Physical Functioning, such as 'unable to do things peers can do', and items in Work/Study and Physical Functioning, such as 'missing study/work due to not feeling well'. Cross-loading is also apparent in Emotional Functioning with Social Functioning: 'feeling angry' and 'feeling blue'.

The splitting of factors is minimal. In my sample Social Functioning and Work/Study Functioning as demonstrated in the proxy-report are split cleanly between factors 1 and 4, and factors 1, 2 and 3 respectively, while in Varni's sample as shown in the self-report Physical Functioning (factors 1 and 2) and Work/Study Functioning (factors 4 and 5) are split.

Proxy-report

Three of the five items of Social Functioning (items 1 to 3) load onto factor 4 in my sample (E4) and factor 5 (V5) in Varni's sample while items four and five in both samples load onto Physical Functioning (Factor 1). These items are 'Hard to keep up with peers' and 'Unable to do things peers do'. The latter, in Varni's sample also cross-loads onto Physical Functioning and demonstrates the link involved between social and physical items.

Study/Work Functioning for Varni's sample (factor 2) and my sample (factor 3) are also split with factor 4 in Varni's sample and factors 1 & 2 in my sample. These two items are 'Missing Study/Work due to not feeling well' or due to a '…doctor or hospital appointment'.

Table 4-6 Generic Core Factor Analysis, Loadings per Item: Self- & Proxy-Report

Generic Core	Self-Report n = 88										Proxy-Report n = 79								
	1		**2**		**3**		**4**		**5**		**1**		**2**		**3**		**4**		**5**
	E1	V2	E2	V1	E3	V5	E4	V3	E5	V4	E1	V1	E2	V3	E3	V2	E4	V5	V4
Physical Functioning																			
1. Hard to walk more than one block	0.84	-0.72	0.09	-0.05	0.16	0.05	0.13	0.00	0.24	0.12	0.90	0.83	0.13	0.04	0.09	-0.11	-0.03	-0.02	0.02
2. Hard to run	0.76	-0.77	-0.08	0.10	0.19	0.12	0.12	-0.06	0.46	0.05	0.89	0.84	0.21	0.07	-0.08	-0.18	0.03	-0.01	0.04
3. Hard to do sports activity/exercises	0.76	-0.78	0.05	0.09	0.23	0.06	0.12	0.01	0.39	0.00	0.88	0.80	0.11	0.10	-0.01	-0.09	0.09	0.01	0.06
4. Hard to lift something heavy	0.69	-0.46	0.22	0.14	0.16	0.05	0.29	0.06	0.27	0.16	0.81	0.75	0.23	0.10	0.15	0.03	0.18	-0.08	0.04
5. Hard to take a bath/shower unaided	0.78	-0.57	0.27	-0.06	-0.12	-0.24	-0.04	0.01	-0.02	-0.14	0.63	0.72	0.02	-0.11	0.48	0.22	-0.14	-0.02	-0.08
6. Hard to do chores around house	0.88	-0.65	0.08	-0.04	0.11	-0.21	0.11	0.01	-0.05	-0.11	0.70	0.74	0.19	-0.02	0.26	0.19	0.25	-0.02	-0.07
7. Hurt or ache	0.65	-0.25	0.34	0.22	0.19	0.03	0.12	0.05	0.19	0.20	0.59	0.31	0.35	0.33	0.30	-0.15	0.15	-0.02	0.29
8. Low energy	0.68	-0.26	0.38	0.39	0.35	-0.03	0.10	0.03	-0.08	0.14	0.57	0.27	0.54	0.37	0.32	-0.03	0.06	-0.04	0.30
Emotional Functioning																			
1. Feel afraid or scared	0.19	-0.03	0.76	0.78	0.08	0.05	0.18	0.11	0.10	-0.18	0.17	0.04	0.82	0.72	0.12	0.00	0.29	0.04	-0.07
2. Feel sad or blue	0.21	0.10	0.68	0.78	0.11	-0.02	0.17	0.07	0.52	0.01	0.25	-0.02	0.80	0.77	0.35	0.05	0.15	0.08	-0.02
3. Feel angry	0.17	0.01	0.52	0.66	0.24	-0.11	-0.03	-0.04	0.61	-0.02	0.11	-0.04	0.72	0.66	0.38	0.09	0.07	0.10	-0.03
4. Trouble sleeping	0.30	-0.13	0.74	0.37	0.18	-0.18	0.08	0.05	-0.08	0.13	0.28	0.04	0.71	0.54	0.18	0.08	0.24	-0.01	0.16
5. Worry about what will happen	0.09	0.00	0.82	0.71	0.16	0.02	0.21	-0.07	0.14	0.04	0.24	-0.03	0.83	0.78	0.07	0.04	0.26	-0.02	-0.05
Social Functioning																			
1. Trouble getting along with peers	-0.02	0.04	0.32	0.01	0.21	-0.18	0.60	0.68	0.30	-0.08	0.13	0.04	0.29	0.09	0.33	0.25	0.62	0.60	-0.16
2. Other youth not wanting to be friends	0.13	0.02	0.15	-0.04	0.15	-0.05	0.84	0.81	0.05	-0.11	0.05	-0.04	0.38	0.06	0.07	0.02	0.82	0.84	-0.02
3. Teased	0.19	0.10	0.10	0.09	0.08	-0.01	0.83	0.75	0.06	0.05	0.15	-0.06	0.09	0.10	0.18	-0.10	0.83	0.81	0.08
4. Unable to do things peers do	0.60	-0.31	0.05	0.07	0.10	0.17	0.33	0.42	0.59	0.18	0.72	0.48	0.20	-0.08	0.36	0.02	0.28	0.41	0.13
5. Hard to keep up with peers	0.45	-0.35	0.12	-0.03	0.17	0.21	0.33	0.42	0.67	0.24	0.71	0.55	0.23	-0.13	0.32	0.15	0.26	0.37	0.12
Study/Work Functioning																			
1. Hard to concentrate class/at work	0.28	-0.06	0.16	0.12	0.83	-0.71	0.19	0.08	0.18	0.04	0.26	0.03	0.31	0.09	0.76	0.81	0.29	0.06	0.01
2. Forget things	0.04	0.00	0.19	0.21	0.86	-0.52	0.21	0.09	0.05	0.14	0.10	0.03	0.31	0.13	0.68	0.73	0.38	-0.03	0.08
3. Trouble keeping up with study/work	0.50	-0.12	0.11	-0.05	0.68	-0.68	0.07	0.14	0.22	0.20	0.26	0.02	0.42	0.04	0.72	0.77	0.26	0.05	0.15
4. Miss class/work - not feeling well	0.62	0.00	0.35	0.00	0.53	-0.14	0.10	-0.05	0.11	0.80	0.49	-0.04	0.53	-0.04	0.45	0.11	0.14	-0.05	0.90
5. Miss class/work - Dr or hospital appt.	0.66	0.06	0.30	-0.03	0.37	-0.07	-0.02	-0.01	0.17	0.85	0.60	-0.03	0.41	-0.02	0.41	0.07	-0.05	0.07	0.86
% of Total Variance	47%		10%		7%		6%		4%		51%		13%		6%		5%		

Note: Principal Components Analysis with Varimax Rotation and an Eigenvalue cut-off at 1.0

Additional cross-loading occurs in Varni's sample for the item "hurt" which loads onto Emotional as well as Physical Functioning. The "low energy" item in my sample also loads onto Emotional as well as Physical Functioning. This is possibly due to carer feelings of frustration and helplessness.

Hence, there was minimal cross-loading and splitting of factors in both Varni's and my sample thereby supporting a clear factor structure, and generation of the given domains.

4.4.1.2. PedsQL Cancer Module

Table 4-7 shows that the Cancer Module set of factor weights (loadings) for self- and proxy-report generated by my sample was best as a six-factor solution, which explained 76% and 84% of the variance for the patient- and proxy-report respectively. As Varni's eight-domain (27 item) instrument was derived through interviews and focus groups by his team and associates, there were no factor weights for comparison. Although an eight-factor solution (not shown) was also generated, the 6-factor model was a better fit with Varni's eight a priori domains. This suggests that the number of domains of the PedsQL Cancer Module could be reduced to an instrument with 6 or 7 domains for the proxy and self-report respectively. For example, the self-report could combine the domains for Treatment Anxiety and Worry, making 7 a priori domains. The instrument for the proxy-report could combine Procedural Anxiety and Treatment Anxiety into one 'Anxiety' domain, and the Pain & Hurt domain could be combined with Nausea into one domain, thereby making it an instrument of 6 domains.

There was one exception to the above fit and this was evident in the self-report scores for the Worry domain. A factor split and cross-loading occurred on the item 'worry that the medical treatments are working' and this was between factor 4 representing Pain/Hurt, and factor 3 representing Treatment Anxiety.

Some other interesting points of the self-report is that Cognitive Problems and Communication load onto factor 1, while Treatment Anxiety and Worry load onto factor 3, remembering that Worry also cross-loads onto factor 4 which it shares with the Pain domain. This interconnection gives some indication of the relationship between the level of pain, anxiety about treatment, and the overarching worry associated when a person has cancer or other serious illness.

For proxies, the items clustered very neatly into 6 factors. Procedural Anxiety and Treatment Anxiety loaded onto factor 1, while Pain and Nausea loaded onto factor 2.

This analysis of the factor structure confirms strong similarity to the structure of Varni's sample, and the construct validity of the instruments in an Australian AYA cancer context. Investigation of Convergent and Divergent validity follows through the use of various correlations.

Table 4-7 Cancer Module Factor Loadings per Item: Self- & Proxy-Report

Cancer Module	Adolescents & Young Adults						Proxy					
	1	2	3	4	5	6	1	2	3	4	5	6
Pain & Hurt Aches in joints and/or muscles	.143	.314	-.043	**.757**	-.025	-.022	-.174	**.652**	.108	.422	.160	.057
Having a lot of pain	.201	.484	.022	**.671**	-.010	.047	.049	**.711**	.140	.390	.232	.232
Nausea Become nauseated during medical treatments	.140	**.755**	-.014	.280	.283	-.131	.431	**.821**	.166	.109	.075	-.080
Food not tasting very good	.058	**.825**	.180	.227	.065	.059	.220	**.809**	.341	.019	.072	.029
Become nauseated while thinking about medical treatments	.199	**.602**	.243	.231	.447	-.078	.586	**.653**	.121	.252	.075	.117
Feeling too nauseous to eat	.287	**.710**	.087	.306	.088	.082	.366	**.788**	.204	.090	.187	.047
Some foods and smells cause nausea	.333	**.726**	.046	-.086	.071	-.005	.258	**.841**	.212	.077	-.036	.132
Procedural Anxiety Problems with Needle Sticks (injections, blood tests, IV) Hurt	.227	.175	.161	.148	**.807**	.100	**.847**	.284	.149	.140	.067	.141
Get anxious about Blood Tests	.058	.294	.506	-.116	**.658**	.205	**.880**	.129	.207	.149	.131	.176
Get anxious about Needle Sticks (i.e. injections, BT and IV's)	.155	.206	.431	.030	**.762**	.077	**.880**	.249	.191	.124	.075	.133
Treatment Anxiety Get anxious while waiting to see the Doctor	.273	.007	**.841**	.095	.195	.010	**.672**	.157	.119	.500	.271	.136
Get anxious about going to the doctor	.250	-.008	**.828**	.007	.306	.033	**.707**	.134	.147	.469	.289	.062
Get anxious about going to the hospital	.305	.036	**.702**	.215	.410	.034	**.684**	.317	.232	.409	.204	.077
Worry Worry about side effects from the medical treatments	-.086	.454	.207	**.557**	.255	.274	.454	.360	.070	**.653**	.139	.162
Worry about whether the medical treatments are working	.109	.198	**.534**	**.547**	.114	.173	.344	.283	.200	**.771**	.093	.117
Worry that the cancer will reoccur	-.061	.246	**.754**	-.028	.035	.276	.383	.099	.225	**.741**	-.015	.087
Cognitive Problems Difficulty figuring out what to do when something is bothersome	**.592**	.209	.319	.406	.107	.054	.269	.235	**.593**	.269	.332	.369
Trouble solving maths problems	**.791**	.149	.123	-.033	.202	.067	.123	.136	**.822**	.108	.212	.152
Trouble writing study papers or reports	**.834**	.219	.192	.044	-.021	.087	.139	.230	**.872**	.102	.225	.066
Difficulty paying attention to things	**.762**	.396	.056	.125	.054	.219	.169	.267	**.799**	.156	.290	.129
Difficulty remembering what he/she read.	**.831**	.260	.190	.006	.132	.016	.179	.244	**.800**	.116	.191	.132
Perceived Physical Appearance Feeling that he/she is not good looking	.163	.279	.440	-.183	-.229	**.574**	.306	.099	.511	.160	-.180	**.605**
Not liking other people to see his/her scars	.074	-.124	-.015	.220	.254	**.799**	.104	.040	.084	.102	.279	**.845**
Being embarrassed about others seeing his/her body	.116	.012	.203	.018	.054	**.894**	.162	.116	.234	.083	.185	**.851**
Communication Difficulty telling the doctors and nurses how he/she feels	**.598**	.069	.238	.459	.162	-.098	.180	.251	.440	.139	**.706**	.259
Difficulty asking the doctors and nurses questions	**.655**	-.117	.051	.519	.046	-.020	.215	.072	.417	.122	**.748**	.205
Difficulty explaining his/her illness to other people	**.539**	-.049	-.101	.461	.211	.244	.225	.171	.402	.060	**.768**	.159
% of total variance (initial Eigenvalue)	37%	12%	9%	7%	6%	5%	51%	11%	9%	5%	4%	4%

4.4.2. Correlations

As hypothesized, the strength of correlations in my sample was comparable to the strength of the corresponding correlations in Varni's sample. To demonstrate this the following correlations were examined:

Between domains within the Generic Core for AYA, and for proxies;

Between domains within the Cancer Module for AYA, and for proxies;

Between the Generic Core & the Cancer Module for AYA, and for proxies.

Patient-proxy concordance was checked using two further correlations:

Within the Generic Core between AYA and proxies;

Within the Cancer Module between AYA and proxies.

4.4.2.1. Convergent and Divergent Validity Correlations:

To check the convergent and divergent validity of the instruments for AYA and proxies the degree of agreement between measurements of the same domain were obtained. The correlations were as follows:

4.4.2.1.1. Between domains within Generic Core for self, and proxies

Table 4-8 displays the correlations between domains within the Generic Core, for self (below the diagonal) and proxies (above the diagonal). All correlations are significant at the 0.001 level (2-tailed) compared with Varni's correlations, which were significant at the 0.01 level (2-tailed). Correlation effect sizes are designated as high $r \geq 0.5$, medium $0.3 \leq r < 0.5$, and small $0.1 \leq r < 0.3$ (Cohen 1988 as cited in Varni et al., 2002a).

The Total (Health Summary) Score, as expected, is highly correlated with all of the domain scores for both the self-report ($r = 0.75$ to 0.95, first column) and the proxy-report ($r = 0.82$ to 0.95, first row) because it is a linear function of all the other domain scores.

Psychosocial Health Summary Score (HSS) is very highly correlated with the domains: Emotional, Social, and Study/Work functioning ($r = 0.84, 0.80, 0.89$ (3rd column) and $r = 0.90, 0.86, 0.93$ (3rd row) for self- and proxy-report, respectively), again because it is a linear function of these domains.

Table 4-8 Generic Core Correlations: AYA versus AYA and Proxy versus Proxy

			Proxy versus Proxy correlations			
Generic Core	**Total Score**	**Physical Health**	**Psycho-Social Health**	**Emotional Function**	**Social Function**	**Study-Work Function**
Total Score		0.88	0.95	0.82	0.85	0.89
Physical Health	0.92		0.70	0.54	0.68	0.66
Psychosocial Health	0.95	0.76		0.90	0.86	0.93
Emotional Functioning	0.75	0.53	0.84		0.65	0.72
Social Functioning	0.79	0.66	0.80	0.54		0.73
Study/Work Functioning	0.87	0.72	0.89	0.58	0.59	

(Left side label: **AYA versus AYA correlations**)

Notes:

1. AYA versus AYA Pearson's correlation coefficients are below the diagonal (in yellow) – Proxy versus Proxy correlations are above the diagonal (in green)
2. Correlation effect sizes: $r \geq 0.5$ high effect, $0.3 \leq r < 0.5$ medium effect, $0.1 \leq r < 0.3$ small effect
3. All significant correlations are at the 0.001 level (2-tailed)

The Physical HSS and the Psychosocial HSS do not contain any common items, yet the correlation is still high at $r = 0.76$ and 0.70 for the self- and proxy-reports respectively. This suggests that if a young person feels in good physical health they are also likely to have good emotional functioning, good social functioning and good study/work functioning. The Total Health Summary score is therefore a good summary of the patient's overall health, although the richness of the separate domains is lost in this single summary score.

Young adult patients report their Emotional Functioning as having the weakest correlation with Physical, Social and Study/Work Functioning ($r = 0.53, 0.54, 0.58$ respectively). Their proxies make a similar assessment for the Physical HSS ($r = 0.54$) but somewhat higher correlations with Social and Study/Work elements $r = 0.65, 0.72$ respectively, suggesting the proxies may overestimate the link between Emotional Functioning and these domains.

I hypothesized that the predominantly external domains Physical Health and Study/Work Functioning would correlate more highly with each other than with predominantly internal domains, i.e. Psychosocial, Emotional and Social Functioning, because issues

about physical concerns are external concepts, which are very different to those of an internalised nature. This hypothesis was not consistently supported by the data, perhaps because Work/Study Functioning straddles the boundary of internal and external issues, as in the item 'I have trouble keeping up with my study/work duties' and 'I miss study/work because of not feeling well'. The domains of the Generic Core demonstrated strong convergent validity due to the strength of agreement between measurements of the same domain. The next section investigates the validity of the Cancer Module.

4.4.2.1.2. Between domains within Cancer Module for self and proxies

Table 4-9 displays the correlations among the domains of the Cancer Module for AYA and for proxies. Overall, the correlation coefficients for the Cancer Module were not quite as strong as those for the Generic instrument, but most were of medium to high effect for both AYA and proxy. As expected within the Cancer Module, Cognitive Problems was highly correlated with Communication for both AYA patient self-report ($r = 0.65$, $p \leq 0.001$) and proxy ($r = 0.73$, $p \leq 0.001$).

Table 4-9 Cancer Module Correlations for AYA and their Proxies

	Cancer Module	Proxy versus Proxy correlations							
		P	N	PA	TA	W	CP	A	C
AYA versus AYA correlations	Pain & Hurt (P)		0.66	0.30**	0.41	0.53	0.46	0.31**	0.40
	Nausea (N)	0.53		0.60	0.61	0.58	0.53	0.33**	0.44
	Procedural Anxiety (PA)	ns	0.47		0.75	0.63	0.47	0.43	0.44
	Treatment Anxiety (TA)	ns	0.35	0.62		0.74	0.50	0.44	0.52
	Worry (W)	0.44	0.51	0.53	0.60		0.46	0.42	0.42
	Cognitive Problems (CP)	0.38	0.51	0.41	0.48	0.35		0.54	0.73
	Perc. Phys Appearance(A)	ns	ns	0.29	0.32	0.42	0.26*		0.51
	Communication (C)	0.49	0.37	0.29	0.39	0.30	0.65	0.23*	

Notes:
1. AYA versus AYA correlation (Pearson's) coefficients are below diagonal (in yellow) – Proxy versus Proxy are above the diagonal (in green)
2. Correlation Coefficients that were not statistically significant = ns
3. Correlation sizes $r \geq 0.5$ high effect, $0.3 \leq r < 0.5$ medium effect, $0.1 \leq r < 0.3$ small effect
4. All significant correlations are at the 0.001 level (2-tailed) except those marked * p-values < 0.05 and **p-values < 0.01

Self-report

Correlation coefficients (below the diagonal) for patients ranged from r = ns, then 0. 23 to 0.65. The highest correlations among AYA patients were between Cognitive Problems and Communications (r = 0.65, p≤0.001), Procedural Anxiety and Treatment Anxiety (r = 0.62, p≤0.001), and highly correlated with Worry were Treatment Anxiety (r = 0.60, p≤0.001), Procedural Anxiety (r = 0.53, p≤0.001), and Nausea (r = 0.51).

These correlations were as expected and clearly indicate the need for specialised multidisciplinary teams (MDT) caring for AYA who understand their developmental needs, concerns and issues, and who can actively assist AYA patients in the additional challenges confronting them as a consequence of their illness, treatment protocols and processes (Bennett, 2001; Bennett et al., 2005; Mitchell et al., 2004; White et al., 2004).

In addition, Worry correlated moderately well with Pain (r = 0.44), Appearance (r = 0.42), Cognitive Problems (r = 0.35) and Communication (r = 0.30). The likely reason the Worry-Pain correlation for AYA (r = 0.44) was only moderately high, could be due to participants with blood disorders, for whom pain is not usually an issue for their illness and treatment, such as Thalassaemia.

There were four AYA correlations that were not statistically significantly different from zero, and these correlations were not expected to be significant, as the literature does not support a relationship between these variables. AYA reported no association between Pain and Anxiety (Procedural and Treatment) or Appearance, and no association between Nausea and Appearance, thereby providing strong cases of divergent validity.

Proxy-report

The among-proxy correlations (above the diagonal of Table 4-9) were all of medium to high effect and ranged between r = 0.3 and 0.75. Like AYA, the highest correlations among proxies were between Procedural Anxiety and Treatment Anxiety (r = 0.75), and between Worry and Anxiety (Treatment, r = 0.74 and Procedural r = 0.63). These correlations were higher than those for the patients (0.62 and 0.60 respectively), which suggest that proxies believe there is a greater link between medical treatments and worry, more so than do the patients. It is possible that the high level of correlation with Worry as

reported by the proxy may also be a reflection of the concern, fear, isolation and helplessness caregivers may feel (Weller, 2004).

The correlation between Cognitive Problems and Communications for proxies (r = 0.73) was higher than for AYA (r = 0.65), which suggests that the greater the patient's problem with cognitive tasks the greater their difficulty in telling medical staff how they feel, asking them questions, or explaining to other people about their illness. These correlations re-enforce the need to provide MDTs trained in AYA health that include well-qualified and experienced specialists able to help AYA patients with their concerns and issues.

Correlations with a high effect were also observed between Worry and Pain (r = 0.53), and Worry and Nausea (r = 0.58) as expected. Accordingly, proxy perception of AYA level of worry about the side effects of treatment, whether the treatments are working, and whether the cancer will return are strongly related to pain and/or nausea. Other expected outcomes having medium effect are the correlation between Worry and each of Cognitive Problems (r = 0.46), Appearance (r = 0.42) and Communication (r = 0.42). Again the element of worry has a similar impact on proxy perception of cognitive problems, appearance and communication.

The above findings demonstrate good convergent and divergent validity of the PedsQL Cancer Module. The correlations between the Generic Core and the Cancer Module for both AYA and their proxies are explored in the next section.

4.4.2.1.3. Between Generic Core & Cancer Module for AYA & proxies

The Generic Core versus Cancer Module correlations ranged from not significant (ns) to correlations with a high effect (ns, then 0.21 to 0.74). As hypothesized there was no association in the self-report between: Social Functioning and each of Anxiety (Procedural and Treatment) and Perceived Physical Appearance; between Physical Health and Perceived Physical Appearance; and between Perceived Physical Appearance and Study/Work Functioning. Divergent validity was also demonstrated in the proxy-report between Procedural Anxiety and Physical Health.

Self-Report

The correlation values between the Generic Core and Cancer Module (CM) for self-report are shown in Table 4-10 and Figure 4.1, and were generally of medium to high effect (r = 0.30 to 0.74, p ≤ 0.05) apart from those which were not statistically significant (ns).

Table 4-10 Generic Core versus Cancer Module Correlations for AYA

		Generic Core					
	Self-Report n = 88	Total Score	Physical Health	Psychosocial Health	Emotional Function	Social Function	Study/Work Function
		Tot	**P**	**Psy**	**Em**	**Soc**	**SW**
Cancer Module	Pain and hurt (P)	**0.72**	**0.67**	**0.68**	**0.54**	**0.54**	**0.63**
	Nausea (N)	**0.71**	**0.63**	**0.69**	0.48	**0.52**	**0.72**
	Procedural Anxiety (PA)	0.30	0.21*	0.34	0.37	ns	0.34
	Treatment Anxiety (TA)	0.32	ns	0.40	0.45	ns	0.35
	Worry (W)	**0.58**	0.46	**0.61**	**0.60**	0.49	0.46
	Cognitive Problems (CP)	**0.59**	0.38	**0.68**	0.49	0.42	**0.74**
	Perc. Phys Appearance (A)	0.25*	ns	0.33	0.48	ns	ns
	Communication (C)	**0.51**	0.36	**0.57**	0.42	0.44	**0.57**

Notes:
1. Pearson's Correlation Coefficients, n = 88
2. Correlation sizes r ≥ 0.5 high effect (bolded), 0.3 ≤ r < 0.5 medium effect, 0.1 ≤ r < 0.3 small effect
3. Correlations which were not significant are denoted as 'ns'
4. All significant correlations are at the 1% level, p≤ 0.01, and '*' means that correlations are significant at the 5% level

Pain/hurt and Nausea (of the Cancer Module) were highly correlated with all domains of the Generic Core for AYA (r = 0.54 to 0.72, and 0.48 to 0.72 respectively, p ≤ 0.05).

Cognitive Problems (r = 0.38 to 0.74) and Worry (r = 0.46 to 0.61) were also correlated to a high to medium degree with the Generic Core domains. This clustering can be seen more clearly in Figure 4.1 (below), which shows that Psychosocial HSS and Work/Study Functioning from the Generic Core (r = 0.34 to 0.74) tend to be more highly correlated with the domains of the Cancer Module; while Anxiety (Procedural and Treatment r ≤ 0.37 and ns) and Appearance (r ≤ 0.45 and ns) from the Cancer Module tend to have only small effect correlations with Generic Core.

The correlation between Cognitive Problems and the domains of the Generic Core have the greatest range (0.38 to 0.74), but these are all still of high to medium effect. The highest correlations of Cognitive Problems with the other domains are with Study/Work

Functioning (r = 0.74, p< 0.001), Psychosocial HSS (r = 0.68, p< 0.001) and the Total Health Summary Score (r = 0.59, p< 0.001). This result is as expected and adds further strength to the validation.

Figure 4-1 Generic Core versus Cancer Module Correlations for AYA
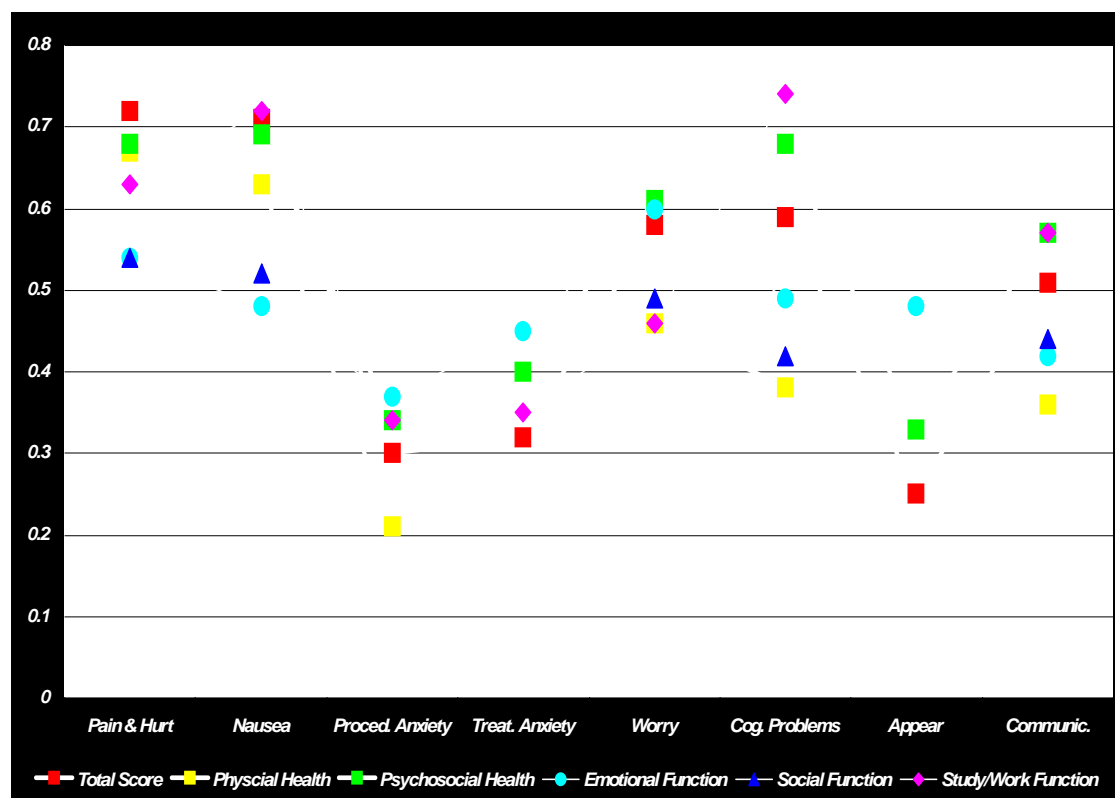
Source: PedsQL^{TM} 4.0 Generic Core and the PedsQL^{TM} 3.0 Cancer Module, Self-report versions

Proxy-Report

Table 4-11 provides the data for the proxy-report correlations between the Generic Core and Cancer Module, which may be clearer in Figure 4.2 below. All these correlations were of medium to high effect (p < 0.001 for most correlations), with one exception: there was no association between Physical Health and Procedural Anxiety, similarly for the self-report data where only a small effect was evident. Again, similar to the Self-Report, Pain/hurt and Nausea (Cancer Module) correlated highly with the domains of the Generic Core (r = 0.55 to 0.80, p ≤ 0.05).

Psychosocial HSS (0.48 ≤ r ≤ 0.77, p < 0.001) and Emotional Functioning (0.49 ≤ r ≤ 0.72, p < 0.001), a subset of Psychosocial HSS, correlated highly with the Cancer Module domains. These were more highly correlated than were AYA Psychosocial HSS (0.33 ≤ r

≤ 0.69, p< 0.002) and Emotional Functioning (0.37 ≤ r ≤ 0.60, p< 0.001) suggesting that these aspects for proxies could be influenced by proxy anxiety.

Table 4-11 Generic Core versus Cancer Module Correlations for Proxies

		Generic Core					
	Proxy-Report **n = 79**	**Total** **Score**	**Physical** **Health**	**Psychosocial** **Health**	**Emotional** **Function**	**Social** **Function**	**Study/Work** **Function**
		Tot	Phy	Psy	Em	Soc	SW
Cancer Module	**Pain and Hurt (P)**	**0.80**	**0.72**	**0.75**	**0.68**	**0.61**	**0.71**
	Nausea (N)	**0.68**	**0.55**	**0.69**	**0.57**	**0.59**	**0.68**
	Procedural Anxiety (PA)	0.39	ns	0.48	**0.51**	0.28*	0.46
	Treatment Anxiety (TA)	0.49	0.28*	**0.56**	**0.61**	0.40	0.48
	Worry (W)	**0.58**	0.42	**0.61**	**0.72**	0.39	0.49
	Cognitive Problems (CP)	**0.66**	0.37	**0.77**	**0.66**	**0.62**	**0.77**
	Perc. Phys Appearance (A)	0.46	0.30	**0.52**	0.49	0.40	0.48
	Communication (C)	0.48	0.23*	**0.59**	**0.52**	0.46	**0.58**

Notes:
1. Pearson's Correlation Coefficient sizes r ≥ 0.5 high (bolded), 0.3 ≤ r < 0.5 medium, 0.1 ≤ r < 0.3 small effect
2. All significant correlations are significant at the 1% level of significance p≤ 0.01, and '*' means that the correlation is significant at the 5% level.

Figure 4-2 Generic Core versus Cancer Module Correlations for Proxies
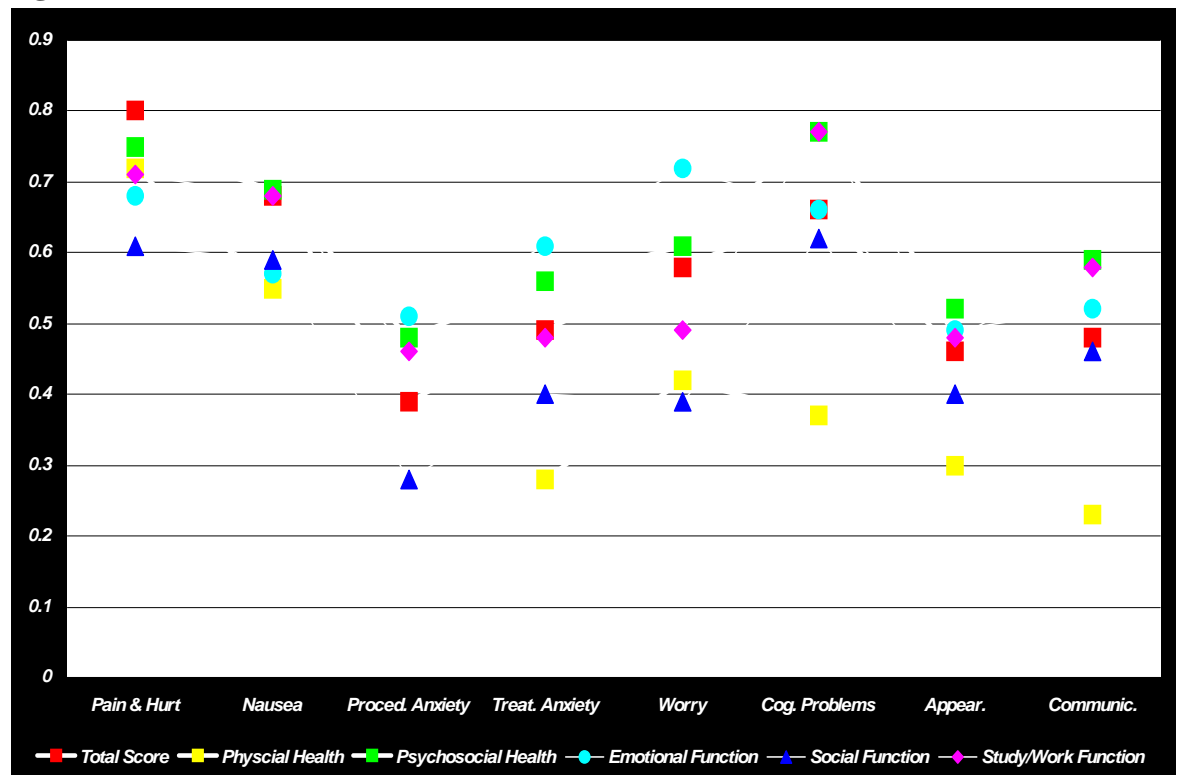
Source: PedsQL™ 4.0 Generic Core and the PedsQL™ 3.0 Cancer Module, Proxy-report versions.

4.4.2.2. Patient –Proxy Concordance

To determine whether other people are able to validly assess patient HRQOL the level of agreement between the patient and their nominated proxy was investigated through cross-informant variance:

(i) Patient-proxy correlations, and (ii) Mean differences for each of the scales of the Generic Core and the Cancer Module, that provide particular insights and permit answers to the following questions:

    a.   How good are proxies at observing and reporting the health of AYA patients?

    b.   How closely correlated are the AYA patient and proxy-reports of HRQOL?

4.4.2.2.1. Correlations within Generic Core between AYA & proxies

Table 4-12 shows the self versus proxy correlations of the PedsQL Generic Core. The most interesting information is on the leading diagonal, which provides the patient-proxy correlation for each domain within the Generic Core. The correlations range from r = 0.56 to 0.85 (p ≤ 0.001) and indicate good patient-proxy concordance. This suggests that proxies are generally good at assessing these particular aspects of young adult patient HRQOL.

Emotional Functioning has the lowest correlation value (r = 0.56) for the self-proxy inter-correlation, which suggests that the nominated proxies of these young adult patients were less able to assess the emotional status of these patients. The external domains may mislead proxies: Social Functioning and Study/Work Functioning, if they use these to help assess emotional functioning of young adults. This result also suggests confirmation that the emotional turbulence, which is often characteristic of AYA, remains an aspect of youth health where young people themselves need to be consulted (Ministry of Health, 2002; Ministry of Youth Affairs, 2002).

The lowest correlation in Table 4-12 is between the proxy assessment of the young adult's Emotional Functioning against self-report Social Functioning (r = 0.37) and vice versa (r = 0.43) is also relatively low. These correlations reflect some of the difficulties inherent for proxies in assessing AYA patient HRQOL. Social Functioning can be influenced by the dynamics between Physical Health and Emotional Functioning, thus making the assessment task more difficult for the proxy.

Table 4-12 Generic Core AYA versus Proxy inter-correlations

		Proxy - Report					
		Total Score	Physical Health	Psycho-social Health	Emotional Function	Social Function	Study-Work Function
AYA Self - Report	Total Score	**0.81**	**0.77**	**0.73**	**0.58**	**0.63**	**0.76**
	Physical Health	**0.77**	**0.85**	**0.62**	0.47	**0.56**	**0.64**
	Psychosocial Health	**0.75**	**0.63**	**0.74**	**0.60**	**0.61**	**0.76**
	Emotional Functioning	**0.55**	0.41	**0.58**	**0.56**	0.43	**0.53**
	Social Functioning	**0.62**	**0.60**	**0.55**	0.37	**0.62**	**0.54**
	Study/Work Functioning	**0.73**	**0.60**	**0.72**	**0.54**	**0.54**	**0.81**

Notes:
1. Pearson's Correlation Coefficient sizes: r ≥ 0.5 high effect (bolded), 0.3 ≤ r < 0.5 medium effect, 0.1 ≤ r < 0.3 small effect
2. The leading diagonal (shaded & underlined) contains the correlations between the AYA and their nominated proxy for each domain
3. All correlations are significant at 0.001 level (2-tailed) p ≤ 0.001

In summary, patient-proxy concordance for all corresponding subscales produced inter-correlations of medium to large effect sizes, as hypothesised. But, how good are proxies at observing the health of young adult patients? This close correlation indicates that proxies are fairly good at observing the health of AYA patients, but their observations do not always coincide with those of the patients themselves. Furthermore, correlations cannot reflect any systematic differences or biases between patients and proxies; these are reflected in the differences between means as presented in Table 4-13 and are also compared with the respective values generated by Varni's sample.

Direction of association of AYA-proxy differences in Generic Core means

Only one of the mean differences between self and proxy was statistically significant from zero, although another was almost significant. This shows there are no large biases, but it is important also to note that in every case the patients' mean scores were higher than the proxies'. This phenomenon is consistent with the literature on proxy measurements. Whether it is a doctor observing a patient, or a parent or caregiver observing the health of their charge, the observer will usually rate HRQOL lower than the patient, themselves (Osoba et al., 2005, p141). It is suggested since HRQOL is "essentially subjective that it

is the individual who experiences that life who is best placed to report on it" (Osoba et al., 2005, p139). However, there are several possible reasons for these differences, for example to reduce their perception of the burden on family and carers, or to please health care staff patients may report their symptoms and psychosocial concerns as being low, or it could be that patients adapt to their illness (Osoba et al., 2005, p141). This apparent "response shift" phenomenon may occur where the person experiencing the loss of health has a psychological coping mechanism in place to readjust their internal coping standard so that they don't feel so bad (King, 2001; Schwartz et al., 2005 in Fayers, 2005 p276 ; Sprangers et al., 1999 as cited in Fayers 2005, p276).

Although the mean differences of the samples were comparable, the spread of Varni's differences (range of SD = 24 to 33) was about twice that of my sample (range of SD = 13 to 22). So while there was about the same degree of bias on average (and in both cases it was small on average), the degree of discrepancy at the individual dyad level was greater in the American sample and less in the Australian sample.

Table 4-13 Direction of Association: Generic Core AYA-Proxy Differences

Generic Core	Self-Report		Proxy-Report		Differences	
	E Mean(SD) n= 88	V Mean(SD) n= 219	E Mean(SD) n= 79	V Mean(SD) n= 336	E Mean(SD)	V Mean(SD)
Total Score	77 (19)	72 (16)	75 (21)	70 (19)	2 (13)	3 (25)
Physical Health	76 (25)	72 (22)	75 (26)	69 (25)	1 (14)	3 (33)
Psychosocial Health	77 (18)	73 (16)	74 (21)	70 (18)	3 (15)	2 (24)
Emotional Functioning	73 (21)	72 (21)	69 (26)	68 (20)	4 (22)[+]	4 (30)
Social Functioning	89 (15)	77 (20)	85 (18)	76 (21)	4 (15)*	1 (29)
Study/Work Function	69 (27)	69 (20)`	69 (27)	66 (23)`	0 (17)	2 (30)

Notes: 1. V is Varni's instrument and sample 2002
2. E is Ewing's sample using Varni's modified instrument
3. Reject the null hypothesis of equality * p = 0.037, + a weak difference p = 0.079
4. Varni's sample size for the Study/Work domain was 191 self-report and 250 proxy-report
4. Rounding to the nearest whole number may have resulted in rounding errors.

Convergent validity is reflected in the Study/Work scale because the items forming this domain have more of an external nature and lend themselves to be measured more readily by proxies or observers (r = 0.81 with minimal mean difference 0(SD = 17), compared

with Varni's medium effect correlation, r = 0.36 and mean difference = 2(SD = 30)). The larger mean difference and weaker correlation in Varni's sample could be due to a significant proportion of his participants being on holiday.

Are the differences statistically significant?

The largest mean difference between patient and proxy in both samples was in assessing the internal domain, Emotional Functioning (mean difference (SD) = 4(22), p = 0.079, and 4(30) for Varni's sample), and this was the domain with the smallest correlation between self and proxy in my sample (r = 0.56) and in Varni's sample (r = 0.54). This demonstrates divergent validity, which again indicates the need to ask young adults themselves about how they feel, as it is hard for proxies or observers to assess the emotional status of AYA patients.

In my sample overall, only one domain, Social Functioning, demonstrated a statistically significant difference between the proxy and self-report (mean (SD) = 4(15), p = 0.037 and the correlation coefficient r = 0.62 p ≤ 0.001). This indicates that overall proxies nominated by the AYA patient can give a fairly reasonable assessment of AYA QOL though they tend to slightly under-estimate their charges' own reports of QOL. The next question of interest was: is there a difference between AYA-proxy assessment in an older AYA and younger AYA? This is considered in the next section.

Is there a significant difference between older and younger patients?

Table 4-14 gives the pattern of differences between the paired self-proxy assessments for a younger AYA patient group (16-19 years, n = 45 pairs) and an older AYA patient group (20-25 years, n = 34 pairs) for the PedsQL Generic Core. While the older group of the Generic Core showed no statistically significant difference between patient and proxy, the younger group demonstrated a significant difference in most domains between patient and proxy QOL assessment at the 5% level. The domains were Total Score (p=0.032), Psychosocial Health (p=0.035), Emotional Functioning (p=0.028), and Social Functioning (p = 0.031).

Proxies of the 16-19 age-group tended to report significantly worse HRQOL than the patient themselves at the 5% level (p<0.05). These results suggests that proxies of the older group were reasonably good at assessing AYA HRQOL and were more accurate

than the proxies of the younger age-group for making these assessments. The next section investigates AYA-proxy concordance and thus proxy validity within the Cancer Module.

Table 4-14 Generic Core Self-Proxy Paired Comparison, p-values for AYA

Generic Core (p – values)	16 – 19yr Proxy< Self, n = 45 pairs	20 - 25yr Self< Proxy, n = 34 pairs
Total Score	0.032	ns
Physical Health	ns	ns
Psychosocial Health	0.035	ns
Emotional Functioning	0.028	ns
Social Functioning	0.031	ns
Study/Work Functioning	ns	ns

Notes: 1. Paired comparison between self- and proxy-report
         2. ns =not statistically significant

4.4.2.2.2. Correlations within Cancer Module between AYA & proxies

Table 4-15 gives the Self-Proxy intercorrelations among the 8 domain scales of the Cancer Module. Apart from the leading diagonal, the correlations were mainly of small to medium effect ranging from $r = 0.23$ to $r = 0.5$, and a number of correlations were not significant (ns), and these were generally as expected. For example, there was no significant correlation between proxy-assessed Pain with: self-assessed Worry; self-assessed Anxiety (Procedural or Treatment); or self-assessed Perceived Physical Appearance. In contrast the strongest correlation, $r = 0.5$ was between proxy Procedural Anxiety and self-report Nausea ($r = 0.5$, $p = 0.001$), and proxy Procedural Anxiety with patient Treatment Anxiety ($r = 0.5$, $p = 0.001$).

Self-Proxy correlations, leading diagonal:

As for the Generic Core, the most interesting information is in the leading diagonal, the patient-proxy concordance correlation for each domain. The correlations are medium to large, ranging from $r = 0.36$ to 0.67 ($p \leq 0.001$). These results suggest that proxies are reasonably good at assessing these aspects of the health of AYA patients, but not as good as the patients themselves. The Self-Proxy correlation for the Worry domain ($r = 0.36$) was the weakest and suggests a discrepancy between the AYA patient's assessment of their worry and the proxy's assessment of the patient's level of worry. To find whether there was any systematic bias in this variation I considered the mean (SD) differences between patient and proxy responses for each domain.

Table 4-15 Cancer Module Self -Proxy Inter-Correlations

		Proxy - Report							
		P	N	PA	TA	W	CP	A	C
Self - Report	**Pain & Hurt (P)**	**0.54**	0.33	ns	ns	0.31	0.31	0.23*	0.28*
	Nausea (N)	0.43	**0.67**	**0.50**	0.48	0.43	0.46	0.32	0.31
	Procedural Anxiety (PA)	ns	0.25*	**0.59**	0.41	0.39	0.32	0.28*	0.27*
	Treatment Anxiety (TA)	ns	0.27*	**0.50**	0.48	0.37	0.30	0.27*	0.31
	Worry (W)	ns	0.33	0.38	0.28*	0.36	0.23*	0.40	ns
	Cognitive Problems (CP)	0.25*	0.34	0.37	0.39	0.35	**0.65**	0.43	0.46
	Perc. Phys Appearance(A)	ns	ns	ns	ns	ns	ns	**0.64**	ns
	Communication (C)	0.26*	0.28*	ns	0.34	0.26*	**0.50**	0.36	**0.60**

Notes: 1. Pearson's Correlation Coefficients, n = 79
2. Correlation sizes: r ≥0.5 high effect (bolded), 0.3≤ r<0.5 medium effect, 0.1≤ r <0.3 small effect
3. The leading diagonal (shaded & underlined) consists of the correlation coefficients between the patient and their nominated proxy for each domain.
4. Most significant correlations are at 0.01 level of significance (2-tailed) i.e. $p ≤ 0.01$ and $*p ≤ 0.05$

Differences between Self- and Proxy-report for the Cancer Module:

Table 4-16 presents the means (SD) of self- and proxy-reports and the self-proxy differences. In the Australian sample, half of the self-report domain scores were higher than the corresponding proxy score, suggesting that in the Generic Core scales proxies tended to overestimate their charges' problems. In contrast, in Varni's sample, proxies gave higher assessments, suggesting they under-estimated the problems faced by AYA patients in all domains except for Pain, Procedural Anxiety and Treatment Anxiety. However, paired sample t-tests on these mean differences failed to reach statistical significance.

These differences were not significant, however the possibility of confounding due to age will be explored at another time because such an analysis does not fit within the validation of the PedsQL instruments and will be the subject of a future paper.

The strength of the correlations within the instruments between AYA, and between Proxies, and the level of AYA-proxy concordance were as hypothesised, while the AYA-

proxy differences were comparable with Varni's sample. Clinical validity is the final test required to establish the validity of the model in an Australian context, and this is investigated in the next section using 'known groups' analysis to test its sensitivity.

Table 4-16 Direction of Association: Cancer Module AYA-Proxy Differences

Cancer Module Scale Scores	Self-Report[1]		Proxy-Report[1]		Differences[2] (Self – Proxy)	
	E n = 88	V n =216-220	E n = 79	V n =327-334	E	V
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean(SD)	Mean(SD)
Pain and hurt	75 (24)	76 (25)	75 (25)	75 (26)	0 (23)	2 (36)
Nausea	84 (21)	76 (23)	83 (25)	78 (24)	1 (19)	-2 (33)
Procedural Anxiety	80 (29)	68 (31)	81 (28)	60 (33)	-1 (26)	8 (45)
Treatment Anxiety	80 (28)	82 (25)	77 (27)	72 (28)	3 (29)	11 (37)
Worry	69 (23)	70 (27)	68 (27)	76 (28)	1 (29)	-6 (39)
Cognitive Problems	73 (25)	71 (22)	76 (27)	74 (22)	-3 (22)	-4 (31)
Perc. Phys Appearance	73 (25)	70 (24)	74 (26)	76 (25)	-1 (22)	-6 (35)
Communication	78 (24)	74 (25)	77 (26)	78 (24)	1 (22)	-4 (34)

Notes: 1. The higher the score the better the HRQOL
2. A positive difference suggests the proxies overestimated the patients' QOL problems
3. A negative difference suggests the proxies underestimate the problem
4. E – Ewing sample; V – Varni's sample
5. The number of items per domain is given with the domain name.

4.5. Sensitivity - Clinical Validity ('Known Groups' analysis)

As mentioned in Sections 3.1.2 and 3.5.4, to investigate the sensitivity of the instruments to clinically important differences and thus prove their clinical validity, AYA patients were classified via the MSAS, into 'known groups' of slight, moderate and severe symptom experience 'in the past week'.

As expected, the HRQOL scores for each domain were inversely related to the level of symptoms experienced by AYA patients. That is, the QOL scales followed the health gradient described by symptom severity groups 1 to 3 (slight, moderate and severe) where increasing symptoms translated to decreasing QOL. Young adult patients with low levels

of symptoms reported higher PedsQL™ scores than those with high levels of symptoms. The proxy-report scores indicated a similar pattern of results.

4.5.1. PedsQL™ Generic Core

Table 4-17 and Table 4-18 provide the means and standard deviations (SD) of each symptom severity subgroup by domain for the self-report and proxy–report, respectively, and provide comparisons between symptom severity groups. The Significant Differences column indicates that the instruments were sufficiently sensitive to detect most differences between symptom severity groups. This was evidenced by the ANOVA where most groups were statistically significantly different to each other ($p \leq 0.01$) in both the self- and proxy-reports.

Table 4-17 Generic Core: Adolescent/Young Adult, Symptom Severity Groups

Generic Core AYA n = 88	Slight n = 37	Moderate n = 39	Severe n = 12	Significant Differences*	
	Mean (SD)	Mean (SD)	Mean (SD)	Groups	P values
Total Score	89 (13)	72 (15)	51 (9)	1 2 3	**1 v. 2, p < 0.001** **2 v. 3, p < 0.001** **1 v. 3, p < 0.001**
Physical Health Score	88 (16)	73 (24)	42 (15)	1 2 3	**1 v. 2, p = 0.007** **2 v. 3, p < 0.001** **1 v. 3, p < 0.001**
Psychosocial Functioning	89 (12)	71 (13)	55 (10)	1 2 3	**1 v. 2, p < 0.001** **2 v. 3, p = 0.001** **1 v. 3, p < 0.001**
Emotional Functioning	87 (15)	65 (15)	53 (19)	1 2 --- 3	**1 v. 2, p < 0.001** 2 v. 3, p = 0.053 **1 v. 3, p < 0.001**
Social Functioning	93 (12)	86 (15)	74 (15)	1-----2 3	1 v. 2, p = 0.074 **2 v. 3, p = 0.027** **1 v. 3, p < 0.001**
Study/Work Functioning	87 (17)	61 (22)	38 (15)	1 2 3	**1 v. 2, p < 0.001** **2 v. 3, p = 0.001** **1 v. 3, p < 0.001**

Note:
1. One-way ANOVA
2. Symptom severity groups: 1 = Slight, 2 = Moderate, and 3 = Severe symptom group
3. The Differences* column indicates the level of significance between symptom severity groups p<0.05 are bolded, whereas e.g. Social Functioning "**1---2 3**" means there is a weak difference between slight and moderate symptom groups (unbolded, 1 v. 2, p = 0.074).

For interpreting the Significant Differences column, groups 1,2 and 3 equate to Slight, Moderate and Severe symptom groups respectively. Dotted lines between groups e.g. in Table 4-17 for self-report, Social Functioning domain groups '1---2' indicate a weak significant difference between groups. Underlining of groups indicates the groups were not significantly different, e.g. in Table 4-18 the proxies report no statistically

significant difference between slight and moderate symptom groups (i.e. '1   2') for Social Functioning (1 v. 2, p = 0.744) or the Physical Health domain (1v. 2, p = 0.3).

For interpreting the Significant Differences column, groups 1, 2, and 3 equate to Slight, Moderate and Severe symptom groups respectively. Dotted lines between groups e.g. in Table 4-17 for self-report, Social Functioning domain groups '1---2' indicates a weak significant difference between groups. Underlining of groups indicates the groups were not significantly different, e.g. in Table 4-18 the proxies report no statistically significant difference between slight and moderate symptom groups (i.e. '1 2') for Social Functioning (1 v. 2, p = 0.744) or the Physical Health domain (1 v. 2, p = 0.3).

Table 4-18 Generic Core: Proxy-Report on Symptom Severity Groups

Generic Core Proxy n =79	Slight n = 35	Moderate n = 34	Severe n = 10	Significant Differences*	
	Mean (SD)	Mean (SD)	Mean (SD)	Groups	P - values
Total Score	84 (16)	73 (20)	47 (18)	1 2 3	**1 v. 2, p = 0.029** **2 v. 3, p = 0.001** **1 v. 3, p < 0.001**
Physical Health Score	83 (20)	74 (26)	48 (31)	<u>1 2</u> 3	1 v. 2, p = 0.3 **2 v. 3, p = 0.01** **1 v. 3, p < 0.001**
Psychosocial Functioning	84 (17)	72 (20)	47 (14)	1 2 3	**1 v. 2, p = 0.011** **2 v. 3, p = 0.001** **1 v. 3, p < 0.001**
Emotional Functioning	80 (24)	66 (23)	41 (16)	1 2 3	**1 v. 2, p = 0.03** **2 v. 3, p = 0.008** **1 v. 3, p < 0.001**
Social Functioning	89 (16)	86 (18)	65 (17)	<u>1 2</u> 3	1 v. 2, p = 0.744 **2 v. 3, p = 0.002** **1 v. 3, p < 0.001**
Study/Work Functioning	84 (18)	63 (26)	36 (16)	1 2 3	**1 v. 2, p < 0.01** **2 v. 3, p = 0.002** **1 v. 3, p < 0.001**

Note:
1. One-way ANOVA
2. Symptom severity groups: 1 = Slight, 2 = Moderate, and 3 = Severe symptom group.
3. The Differences* column indicates the significance level between symptom severity groups, p<0.05 are bolded, whereas e.g. Social Functioning "**1   2   3**" means there is no significant difference between slight and moderate symptom groups (unbolded,1 v. 2, p = 0.744).

It is interesting to note that the highest QOL scores for both self- and proxy-report were in Social Functioning, and Social Functioning remained consistently the highest score compared to other domains across each symptom severity group, and ranged from 74(15) to 93(12) and 65(17) to 89(16) for self and proxy respectively. On the other hand, Study/Work Functioning was shown to have the lowest mean QOL score for all symptom groups of the self-report ranging from 38(15) to 87(17), and most symptom groups as

reported by proxy 36(16) to 84(18). The low study/work scores suggest a high level of concern and difficulty faced by many AYA patients who may be struggling to complete or further their education, or find appropriate employment to achieve financial independence. Study/Work Functioning was the only domain that breached the homogeneity of variance assumption for the Generic Core and was considered not to adversely affect the sensitivity of this instrument. As previously found, the AYA patient usually reported a slightly higher QOL score than the proxy.

These results provide strong evidence of the sensitivity of the PedsQL^{TM} Generic Core, as it was able to differentiate between groups of different symptom experience, and thereby prove its clinical validity. The final section of this validation study explores the sensitivity of the Cancer Module.

4.5.2. PedsQL^{TM} Cancer Module

The same 'known groups' as identified by MSAS were utilised for testing the sensitivity of the Cancer Module. However, due to breaches of homogeneity of variance for the Cancer Module over most domains I used the Kruskal-Wallis (K-W) Non-Parametric test to check for differences between the symptom groups. This test indicated that there was a significant difference between most symptom groups ($p < 0.001$) within each domain. To extract more detail about the level of significance between each pair of groups within each domain, the Mann-Whitney U test was used.

Table 4-19 details the domains that were in breach of the homogeneity of variances assumption. The Levene Statistic shows that the variances were significantly different in all domains except for Pain/Hurt ($p = 0.233$, $p= 0.208$), and Worry ($p = 0.159$, $p= 0.382$) for AYA and proxy respectively, and proxy Treatment Anxiety proxies only ($p = 0.496$).

The QOL scores for the Cancer Module follow the health gradient. Table 4-20 and Table 4-21 give the mean and standard deviation (SD) of each symptom group by domain of the Cancer Module for self and proxy respectively, as well as whether significant differences were evident between these groups. As in the Generic Core, QOL was inversely related to symptom level for both self- and proxy-report. The exception was Perceived Physical Appearance as reported by the young adults where groups 2 and 3 were tied (2 3). The Significant Differences column worked in a similar way as

explained for the Generic Core but with a variation due to the non-parametric nature of the Cancer Module data.

Table 4-19 Cancer Module: Test of Homogeneity of Variances

Cancer Module	Self-Report				Proxy-Report			
	Levene Statistic	df1	df2	Sig. p =	Levene Statistic	df1	df2	Sig. p =
Pain and hurt	1.483	2	85	.233	1.602	2	76	.208
Nausea	9.276	2	85	**.000**	6.388	2	76	**.003**
Procedural Anxiety	15.369	2	85	**.000**	5.873	2	76	**.004**
Treatment Anxiety	10.980	2	85	**.000**	.707	2	76	.496
Worry	1.882	2	85	.159	.975	2	76	.382
Cognitive Problems	3.541	2	85	**.033**	2.758	2	76	**.070**
Perceived Phys. Appearance	3.488	2	85	**.035**	2.506	2	76	**.088**
Communication	5.372	2	85	**.006**	4.445	2	76	**.015**

Note: The Levene Statistic was considered significant when p < 0.1, (bolded).

Self-Report

The Significant Differences column under "Groups" in Table 4-20 shows a clear distinction between patient symptom severity groupings for Pain, Nausea, and Worry and a very significant difference between patients reporting slight and moderate symptoms in all domains. A statistically weak significant difference was evident between moderate and severe groups for Procedural Anxiety (2 v. 3, 2---3, p = 0.098), but there was no significant difference between moderate and severe symptom groups for Treatment Anxiety, Cognitive Problems, Perceived Physical Appearance and Communication i.e. 2 3, all were tied.

It is noted that the worst QOL scores of the severe symptom group for self-report were: Worry 47(26), Nausea 48(21), and Pain 50(23). This was an expected result since worry tends to increase with symptoms such as pain, and most AYA had a cancer diagnosis requiring the usual cancer treatments, which cause nausea.

Proxy-Report

The Kruskal-Wallis Test shows that there is a significant difference between symptom severity groups (p ≤ 0.028) for all domains except Communication. Also the Mann-Whitney U Test (see Table 4-21) shows that a clear split (under Groups) is evident

between pairs of these symptom groups within each domain, except between moderate and severe symptom groups for Perceived Physical Appearance (2   3), and a weak difference for Cognitive Problems (2---3, p = 0.068).

Table 4-20 Cancer Module: AYA Symptom Severity Groups

Cancer Module AYA n=88	Slight n = 37	Moderate n = 39	Severe n = 12	Significant Differences*		
	Mean (SD)	Mean (SD)	Mean (SD)	Groups	p - values Mann-Whitney	Krus-W
Pain & Hurt	88 (18)	71 (22)	50 (23)	1 2 3	1 v. 2, p<0.001 2 v. 3, p=0.014 1 v. 3, p<0.001	p<0.001
Nausea	95 (11)	84 (16)	48 (21)	1 2 3	1 v. 2, p<0.001 2 v. 3, p<0.001 1 v. 3, p<0.001	"
Procedural Anxiety	93 (15)	75 (31)	56 (38)	1 2---3	1 v. 2, p=0.002 2 v. 3, p=0.098 1 v. 3, p<0.001	"
Treatment Anxiety	92 (16)	73 (31)	65 (32)	1 2 3	1 v. 2, p=0.001 2 v. 3, p=0.442 1 v. 3, p =0.001	"
Worry	80 (20)	67 (18)	47 (26)	1 2 3	1 v. 2, p=0.001 2 v. 3, p=0.007 1 v. 3, p<0.001	"
Cognitive Problems	88 (16)	64 (23)	54 (28)	1 2 3	1 v. 2, p<0.001 2 v. 3, p=0.163 1 v. 3, p =0.001	"
Perceived Physical Appearance	84 (19)	63 (27)	68 (28)	1 2 3	1 v. 2, p=0.001 2 v. 3, p=0.546 1 v. 3, p = 0.048	p=0.003
Communication	92 (16)	69 (23)	63 (25)	1 2 3	1 v. 2, p<0.001 2 v. 3, p=0.481 1 v. 3, p<0.001	p<0.001

Notes:
1. The Kruskal-Wallis non-parametric test, used to decide significance between symptom groups
2. Mann-Whitney U Test used for detail of significance between pairs of groups within domains.
3. Significant Differences between symptom severity groups: 1 = Slight, 2 = Moderate, 3 = Severe.
4. Symptom severity cut-off points determined by MSAS-PHYS at 0 - 0.2; 0.2 - 1; 1+

The poorest HRQOL scores in the severe symptom group as reported by the proxies were in keeping with the self-report i.e. Worry 34(23), Nausea 43(28), and Pain 46(28), but in addition to these the proxies also rated Procedural Anxiety 48(33) as being of more concern than did AYA 56(38), which is most likely due to proxy concern and feelings of helplessness.

The above investigations have confirmed that the instruments are sensitive to clinically important differences between groups of patients in the AYA and proxy versions of the

Generic Core and Cancer Module and hence the Clinical validity of these instruments is confirmed.

Table 4-21 Cancer Module: Proxy-Report on AYA Symptom Severity Groups

Cancer Module Proxy n = 79	Slight n = 35	Moderate n = 34	Severe n = 10	Significant Differences		
	Mean (SD)	Mean (SD)	Mean (SD)	Groups	p - values	
					Mann-Whitney	Krus-W
Pain & Hurt	86 (18)	72 (23)	46 (28)	1 2 3	1 v. 2, p=0.009 2 v. 3, p=0.009 1 v. 3, p<0.001	P<0.001
Nausea	94 (16)	84 (20)	43 (28)	1 2 3	1 v. 2, p=0.009 2 v. 3, p<0.001 1 v. 3, p<0.001	P<0.001
Procedural Anxiety	92 (17)	80 (28)	48 (33)	1 2 3	1 v. 2, p=0.037 2 v. 3, p=0.005 1 v. 3, p<0.001	P<0.001
Treatment Anxiety	88 (22)	74 (25)	51 (29)	1 2 3	1 v. 2, p=0.003 2 v. 3, p=0.020 1 v. 3, p=0.001	P<0.001
Worry	79 (22)	66 (24)	34 (23)	1 2 3	1 v. 2, p=0.016 2 v. 3, p=0.001 1 v. 3, p<0.001	P<0.001
Cognitive Problems	85 (20)	73 (28)	56 (31)	1 2---3	1 v. 2, p=0.041 2 v. 3, p=0.068 1 v. 3, p=0.002	P=0.005
Perceived Physical Appearance	82 (21)	69 (28)	63 (30)	1 2 3	1 v. 2, p=0.027 2 v. 3, p=0.506 1 v. 3, p=0.029	P=0.028
Communication	84 (21)	74 (27)	64 (36)	1---2 3	1 v. 2, p=0.075 2 v. 3, p=0.439 1 v. 3, p=0.097	P=0.101

Notes:
1. The Kruskal-Wallis non-parametric test, used to decide significance between symptom groups
2. Mann-Whitney U Test used for detail of significance between pairs of groups within domains.
3. Significant Differences between symptom severity groups: 1 = Slight, 2 = Moderate, 3 = Severe.
4. Symptom severity cut-off points determined by MSAS-PHYS at 0 - 0.2; 0.2 - 1; 1 – 4.

Chapter 5

5. Conclusion

In concluding, I summarise my work, note its implications and limitations, and speculate on further research.

This study presents the measurement properties for Varni's instruments PedsQL™ 4.0 Generic Core and PedsQL™ 3.0 Cancer Module (Varni et al., 2002a) modified for adolescent and young adult (AYA) aged patients and their nominated proxy. At the time of the study's commencement, I considered these to be the most appropriate generic and cancer specific HRQOL measurement instruments for three reasons: they included role functioning items that were appropriate to this age range; they had been shown to be feasible, reliable and valid in paediatric populations; and they were brief. The instruments' limitation is in their neglect of issues concerning sexuality and fertility. However, I plan to include such domains in the proposed substantive study.

Recruitment through outpatient clinics, in-patient clinics and via telephone was shown to be feasible at three Sydney hospitals, and an initial letter of introduction proved helpful. There were more AYA females in each age-group (M: F = 37:51); a greater number of younger patients than older young adults (48 v. 40), and about three-quarters of the AYA patients had a cancer diagnosis.

Cronbach's Alpha internal consistency reliability values for both instruments were the same or better than Varni's alpha values. The study values for the PedsQL Generic Core ranged from 0.81 to 0.96 inclusive for self-report and proxy-report. Values for the PedsQL Cancer Module ranged from 0.75 to 0.90 for self-report and 0.83 to 0.98 for proxy-report. This demonstrated good reliability and consistency, exceeding the recommended minimum alpha coefficient standard of 0.70 for group comparisons. The PedsQL Generic Core Total Score for both self-report and proxy-report exceeded the alpha value of 0.90 recommended for individual patient analysis, thereby making the Total Score suitable as a summary score for primary analysis of HRQOL outcome and useful in clinical trials and for other group comparisons. The Physical and Psychosocial Health Summary Scores are recommended for secondary analyses, while the subscales

Emotional, Social and Study/Work Functioning can be used to investigate specific domains of functioning.

The items of the Generic Core and Cancer Module had minimal missing responses, illustrating that participants were willing and able to provide good quality data about the AYA's HRQOL, albeit with some follow-up prompting. There were no floor effects and minimal ceiling effects, which was in keeping with Varni's results and other HRQOL studies.

Construct validity was demonstrated through factor analysis, correlations and descriptive statistics. The factor structure for the PedsQL Generic Core was comparable to that of Varni's sample where the data fell into clear factors with minimal cross-loading or splitting of factors. The factor analysis of the Cancer Module for the study sample supported a clear factor structure, which generated the given domains comparable with the factor groupings that Varni had arrived at through focus groups and interviews alone.

The correlations between domains within the PedsQL Generic Core for the self-report and proxy-report indicated that the physically based external domains correlated significantly more highly with each other than the internal, psychological domains. Apart from the Total Score, the self-versus-self correlations tended to be smaller than the proxy-versus-proxy correlations for both the Generic Core and the Cancer Module. The Total Score provided a good summary score and correlated highly with all domains, which was expected as it was a linear function of the domain scores. Similarly, Psychosocial Functioning, which was a linear function of Emotional, Social and Work/Study Functioning, correlated highly with these three domains. In addition, Physical Functioning correlated highly with Psychosocial Functioning, suggesting that AYA who feel good physically are likely to have good emotional functioning, good Social Functioning and good Study/Work Functioning. Furthermore, the internal domain Emotional Functioning has the smallest correlations with Physical, Social and Work/Study Functioning indicating the difficulty involved in assessing Emotional Functioning of AYA.

The correlations within the PedsQL Cancer Module between domains for the self-report and for the proxy-report were not as high as those of the Generic Core but were nevertheless medium to high in size. The proxy-versus-proxy correlations were

consistently the same or higher than the self-versus-self correlations. The highest AYA correlations were between Cognitive Problems and Communication, Procedural Anxiety and Treatment Anxiety, and between Worry and Treatment Anxiety and as expected, were reflected similarly by the proxies. However, pain-worry correlation was only moderately high which is possibly due to a proportion of the sample having a blood disorder.

Patient-proxy concordance as assessed by correlations tended to be moderate to high among the scales of the PedsQL Generic Core and somewhat weaker degree among the Cancer Module scales. However, proxies tended to assess HRQOL at a poorer level than the patient themselves for all the Generic Core scales, and this bias was more pronounced among the younger age-group. The older group (20-25years old) demonstrated no significant difference between self and proxy of the Generic Core while the younger group demonstrated significant differences in Emotional Functioning, Social Functioning, Psychosocial Functioning and the Total Score. Emotional Functioning and Worry (Cancer Module) further demonstrated the inherent difficulty proxies have in assessing the internal dimensions for AYA patients, and further highlights the need to ask the patient for their perspective of their HRQOL whenever possible, while assessment of the external dimensions (i.e. Physical Health Summary Score and Study/Work domain) could be achieved with more accuracy. Although patient self-report is the standard for measuring HRQOL, the proxy's perception, particularly in the younger 16-19 age-group, may be an influential factor in health care utilisation, and can be used if the patient is unable to complete the HRQOL assessment. However, the difficulties in assessing the internal domains of AYA, such as emotional functioning, need to be considered when implementing HRQOL research studies.

The correlations between the scales of the Generic Core and those of the Cancer Module, for both self-report and proxy-report, demonstrated convergent validity in terms of the high correlation of the causal variables, Pain and Nausea (Cancer Module) with the QOL domains of the Generic Core (Fayers et al., 2000). The correlation between Psychosocial HSS and Emotional Functioning was much greater for the proxy responses than those of the AYA. A possible explanation is that proxy assessment of AYA HRQOL may be influenced by proxy anxiety for the AYA patient, however this cannot be tested with my data, as I did not have measures of proxy anxiety. Divergent validity was demonstrated in

the self-report where there was no association between: Social Functioning and three domains, Procedural Anxiety, Treatment Anxiety and Perceived Physical Appearance; Appearance and Physical Health; and Appearance and Study/Work Functioning. For the proxy-report divergent validity was demonstrated between Procedural Anxiety and Physical Health only.

Discriminant validity was demonstrated by the sensitivity of the PedsQL Generic Core and PedsQL Cancer Module instruments to hypothesised gradients in QOL expected in groups of patients with slight, moderate and severe symptom experience. MSAS was used to define these "known Groups". The results were consistent with the conceptualisations of disease-specific symptoms as causal indicators of HRQOL in that HRQOL was inversely related to symptom level (Fayers et al., 2000, p46 & p66) reported by AYA and their proxies. This was shown in the ANOVA utilised for the PedsQL Generic Core, and the Kruskal-Wallis Test and Mann-Whitney U test for the PedsQL Cancer Module, i.e. increasing symptoms translated to decreasing QOL. (Notably, the AYA usually reported a slightly higher QOL score than the proxy even in these symptom strata.) The results provided strong evidence of the clinical validity of the PedsQL^{TM} Generic Core and the Cancer Module, as these instruments were sufficiently sensitive to differentiate between groups of different symptom experience.

In overview these results provide strong evidence that the instruments exhibit adequate validity, reliability and sensitivity for use in the Australian context with the target group of 16-25 year old AYA. The results will be strengthened further in regard to illness burden, when the Impact Report, Preferences for Support Services and Satisfaction Surveys are also analysed in conjunction with the HRQOL instruments.

The limitations of the instruments are that they been field tested only in Australia. However, their content is likely to generalise to New Zealand, the United Kingdom, America, Canada and Europe due to similarities in culture and health services. Further research and field-testing is required, particularly after the inclusion of sexuality and fertility domains. Test-retest reliability and responsiveness were not reported, as this work was conducted within the time and budgetary constraints of a Masters research project. The sample was one of convenience and information on non-participants was limited, so generalisability could not be fully assessed. Finally, Varni's instruments (held by the

MAPI Research Trust, France) require author permission and have licensing costs attached except for unfunded student research.

To achieve better social and psychological outcomes for cancer patients, and improved quality of life and survival, the scope and application of research need to be widened and a mobilisation of activities and resources needs to take place (Weller, 2004). Weller, also warns of the current fragmentation of behavioural and social science research, and of little cross-country participation, and that further change needs to be generated in health systems and health policy, and partnerships fostered to advocate for improved resources (Bleyer, 2002b; Weller, 2004). Appropriate models of care for AYA with cancer need to be developed and implemented (White, 2002), with possible extension through to 35year olds. Also, for positive change to occur, change in the attitude of some physicians toward AYA is needed (Bennett et al., 2005). The Multi-Disciplinary Team (MDT) needs to be able to relate to AYA and provide assurances and understanding of the difficulties they face. In addition the education of the public, the health providers, legislators, insurers (Bleyer, 2002b) and educational institutions needs to take place to better cater to the needs of AYA. Furthermore, in addition to the required medical treatment, the integration of resources, infrastructure and support from expert researchers to attract investment, and better coordination of activities (Weller, 2004) is essential for an AYA patient and family centred model of care that incorporates all "influential factors in adolescent behaviors, so approaches must be developed which cross education, social services and the justice system" (Viner et al., 2005)

An encouraging and recent development is that the theme of the International Psycho-Oncology Society (IPOS) Conference 2006 asserts "an imperative need for a continuous dialogue and constructive interaction among health care professionals involved in cancer care, through their active participation in research, education and clinical assistance". This theme highlights further the strong need for an AYA research emphasis and framework to build, develop and constantly update improved knowledge about the psychosocial needs of AYA patients and their families; plus appropriate data collection to enable ease of identification of AYA patient needs and monitor progress.

This thesis focuses on the validation of the HRQOL instruments modified for the AYA patient group to assist in the provision of a set of appropriate instruments for further

investigation of this age-group of patients – a small but vital part in the quest for better health outcomes for AYA with cancer and blood disorders. These new versions of the PedsQL™ Generic Core and the PedsQL™ Cancer Module for AYA patients have demonstrated construct validity, clinical validity and reliability for use as outcome measures in clinical trials, clinical practice and future health research into adolescents and young adults in the Australasian and international context.

Where to next in the bigger AYA picture?

The development of key policy documents such as NSW Health's Optimising Cancer Care in NSW (NSW Department of Health, 2003), the NZ Cancer Control Strategy (New Zealand Ministry of Health et al., 2003), the NZ Cancer Control Strategy: Action Plan 2005-2010 (Cancer Control Taskforce, 2005), and Australia's Senate Committee Report on Cancer (Senate Community Affairs References Committee Secretariat, 2005) suggest that stakeholders are commencing mobilisation for a collaborative effort to provide timely, age-appropriate support (emotional, psychological, social, spiritual, financial, palliative care and rehabilitation – education, employment and insurance needs) for reducing the illness burden on AYA patients, siblings, parents and carers living with cancer. In addition to policy, early diagnosis, easy entry into appropriate clinical trials, age-appropriate facilities staffed by health professionals trained in AYA health and advocacy will certainly help in conjunction with the development of AYA tools, such as these for measuring AYA HRQOL, satisfaction, preferences for different types of support, the impact of illness and treatment, and a needs assessment to gather essential data. AYA instrument development is a vital step in providing guidance for addressing the issues and unmet needs of AYA cancer patients and their families, and hence potentially reduce illness burden.

However, progress for AYA will continue to be fraught with difficulty without a national policy on transition care for AYA, clinical guidelines for the psychosocial care of AYA cancer patients and families, adult and paediatric health service collaboration on AYA care and treatment, development of an AYA multi-disciplinary centre of excellence for a cancer (and blood disorder) service in each state and territory of Australia, and in New Zealand that can provide local delivery of expertise in psychosocial support and treatment (where it is safe to do so), AYA focused cancer and palliative care nurses and rehabilitative support for AYA patients to continue or resume

study and/or work. However, this must be matched with appropriate funding to implement these milestones and changes, and provide on-going research and evaluation to monitor and guide improvement and address the current shortfall in care, positive outcomes and facilities for AYA living with cancer.

This AYA population has a right to be supported in a way that is appropriate to their developmental needs. This could be achieved through Centres of Excellence to oversee the treatment (including access into clinical trials for AYA) and support of AYA by clinical and psychosocial experts at local delivery (where this is possible and safe), with close collaboration between the health system (seamlessly between paediatric, adult, and AYA services), the education system, the justice system, WINZ/Centrelink and support from current and future employers and insurers to develop a comprehensive and integrated model of care that can also cope with providing appropriate and timely information and psychosocial support, and can deal with the issues of fertility, relationships, palliative care and post-treatment care.

In addition, given the poor survival of patients 15-40 years old (Figure 1-1), it might be appropriate to extend the AYA age-group to include patients up to 35years and stratify into about four age-groups for comparison.

Finally, since significantly more AYA will become survivors as treatment improves, they too will need support in planning a future after cancer, a chance to fulfil their personal goals and dreams, and live to become productive members of society, enriching communities, and providing encouragement to the growing number of first and second generation cancer patients.

Glossary

Access. The ability of people to make use of health services and support available.

Adolescence. The period between childhood and maturity (Oxford Dictionary) and for the purposes of this study to be roughly the period between 12 & 25 years of age.

Adolescent. A young person aged approximately between 12 and 25 years old. See also youth, young adult.

Blood disorder. A general term to describe the full range of disorders and diseases of the blood.

Cancer. A disease where malignant growths form.

Caregiver. A voluntary person or carer is usually a family member who looks after a person with a health problem or disability and who is unpaid. As defined in the NZ Cancer Control Strategy 2003, p67 (New Zealand Ministry of Health et al., 2003)

Ceiling and floor effects. This is a clustering of scores at either end of a scale, and results from a lack of precision at the extremities, which may cause scale insensitivity in cross-sectional data, and lack of responsiveness to change in longitudinal data (as it is not possible to distinguish between participants at the extremes).

Centrelink. Provider of social security payments and services to the Australian public.

Chemotherapy. The treatment or control of cancer using anti-cancer drugs.

Chronic illness. People with any on-going long-term or recurring condition that can have a significant impact on a person's life.

Co-morbidity. An additional illness or disorder e.g. Downs Syndrome.

Construct validity. The degree to which an instrument measures the construct it was designed to measure. Construct validity refers to the degree to which inferences can legitimately be made from the operationalisations in your study to the theoretical constructs on which those operationalisations were based (Fayers, Machin, p50).

Convergent validity. Correlation among latent variables that are hypothesised to be similar.

Correlation. The mean of the product of the standardised data values.

Cronbach's alpha. This is a measure for checking the consistency of answers from participants within each scale (Internal Reliability). It is a summary statistic about items summarising across scales.

Cross-informant variance. Patient and proxy give a similar assessment of patient health.

Dimension. An aspect of measurement. Each dimension may consist of several items.

Discriminant validity. Discriminates between known groups by a lack of correlation between groups known to be different.

Divergent Validity. A lack of correlation between latent variables, which are hypothesised as dissimilar.

Dyad. The patient and proxy pair.

Eigenvalues. A measure of how much of the variation in the data is accounted for by each factor, thus indicating the importance of each by explaining the variability and correlations in the data sample. They are usually scaled so that the sum of the eigenvalues equals the number of items.

End-of-life care. Care given to a person needing more than Palliative Care in preparation for dying.

Factor Analysis. A statistical technique that attempts to identify groups of variables in a correlation matrix such that there are strong correlations amongst all the variables within a group but weak correlations between those outside the group.

Greater Metropolitan Clinical Taskforce (GMCT) has an ongoing commitment to facilitating clinical networks to improve health care in NSW. GMCT manages networks set up by the Greater Metropolitan Transition Taskforce (GMTT) and new clinical networks approved by the Director-General, NSW Health.

Working groups of dedicated clinicians (doctors, nurses and allied health professionals) specialising in particular areas, together with consumers and managers, guide the process of consultation, research and planning.

Following the Greater Metropolitan Services Implementation Group's report, the taskforce made numerous recommendations for improvements to the public hospital system. With $64.6 million in recurrent funding from the Government, many of these improvements are now being implemented.

Health-Related Quality of Life (HRQOL) emphasizes the domains of physical and psychological well-being.

Health status. A description and/or measurement of the health of an individual or population.

Incidence of cancer. Number of new cancer cases.

Instrument. A device for measuring.

Internal reliability. Measure for checking the consistency of responses to items within a scale (see Cronbach's alpha, p116).

Items. Questions in a survey or questionnaire.

Known Groups. Known to differ by clinical criteria.

Late-effects. Any adverse effect that does not resolve after completion of therapy or any new problem that becomes evident after completion of therapy. Most of these effects are not detectable at completion of therapy but become evident some time later (www.emedicine.com/ped/topic2591.htm accessed 30/06/2006).

Leukaemia. Cancer of the blood and although it is a type of blood disorder, I have classified it under cancer.

Likert scale. A set of ordered categories with labels such as, "never", "almost never", "sometimes", "often", and "almost always".

Measurement instruments. Tools, such as surveys and questionnaires for measuring outcomes.

Morbidity. Illness or disease.

Mortality. Death.

Multidimensional construct. An amalgamation of two or more domains for example, to explain HRQOL where the domains may target physical, emotional and social functioning.

Multidisciplinary team (MDT). This consists of nurse physician, social worker, physiotherapist, occupational therapist, pharmacist, bereavement counsellor, spiritual worker and dietician (New Zealand Ministry of Health, 2001).

Palliative care. An approach for improving the quality of life of patients and families facing the problems associated with life-threatening illness, through the prevention and relief of suffering by means of early identification, assessment and treatment of pain and other problems, physical, psychosocial and spiritual.

Parent. Young adults may still be partially or wholly dependent, financially and/or emotionally, on an older adult or caregiver, usually a biological parent, foster parent, grandparent, aunt or uncle, guardian, or may be a sibling, or spouse. I use the term "parent" to denote this person whom the young person nominates.

Patient. The adolescent or young adult (AYA) with the cancer or blood disorder.

Patient-Proxy dyad. The research participant pair consisting of the AYA patient and their nominated caregiver.

Proxy. In this case, a parent or caregiver as nominated by the patient. In this context the proxy could be the patient's biological mother or father, a grandparent, a foster parent, spouse, caregiver, sibling or any other very close and respected adult.

Psycho-Oncology. The study, understanding and treatment of social psychological, emotional, spiritual, quality-of-life and functional aspects of cancer as applied across the cancer continuum (Minister of Health, 2003).

Quality of Life (QOL). An individual's perception of their position in life in the context of the culture in which they live, and in relation to their goals, expectations and standards. The term incorporates concepts of physical and psychological well-being, levels of independence and autonomy, social relationships and support, and spirituality. HRQOL emphasizes the domains of physical and psychological well-being.

Radiation therapy. The use of radiation to destroy cancer cells.

Reliability. See Cronbach's alpha p117.

Response shift. The phenomenon where patients with progressive disease may tend to adjust or recalibrate their assessment of their HRQOL in response to worsening health.

Responsiveness to change over time. The ability of a scale to detect changes to a patient's condition i.e. when a patient improves or deteriorates – similar to sensitivity (Fayers et al., 2000).

Sensitivity – 'known groups' analysis is the ability to detect differences between groups known to be different.

Stakeholders. Organisations/groups with a direct interest and involvement in aspects of cancer control.

Strategy. A plan or course of action for achieving targets.

Support services. Support for patients and their families in terms of their psychosocial needs e.g. emotional, psychological, financial, rehabilitative and spiritual needs.

Transition Care is the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-orientated health care systems.

Transition Care for Young People with Chronic Childhood Illnesses (TCYPCCI). The Transition Care for Young People with Chronic Childhood Illnesses Group was convened following a proposal put to GMTT for the development of a co-ordinated Sydney-wide network of centres interested in the management of thalassaemia patients (a blood disorder). Stakeholders, including paediatric and adult clinicians and consumers, met in December 2002 to identify the issues arising during the transfer of care from the paediatric setting to adult health care services. Consultation revealed that complex problems were experienced by AYA from a range of specialty and special needs groups.

A successful proposal for recurrent funding resulted in appointment of a Program Manager in May 2004. Three Transition Coordinators commenced in October/Nov 2004. These positions are based at Royal Prince Alfred, John Hunter and Westmead Hospitals. They work closely with the tertiary paediatric units within the 3 state-wide paediatric networks. The initial focus is on identifying gaps in transition services and on working closely with clinicians, young people and their families to determine what is needed.

Thalassaemia. A blood disorder usually requiring daily intervention.

Varimax rotation. A method for explaining the percentage of variability - the resulting factor weights are equal to the correlations between the factors and the items, and helps with identification of interpretable patterns. Varimax is one of the most commonly used methods, and attempts to minimise the number of variables with high loadings on each factor to simplify the overall structure.

Young person. A person aged between 12 and 25. This age range is based on the definition used in the Youth Advisory Council Act 1989 (See Youth below).

Youth. A synonym for 'young people' or adolescent and young adult (AYA) aged people. Due to inconsistency in definitions of 'adolescent', 'young person', 'young people', 'young adult' and 'youth', for simplicity in this study we will consider these terms as synonyms unless stated otherwise.

Young adult. Young adults are a subset of young people, defined for this study as being aged between 16 and 25 years (see youth).

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Appendices

Appendix A. Cancer incidence in NZ & Australia

Appendix B. The Centre for Children's Cancer & Blood Disorders (CCCBD) Family Newsletter

Appendix C. (i) Form letter to potential participants treated at Prince of Wales Hospital and the Royal Hospital for Women.

(ii) Participant Information Sheet, Consent Form & Questionnaire for:

      a. AYA Self-Report Survey Pack

      b. Proxy-Report Survey Pack

Appendix A

Table A- 1 New cancer diagnoses in Australia of AYA 12-24years inclusive, for the period 1997-2001

Year	Males	Females	Totals
1997	162	134	296
1998	160	144	304
1999	147	135	282
2000	169	130	299
2001	178	141	319

Notes: All cancers excluding non-melanoma skin cancers (squamous cell carcinomas of the skin and basal cell carcinomas of the skin).

Source: Elizabeth Tracey, Manager NSW Central Cancer Registry.

Table A- 2 Incidence of all cancers (ICD-10 C00-C97), persons aged 12-24, for the period 1997-1999 in Australia

Year	Males	Females	Totals
1997	521	478	999
1998	491	435	926
1999	487	445	932

Table A- 3 NZ Cancer Registrations 1994-99, Total Registrations Sex by Age

Year	Age brackets			
	10 - 15	15 - 20	20 - 25	10 - 25
1994				
Total	34	59	117	**210**
Male	16	31	57	104
Female	18	28	60	106
1995				
Total	39	62	113	**214**
Male	16	39	60	115
Female	23	23	53	99
1996				
Total	33	57	104	**194**
Male	16	27	60	103
Female	17	30	44	91
1997				
Total	40	50	79	**169**
Male	21	28	44	93
Female	19	22	35	76
1998				
Total	31	47	104	**182**
Male	15	20	50	85
Female	16	27	54	97
1999				
Total	36	63	105	**204**
Male	26	27	53	106
Female	10	36	52	98

Notes: Source: New Zealand Health Information Service (NZHIS)

The mean number of cancer incidences over the 6 years 1994-1999 is 195.5 cases/year. This equates to approximately 200 young people aged 10-25 years, are diagnosed with cancer each year in New Zealand.

Appendix B

Latest Research: Support Services for Young People 16 to 24 years

A new project has started in the Centre for Children's Cancer and Blood Disorders (CCCBD) at Sydney Children's Hospital, in conjunction with the University of Technology, Sydney (UTS).

The project focuses on a frequently overlooked age group: patients aged 16 to 24 years who often feel caught between the paediatric and adult healthcare groups. We are looking at how we might be able to reduce the impact of serious illness on young people and their families, and whether or what type of support services need to be developed. So don't be surprised if you hear from us if you have child in this age category.

One of the main aims of this study is to identify the most desirable mix of support services from both the patient and caregiver perspective. Participation involves completing some tick-the-box style questionnaires and to write comments about the personal impact of illness and treatment. The questionnaires cover topics such as Quality of life, and satisfaction with services. An exciting aspect of this project is that, in asking about which types of support services families prefer, we will be using a questionnaire method new to health sciences, but commonly used in marketing, business and economics. It requires respondents to simply choose between various combinations of possible support services.

We have already begun data collection in the CCCBD outpatient clinics and wards, and plan to commence telephone recruitment shortly - hopefully before you receive this newsletter!

If you don't hear from us, and would like to take part, then please contact Jane Ewing from UTS,   or Belinda Goodenough in the CCCBD (   ).

We'll plan to give updates of this study to the Family Newsletter. We hope that all participants enjoy contributing to this important research, and we look forward to meeting many of you in the near future.

Source: Centre for Children's Cancer & Blood Disorders (CCCBD) Family Newsletter

Appendix C

1. Form letter to potential participants treated at the Prince of Wales Hospital and the Royal Hospital for Women.

2. The Survey Pack contents:

❖ **Self-report**

- o Participant Information Sheets

- o Consent Forms

- o Questionnaires

❖ **Proxy-report**

- o Participant Information Sheets

- o Consent Forms

- o Questionnaires

Re: Support services for Young Adults aged 16 to 25 years

A new project has commenced at the Royal Hospital for Women and Prince of Wales Hospital in conjunction with Sydney Children's Hospital and the University of Technology, Sydney (UTS).

The project focuses on a frequently overlooked age group who often feel caught between the paediatric and adult healthcare groups. We are looking at how we might be able to reduce the impact of illness on young adults and their families, and whether or what type of support services need to be developed.

One of the main aims of this study is to identify the most desirable mix of support services from both the patient and caregiver perspective. Participation involves completing some tick-the-box style questionnaires and to write a few comments about the personal impact of illness and treatment. The questionnaires cover topics such as Quality of life, and satisfaction with services. An exciting aspect of this project is that, in asking about which types of support services young adults and their families prefer, we will be using a questionnaire method new to health sciences, but commonly used in marketing, business and economics. It requires respondents to simply choose between various combinations of possible support services.

We are currently collecting data at the outpatient clinics and wards of the participating hospitals. We are also recruiting by telephone.

Jane Ewing from the UTS will call you in the next few days to discuss the project with you. If you don't hear from us (maybe your phone number has changed) and would like to take part, please feel free to call Jane on ▬▬▬▬▬ or 9514 2240.

We hope that all participants enjoy contributing to this important research, and we look forward to meeting many of you in the near future.

Thank you

SUBJECT INFORMATION STATEMENT AND CONSENT FORM
Support Services for Young People with
Cancer or a Blood Disorder

You are invited to take part in a study of support services for young people (16-25 years old) living with a diagnosis of cancer or a blood disorder. We hope to learn more about how to reduce the impact of illness on young people and their families, and whether you think there are additional services that should be developed. Throughout this project the terms "young people" and "young adult" will be used to mean 16 to 25 year old people.

This study forms Jane's Master of Mathematical Science degree. You were selected as a possible participant in this study because you are aged between 16 and 25; were diagnosed with cancer or a blood disorder; and are on the database of either the Centre for Children's Cancer and Blood Disorders (CCCBD) at Sydney Children's Hospital, or the Prince of Wales Hospital or the Royal Hospital for Women.

Participation in this study is entirely voluntary. You are not obliged to participate, and if you do participate, you can withdraw at any time. Whatever your decision, it will in no way affect your medical care or the support services you use.

If you decide to participate, the researcher will describe what is required, and you will be asked to complete a **Consent Form** and **questionnaires** about**:**
1. Your **quality of life (PedsQL),** (which takes about 4 minutes to complete);
2. Your **satisfaction** as a patient (which takes about 5 minutes to complete);
3. Your **preferences** for the different types of support service. (This may take about 20 minutes to complete);
4. A **symptom assessment scale** (which takes about 5 minutes to complete); and
5. An opportunity to express the **impact** your illness has had on your life.

The Preferences Survey is a different kind of questionnaire. It consists of choosing between each of 16 pairs of realistic scenarios that describe different combinations of support service. Each pair consists of a different mix of different types of support service, which you may or may not prefer, to your current support services. An example of how to complete this survey is enclosed.

While it is intended that this study should be of great value in helping with the planning of future support services and their co-ordination, we cannot and do not hold that you will gain any benefit by participating in this study.

Since one of the main aims of this study is to help formulate a desirable mix of support services, which we hope can be of help to young adults and their families, this study gathers and compares the views of the young adult with those of their nominated parent,

guardian, caregiver or partner/spouse. For ease of terminology however, we will use the word 'parent' to describe this person. Therefore, we need you to nominate one 'parent' to participate by completing a similar Consent Form and questionnaires (as above).

There are no right or wrong answers. Please feel free to ask for help from a family member, friend, or Jane Ewing (9514 2240) if you should need help in completing the surveys. However, it is essential that you do not discuss your answers until completed because it is **your own view** on the different issues that is important.

All aspects of the study will be strictly confidential and only the researcher (Jane), her Supervisor (Dr Narelle Smith 9514 2239) and Co-supervisor (Dr Madeleine King 9514 4746) will have access to the names of individuals participating and the information you provide. Individual participants will not be identifiable in any reports or publications emanating from this study.

Whether you take part in this study or not, it will not make any difference to the medical care you receive at the hospital. If you decide to take part in the study, you can still withdraw at any time and this will not make any difference to your medical care either. If you have any questions about the research at any time or would like to know more, Jane (telephone 9514 2240) will be happy to answer them.

Should participation in this study raise questions about your current use of support services, you are welcome to discuss your needs with your usual Social Worker from the Social Work Department. Please phone 9382 1111 (the main hospital number) and ask the telephonist to 'page' your social worker, called…………………...

The data from this study will be stored in a locked room at the Faculty of Science at UTS, on PC and files held in a locked filing cabinet for 5 years. Disposal will be by shredding or erasure via magnetic corruption of relevant computer files. Once the data has been encoded, the file linking names with study ID's will be stored separately from the data. The data will be identified by study ID code only.

Please read the questions carefully before responding. Once you have completed the (pink) **Consent Form** and (gold) **questionnaire booklet**, please return them in the envelope provided.

This **Subject Information Statement** and the attached copy of the **Consent Form** and **Revocation of Consent** are for you to keep. The latter is for use should you wish to withdraw from the study, later on.

Thank you for your time and help in making this study possible.

Complaints:

Sydney Children's Hospital and Prince of Wales Hospital: Complaints may be directed to the Research Ethics Secretariat, South Eastern Sydney Area Health Service – Eastern Section, Prince of Wales Hospital, RANDWICK NSW 2031 AUSTRALIA (Phone 9382 3587, fax 93822813, email brehenyk@sesahs.nsw.gov.au).

This study has been approved by the **University of Technology, Sydney**, Human Research Ethics Committee. If you have any complaints or reservations about any aspect of your participation in this research, which you cannot resolve with the researcher, you may contact the Ethics Committee through the Research Ethics Officer, Ms Susanna Davis (ph: 02 - 9514 1279, Susanna.Davis@uts.edu.au). Any complaint you make will be treated in confidence and investigated fully and you will be informed of the outcome.

CONSENT FORM
Support Services for Young People with Cancer or a Blood Disorder

You are making a decision to voluntarily participate in this research project. Your signature indicates that you have read and understood the information statement provided, have been verbally informed about the study, have had a chance to ask questions, and consent to completing the questionnaires.

_____   _____
Signature of subject   Signature(s) of investigator(s)

_____   _____
Please PRINT name   Please PRINT name

_____   _____
Date   Date

_____
Telephone/Mobile

Please print your nominated parent's name:

_____
         First Name   Last Name

REVOCATION OF CONSENT

I hereby wish to WITHDRAW my consent to participate in the research proposal described above and understand that such withdrawal WILL NOT make any difference to my medical care or my relationship with the Hospital or my medical attendants.

_____   _____
Signature   Date

_____   _____
Please PRINT name   [Date Received – Office use only]

If you no longer wish to take part in this research, **please complete and send this form to:**
Dr Narelle Smith, Co- Investigator, Department of Mathematical Sciences, Faculty of Science, University of Technology, Sydney (UTS) PO Box 123, Broadway 2007, Sydney, NSW.

[PLEASE KEEP THIS COPY OF THE FORM FOR YOUR RECORDS]

Patient Code:_____

REVOCATION OF CONSENT

(Use this form if you no longer wish to take part in this study)

Support Services for Young People with Cancer or a Blood Disorder

I hereby wish to WITHDRAW my consent to participate in the research proposal
described above and understand that such withdrawal WILL NOT make any difference
to my medical care or my relationship with the Hospital or my medical attendants.

_____   _____
Signature   Date

_____   _____
Please PRINT name   [Date Received – Office use only]

If you <u>no longer wish to take part</u> in this research, **please complete and
send this form to:**

Dr Narelle Smith, Co-Investigator,
Department of Mathematical Sciences,
Faculty of Science,
University of Technology,
Sydney (UTS)
PO Box 123 Broadway 2007,
Sydney, NSW.

UTS in conjunction with South Eastern Sydney Area Health Service - Eastern Section, CCCBD, Sydney Children's & Prince of Wales Hospitals, and the Royal Hospital for Women

CONSENT FORM   Patient Code:_____

Support Services for Young People with Cancer or a Blood Disorder

You are making a decision to voluntarily participate in this research project. Your signature indicates that you have read and understood the information statement provided, have been verbally informed about the study, have had a chance to ask questions, and consent to completing the questionnaires.

_____   _____
Signature of subject   Signature(s) of investigator(s)

_____   _____
Please PRINT name   Please PRINT name

_____   _____
Date   Date

_____
Telephone/mobile

  Please print your nominated parent's name:

_____
        First Name   Last Name

[PLEASE SEND OR GIVE THIS FORM BACK TO JANE]

Date:

Date Returned:

Support Services for Adolescents and Young Adults with Cancer or a Blood Disorder

Questionnaires for Adolescents and Young Adults

UTS and CHERE in conjunction with the South East Sydney Area Health Service (Eastern Section), the Centre for Children's Cancer and Blood Disorders, Sydney Children's Hospital, the Prince of Wales Hospital and the Royal Hospital for Women.

How to complete this survey

Thank you for taking time to complete this important survey. The instructions for filling it out are provided with each questionnaire. Please answer every question. If you are unsure about how to answer a question, mark the response for the closest answer to how you feel, or the answer that is the best you can remember.

Please do not discuss with others the answers you should give because it is <u>what you think</u> that is important to us. However, once you have completed the booklet please feel free to discuss your answers with others if you wish, but please leave your answers unaltered. Thank you.

The questionnaires ask you to think about different time periods: the last month, the last week, your cumulative experience, and your current choices. Please check the time period carefully for each questionnaire.

Please remember that participation is voluntary and all information is confidential to the researcher and her supervisors at UTS.

To fill in the questions either mark a box or write your answer in the space provided.

If you need help to answer any questions, please phone
Jane Ewing on the number
9514 2240

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PedsQL™

Pediatric Quality of Life Inventory

Version 4.0

YOUNG PERSON'S REPORT (ages **16-24**)

DIRECTIONS

On the following page is a list of things that might be a problem for you.
Please tell us **how much of a problem** each one has been for you
during the **past ONE month** by circling:

> **0** if it is **never** a problem
> **1** if it is **almost never** a problem
> **2** if it is **sometimes** a problem
> **3** if it is **often** a problem
> **4** if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the past **ONE month**, how much of a **problem** has this been for you …*

About My Health and Activities *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than one block	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activity or exercise	0	1	2	3	4
4. It is hard for me to lift something heavy	0	1	2	3	4
5. It is hard for me to take a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I hurt or ache	0	1	2	3	4
8. I have low energy	0	1	2	3	4

About My Feelings *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad or blue	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

How I Get Along with Others *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting along with other young people	0	1	2	3	4
2. Other young people do not want to be my friend	0	1	2	3	4
3. Other young people tease me	0	1	2	3	4
4. I cannot do things that other young people my age can do	0	1	2	3	4
5. It is hard to keep up with my peers	0	1	2	3	4

About Study/work *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class/at work	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my study/work duties	0	1	2	3	4
4. I miss study/work because of not feeling well	0	1	2	3	4
5. I miss study/work to go to the doctor or hospital	0	1	2	3	4

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PedsQL ™

Cancer Module

Version 3.0

YOUNG PERSON'S REPORT (ages **16-24**)

DIRECTIONS

Young people with cancer sometimes have special problems. Please tell us **how much of a problem** each one has been for you during the **past one month** by circling:

        0 if it is **never** a problem
        1 if it is **almost never** a problem
        2 if it is **sometimes** a problem
        3 if it is **often** a problem
        4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

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*In the past **one month,** how much of a **problem** has this been for you …*

Pain and Hurt (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
I ache or hurt in my joints and/or muscles	0	1	2	3	4
I hurt a lot	0	1	2	3	4

Nausea (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
I become sick to my stomach when I have medical treatments	0	1	2	3	4
Food does not taste very good to me	0	1	2	3	4
I become sick to my stomach when I think about medical treatments	0	1	2	3	4
I feel too sick to my stomach to eat	0	1	2	3	4
Some foods and smells make me sick to my stomach	0	1	2	3	4

Procedural Anxiety (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
Needle sticks (i.e. injections, blood tests, IV's) hurt me	0	1	2	3	4
I get scared when I have to have blood tests	0	1	2	3	4
I get scared about having needle sticks (i.e. injections, blood tests, IV's)	0	1	2	3	4

Treatment Anxiety (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
I get scared when I am waiting to see the doctor	0	1	2	3	4
I get scared when I have to go to the doctor	0	1	2	3	4
I get scared when I have to go to the hospital	0	1	2	3	4

Worry (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
I worry about side effects from medical treatments	0	1	2	3	4
I worry about whether or not my medical treatments are working	0	1	2	3	4
I worry that my cancer will come back or relapse	0	1	2	3	4

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*In the past **one month,** how much of a **problem** has this been for you …*

Cognitive Problems (problems with…)	Never	Almost Never	Some-times	Often	Almost Always
It is hard for me to figure out what to do when something bothers me	0	1	2	3	4
I have trouble solving math problems	0	1	2	3	4
I have trouble writing study papers or reports	0	1	2	3	4
It is hard for me to pay attention to things	0	1	2	3	4
It is hard for me to remember what I read	0	1	2	3	4

Perceived Physical Appearance *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
I feel I am not good looking	0	1	2	3	4
I don't like other people to see my scars	0	1	2	3	4
I am embarrassed when others see my body	0	1	2	3	4

Communication *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
It is hard for me to tell the doctors and nurses how I feel	0	1	2	3	4
It is hard for me to ask the doctors and nurses questions	0	1	2	3	4
It is hard for me to explain my illness to other people	0	1	2	3	4

Patient Satisfaction Survey:

For Adolescents and Young Adults

Please let us know what <u>you</u> think.

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Patient Satisfaction Survey:
for Adolescents & Young Adults

Please rate your level of satisfaction with each of the following issues by ticking one box in each row:

Part 1	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
Scale 1: General Satisfaction					
1. The overall care you are receiving.	☐	☐	☐	☐	☐
2. How friendly and helpful the staff is.	☐	☐	☐	☐	☐
3. The way you are treated at the hospital.	☐	☐	☐	☐	☐
Scale 2: Information					
4. How much information was provided to you about your diagnosis.	☐	☐	☐	☐	☐
5. How much information was provided to you about the treatment and course of your disease.	☐	☐	☐	☐	☐
6. How much information was given to you about the side effects of your treatment.	☐	☐	☐	☐	☐
7. How soon information was given to you about your test results.	☐	☐	☐	☐	☐
8. How often you are updated about your disease and health.	☐	☐	☐	☐	☐
Scale 3: Inclusion of Family					
9. The sensitivity shown to your family during your treatment.	☐	☐	☐	☐	☐
10. The willingness to answer questions that you and your family may have.	☐	☐	☐	☐	☐
11. The effort to include your family in discussion of your care and other information about your disease.	☐	☐	☐	☐	☐
12. How much time the staff gave you to ask any questions you may have had about your disease and treatment.	☐	☐	☐	☐	☐

Scale 4: Communication

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
13. How well the staff explained your disease and treatment to you in a way that you could understand.	☐	☐	☐	☐	☐
14. The time taken to explain your disease and treatment to you in a way you could understand.	☐	☐	☐	☐	☐
15. How well the staff listens to you and your concerns.	☐	☐	☐	☐	☐
16. The preparation provided for your family about what to expect during tests and procedures.	☐	☐	☐	☐	☐
17. The preparation provided for you about what to expect during tests and procedures.	☐	☐	☐	☐	☐

Scale 5: Technical skills

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
18. How well the staff responds to your needs.	☐	☐	☐	☐	☐
19. Efforts to keep you comfortable and as pain-free as possible.	☐	☐	☐	☐	☐
20. How quickly the staff responds to your nausea.	☐	☐	☐	☐	☐
21.How much time the staff took to help you with going back home.	☐	☐	☐	☐	☐

Scale 6: Emotional Needs

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
22. The amount of time given to you to talk about your feelings, and ask any questions you may have.	☐	☐	☐	☐	☐
23. The amount of time spent helping you with going back to your studies or work.	☐	☐	☐	☐	☐
24. The amount of time spent attending to your emotional needs.	☐	☐	☐	☐	☐
25. The amount of time spent attending to your family's or support person's emotional needs.	☐	☐	☐	☐	☐

Part 2

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied

Scale 1A: General Satisfaction

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
1. Staff efficiency in referring you to the most suitable support services available.	☐	☐	☐	☐	☐
2. How staff relate to you.	☐	☐	☐	☐	☐
3. The ward was suitable for people my age.	☐	☐	☐	☐	☐

Scale 2A: Information

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
4. Information given to you about therapies such as dietary, herbal, massage, relaxation to complement your main treatment.	☐	☐	☐	☐	☐
5. Information given to you about accessing services for emotional support.	☐	☐	☐	☐	☐
6. Information given to you about accessing financial support services.	☐	☐	☐	☐	☐

Scale 3A: Inclusion of family

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
7. The level of cultural sensitivity shown to you and your family.	☐	☐	☐	☐	☐
8. The involvement of your family is at the level you want.	☐	☐	☐	☐	☐

Scale 4A:Communication

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
9. How well the support staff explained the support services available to you and your family.	☐	☐	☐	☐	☐

Scale 5A: Technical skills

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
10. How efficiently staff respond to unplanned admissions caused by your illness or the prescribed treatment.	☐	☐	☐	☐	☐

Scale 6A: Emotional needs

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
11. The amount of staff effort in linking you with people of similar age who face similar health problems.	☐	☐	☐	☐	☐

Scale 7A: Financial Needs

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
12. The amount of attention initiated by staff in helping you address any financial needs caused by your illness.	☐	☐	☐	☐	☐

Scale 8A: Current state of well-being

	Very Poorly	Poor	Average	Good	Very Good
13. Please circle one of the faces to show how you are feeling in yourself today.	☹	☹	😐	☺	😄

Scale 9A: Comments

Preferences for Support Services

The views of Young Adults

Preferences for Support Services

This questionnaire is about support services (in addition to your medical and nursing care). It describes some hypothetical (or imaginary) support services.

We want you to imagine which support services would help you, and which mix you would choose (columns "**Mix A**" or "**Mix B**" – see example). We have included some definitions or meaning of terms on the next page that you might find useful.

Remember that your answers will not alter your current use of support services, and that it is your views that are important.

Example:

Support Services:	**Mix A:**	**Mix B:**
Cultural/Ethnic support	No	Yes
Spiritual support	**No**	**No**
A person who can advise you about available financial support and how to access it	**No**	**Yes**
Support to help you return to your studies and/or work	**Yes**	**No**
Emotional support **for you**	**Yes, provided by a counsellor and peer support**	**Yes, provided by a counsellor only**
Emotional support for **your family**	**Yes, provided by a peer support only**	**No**

Which mix of support services would you choose?
(Please mark one box only)

Mix A ☐

Mix B ☒

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DEFINITIONS and TERMS

1. Cultural/Ethnic support
Whether staff discuss options with you regarding your cultural needs and those of your family, in providing suitable support from appropriate cultural and ethnic groups that might be helpful.

2. Spiritual support
Whether staff discuss options with you, regarding your spiritual needs and those of your family, in providing suitable support from appropriate spiritual or religious groups that might be helpful.

3. Financial support
Whether the hypothetical support service includes a person who can advise you of your eligibility for financial assistance (either through Government or private funding). Would you prefer a service where you were informed about the available financial support, and how to access it?

4. Rehabilitation
Whether the staff discussed with you and provided appropriate referral as agreed for suitable rehabilitative support, to achieve a smooth return to your study or work. For example, it could be you are referred for assistance regarding mobility issues or for other equipment; or perhaps in helping your peers or work colleagues to understand your medical problem (if you wish); or providing referral for specialist subject teacher(s) to assist you in catching up with your study; or any other help to make your transition back to work or study that much smoother.

5. Emotional support for you
Whether emotional support is offered to you, by way of a counsellor/psychologist only, peer support only, a combination of a counsellor/psychologist and peer support, or none of these. (In this context a peer is considered as a person with similar experience and is a non-professional, whereas a counsellor/psychologist is a professional adviser)

6. Emotional support for your family
Whether emotional support is offered to your family, by way of a counsellor/psychologist only, peer support only, a counsellor/psychologist and peer support, or none of these.

- **Cultural** means, the ideas, customs and art of a particular society or group.
- **Ethnic** means, "of or relating to a human group with racial, religious, and linguistic characteristics in common"
- **Spiritual** means, "relating to a person's beliefs as opposed to his or her physical or material needs" or "relating to religious beliefs"
- **Religious** means 'of religion', which is "belief in or worship of a supernatural power or powers considered to be divine or to have control over human destiny", or "any formal expression of such belief: the Christian religion"
- **Peer** means, "a person of equal social standing, rank, age, etc"
- **Counsellor** means, "an adviser, a person giving professional guidance on personal problems"

Scenario 1

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	No	Yes, provided by a counsellor
Emotional support for your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 2

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	No	Yes, provided by a counsellor
Emotional support for your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 3

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 4

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 5

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	No	Yes, provided by a counsellor
Emotional support for your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 6

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	No	Yes, provided by a counsellor
Emotional support for your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A 　　　☐

Mix B 　　　☐

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A 　　　☐

Mix B 　　　☐

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a counsellor and a peer	No
Emotional support for your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a counsellor and a peer	No
Emotional support for your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 11

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ⬜.

Mix B ⬜.

Scenario 12

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ⬜.

Mix B ⬜.

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a counsellor and a peer	No
Emotional support for your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A   ⬜.

Mix B   ⬜.

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	No	Yes
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a counsellor and a peer	No
Emotional support for your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A   ⬜.

Mix B   ⬜.

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	No	Yes
Emotional support for you	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Support Services:	Mix A:	Mix B:
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you about available financial support and how to access it.	Yes	No
Support to help you return to your studies and/or work	Yes	No
Emotional support for you	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

You are nearly at the end
of the survey!

Symptom Assessment Scale

Section 1:

We have listed 22 symptoms below. Please read each one carefully. If you have had the symptom during the past week, circle YES. If YES, let us know how OFTEN you had it, how SEVERE it was usually and how much it BOTHERED OR DISTRESSED by circling the appropriate answer. If you DID NOT HAVE the symptom circle NO.

During the past WEEK did you have any:

1. Difficulty concentrating or paying attention? YES / NO

If YES: How **often** did you have it? ……………………
How **severe** was it usually? ……………………
How much did it **bother** or distress you? …

Almost never	Sometimes	A lot	Almost always
Slight	Moderate	Severe	Very severe
Not at all	A little bit	Somewhat	Quite a bit	Very much

2. Pain?   YES / NO

If YES: How **often** did you have it? ……………………
How **severe** was it usually? ……………………
How much did it **bother** or distress you? ……

Almost never	Sometimes	A lot	Almost always
Slight	Moderate	Severe	Very severe
Not at all	A little bit	Somewhat	Quite a bit	Very much

3. Lack of energy?   YES / NO

If YES: How **often** did you have it? ……………………
How **severe** was it usually? ……………………
How much did it **bother** or distress you? ……

Almost never	Sometimes	A lot	Almost always
Slight	Moderate	Severe	Very severe
Not at all	A little bit	Somewhat	Quite a bit	Very much

4. Cough?   YES / NO

If YES: How **often** did you have it? ……………………
How **severe** was it usually? ……………………
How much did it **bother** or distress you? ……

Almost never	Sometimes	A lot	Almost always
Slight	Moderate	Severe	Very severe
Not at all	A little bit	Somewhat	Quite a bit	Very much

5. Feeling of being nervous?   YES / NO

If YES: How **often** did you have it? ……………………
How **severe** was it usually? ……………………
How much did it **bother** or distress you? ……

Almost never	Sometimes	A lot	Almost always
Slight	Moderate	Severe	Very severe
Not at all	A little bit	Somewhat	Quite a bit	Very much

6. Dry mouth?   YES / NO

If YES:		Almost never	Sometimes	A lot	Almost always	
How **often** did you have it? ……………						
How **severe** was it usually? ……………						
How much did it **bother** or distress you? ……						

For each line the scales read:
- How **often**: Almost never — Sometimes — A lot — Almost always
- How **severe**: Slight — Moderate — Severe — Very severe
- How much did it **bother**: Not at all — A little bit — Somewhat — Quite a bit — Very much

7. Nausea or feeling like you could vomit?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

8. A feeling of being drowsy?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

9. Numbness/tingling or pins & needles feeling in hands or feet?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

10. Difficulty sleeping?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

11. Problems with urination or 'peeing'?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

12. Vomiting or throwing?   YES / NO
If YES: How **often** did you have it? ……………   Almost never   Sometimes   A lot   Almost always
How **severe** was it usually? ……………   Slight   Moderate   Severe   Very severe
How much did it **bother** or distress you? ……   Not at all   A little bit   Somewhat   Quite a bit   Very much

13. Shortness or Breath? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?...... Not at all / A little bit / Somewhat / Quite a bit / Very much

14. Diarrhea or loose bowel movement? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?..... Not at all / A little bit / Somewhat / Quite a bit / Very much

15. Feelings of sadness? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?..... Not at all / A little bit / Somewhat / Quite a bit / Very much

16. Sweats? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?...... Not at all / A little bit / Somewhat / Quite a bit / Very much

17. Worrying? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?..... Not at all / A little bit / Somewhat / Quite a bit / Very much

18. Itching? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?.... Not at all / A little bit / Somewhat / Quite a bit / Very much

19. Lack of appetite or not wanting to eat? **YES / NO**

If YES:
How **often** did you have it? .... Almost never / Sometimes / A lot / Almost always
How **severe** was it usually? .... Slight / Moderate / Severe / Very severe
How much did it **bother** or distress you?.. Not at all / A little bit / Somewhat / Quite a bit / Very much

20. Dizziness?

If YES: **YES / NO**

How **often** did you have it?............... Almost never   Sometimes   A lot   Almost always

How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

21. Difficulty swallowing?   **YES / NO**

If YES:

How **often** did you have it?............... Almost never   Sometimes   A lot   Almost always

How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

22. Feelings of being irritable?   **YES / NO**

If YES:

How **often** did you have it? .... Almost never   Sometimes   A lot   Almost always

How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

Section 2:

We have listed 8 symptoms below. Please read each one carefully. If you have had the symptom during this past week, let us know how how SEVERE it was usually and how much it BOTHERED or DISTRESSED you by circling the appropriate answer. If YOU DID NOT HAVE the symptom circle NO.

During the last week did you have any:

1. Mouth sores?   **YES / NO**

If YES: How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

2. Change in the way food tastes? YES / NO

If YES: How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

3. Weight loss?   **YES / NO**

If YES: How **severe** was it usually?............... Slight   Moderate   Severe   Very severe

How much did it **bother** or distress you?............ Not at all   A little bit   Somewhat   Quite a bit   Very much

4. Hair loss? YES / NO

If YES: How **severe** was it usually?…………………

Not at all	Slight	Moderate	Severe	Very severe

How much did it **bother** or distress you?…………

Not at all	A little bit	Somewhat	Quite a bit	Very much

5. Constipation or uncomfortable because bowel movements are less often? YES / NO

If YES: How **severe** was it usually?…………………

Not at all	Slight	Moderate	Severe	Very severe

How much did it **bother** or distress you?…………

Not at all	A little bit	Somewhat	Quite a bit	Very much

6. Swelling of arms or legs? YES / NO

If YES: How **severe** was it usually?…………………

Not at all	Slight	Moderate	Severe	Very severe

How much did it **bother** or distress you?…………

Not at all	A little bit	Somewhat	Quite a bit	Very much

7. "I don't look like myself" YES / NO

If YES: How **severe** was it usually?…………………

Not at all	Slight	Moderate	Severe	Very severe

How much did it **bother** or distress you?…………

Not at all	A little bit	Somewhat	Quite a bit	Very much

8. Changes in skin? YES / NO

If YES: How **severe** was it usually?…………………

Not at all	Slight	Moderate	Severe	Very severe

How much did it **bother** or distress you?…………

Not at all	A little bit	Somewhat	Quite a bit	Very much

IMPACT SCALE – Please comment

To what extent and in what way has your illness and treatment:

(a) disrupted your ability to:

      (i) Pursue your study (or goals)?

      (ii) Earn income?

(b) enhanced your ability to:

      (i) Pursue your study (or goals)?

      (ii) Earn income?

(c) interfered with:

      (i) Family relationships?

      (ii) Personal relationships?

(d) strengthened:

      (i) Family relationships?

      (ii) Personal relationships?

---

*In the past **12 months**, have you, the young adult patient had…*

Any **OVERNIGHT VISITS** to the hospital?	Any **EMERGENCY ROOM/URGENT CARE** visits?
☐ NO ☐ YES	☐ NO ☐ YES
IF YES, … How many times? ⎣_⎦	**IF YES,** … How many times? ⎣_⎦
What was wrong? [＿＿＿＿＿]	What was wrong? [＿＿＿＿＿]

*In the past **30 days**…*

How many days did you miss study/work due to physical or mental health? ⎣_⎦

How many days were you sick in bed or too ill to take part in fun activities? ⎣_⎦

How many days did you need someone to care for you due to physical or mental health? ⎣_⎦

About your condition and treatment, please tick as many as apply to you

My condition or disease	Treatments over past month	Treatments I am expecting over next month:
☐ ongoing	☐ chemotherapy	☐ chemotherapy
☐ in remission	☐ radiation ☐ chelation	☐ radiation ☐ chelation
☐ in relapse (recurred)	☐ surgery ☐ transfusion	☐ surgery ☐ transfusion
☐ cured	☐ transplant	☐ transplant
	☐ no treatment	☐ no treatment

Illness:　　☐ Cancer　　　　☐ Blood Disorder　　　Other _____

INFORMATION ABOUT YOU, THE YOUNG ADULT PATIENT

Gender:　　　☐ male　　　　☐ female

Ethnic Group or Race:
　　　　☐ Aboriginal/Torres Strait　　☐ Maori/Cook Islands　　☐ Pacific Islander
　　　　☐ Asian　　　　☐ European descent
☐ Other_____

Post Code:　☐☐☐☐

Marital Status:　☐ Single　　　　☐ Separated　　　　☐ Living with someone
　　　　　　　　☐ Married　　　　☐ Divorced　　　　☐ Widowed

Highest Level　☐ 6th grade or less　　　　☐ High School Certificate (HSC)
of Education:　☐ 7th-9th grade or less　　　☐ Some tertiary certification course
　　　　　　　☐ 9th-12th grade or less　　　☐ Graduate or Professional Degree

Are you currently:　　☐ *studying?*　　　☐ *working?*　　　☐ *working and studying?*

What is your relationship to your nominated 'parent' (please tick one box)?
☐ Daughter, ☐ Step-daughter, ☐ Foster-daughter ☐ Grand-daughter ☐ Niece ☐ Spouse ☐ Partner
☐ Son, 　　☐ Step-son, 　　☐ Foster-son 　　☐ Grand-son 　　☐ Nephew ☐ Other………………

Thank you for taking the time to fill in this survey.

Please check that you have answered every question before
you hand it back to Jane or post it to her. If posting please use the pre-addressed
reply-paid envelope provided.

We appreciate your help with this important research.
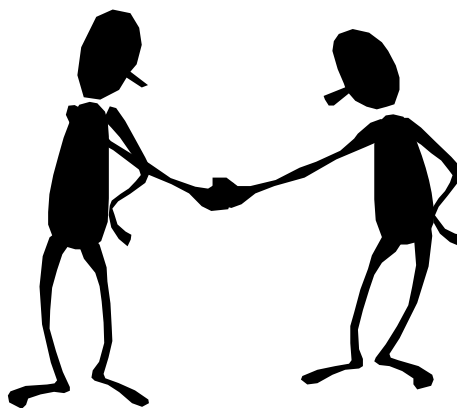
Postal Address: Jane Ewing, Dept of Mathematical Sciences, Faculty of Science,
University of Technology, Sydney (UTS)
PO Box 123 Broadway, Ultimo, Sydney 2007, NSW

SUBJECT INFORMATION STATEMENT AND CONSENT FORM

Nominated Parent

Support Services for Young People with Cancer or a Blood Disorder

As the nominated parent/guardian of your child you are invited to take part in a study of support services about young people living with a diagnosis of cancer or a blood disorder. We hope to learn more about how to reduce the impact of illness on young people and their families, and whether you think there are additional services that should be developed. Throughout this project the terms "young people" and "young adult" will be used to mean 16 to 25 year old people.

Since one of the main aims of this study is to help formulate a desirable mix of support services, which we hope can be of help to young adults and their families, this study gathers and compares the views of the young adult patient with those of their nominated parent or guardian or partner/spouse or caregiver. For ease of terminology, however, we will use the word "parent" to describe this person.

This study forms Jane Ewing's Master of Mathematical Science degree. Your child was selected as a possible participant in this study because he/she: is aged 16 to 25 years; was diagnosed with cancer or a blood disorder; and is on the database of the Centre for Children's Cancer and Blood Disorders (CCCBD) at Sydney Children's Hospital, or Prince of Wales Hospital or the Royal Hospital for Women.

Participation in this study is entirely voluntary. You are not obliged to participate, and if you do participate, you can withdraw at any time. Whatever your decision, it will in no way affect the medical care of your child or the support services used.

If you decide to participate, the researcher will describe what is required, and you will be asked to complete a slightly different version of the **Consent Form** and **questionnaires** to those of your child. The questionnaires are about**:**
   1. Your perception of the **quality of life (PedsQL) of your** child**,** (which takes about 4 minutes to complete);
   2. Your **satisfaction with services** as a parent (This takes about 5 minutes to complete);
   3. Your **preferences** for the different types of support service, which you think would be helpful. (This may take about 20 minutes to complete); and
   4. An opportunity to express the **impact** your child's illness has had.

The Preferences Survey is a different kind of questionnaire. It consists of choosing between each of 16 pairs of realistic scenarios that describe different combinations of support service. Each pair consists of a different mix of different types of support service, which

you may or may not prefer, to your current support services. An example of how to complete this survey is enclosed.

While it is intended that this study should be of great value in helping with the planning of future support services and their co-ordination, we cannot and do not hold that you or your child will gain any benefit by participating in this study.

There are no right or wrong answers. Please feel free to ask for help from a family member, friend, or Jane Ewing (9514 2240) if you should need help in completing the surveys. However, it is essential that you do not discuss your answers until completed because it is **your own view** on the different issues that is important.

All aspects of the study will be strictly confidential and only the researcher (Jane), her Supervisor (Dr Narelle Smith 9514 2239) and Co-supervisor (Dr Madeleine King 9514 4746) will have access to the names of individuals participating and the information you provide. Individual participants will not be identifiable in any reports or publications emanating from this study.

Whether you take part in this study or not, it will not make any difference to the medical care your child receives at the hospital. If you decide to take part in the study, you can still withdraw at any time and this will not make any difference to the medical care either. If you have any questions about the research at any time or would like to know more, Jane Ewing (telephone 9514 2240) will be happy to answer them.

Should participation in this study raise questions about your current use of support services, you are welcome to discuss your family's needs with your child's usual Social Worker from the Hospital Social Work Department. Please phone 9382 1111 (the main hospital number) and ask the telephonist to 'page' your social worker, who is called …………………..........

The data from this study will be stored in a locked room at the Faculty of Science at UTS, on PC and files held in a locked filing cabinet for 5 years. Disposal will be by shredding or erasure via magnetic corruption of relevant computer files. Once the data has been encoded, the file linking names with study ID's will be stored separately from the data. The data will be identified by study ID code only.

Please read the questions carefully before responding. Once you have completed the (pink) **Consent Form,** and (pale green) **questionnaire booklet**, please return them in the envelope provided. The **Subject Information Statement** and attached copy of the **Consent and Revocation of Consent Form** is for you to keep.

Thank you for your time and help in making this study possible.

Complaints:

CONSENT FORM

Support Services for Young People with
Cancer or a Blood Disorder

You are making a decision to voluntarily participate in this research project. Your signature indicates that you have read and understood the information statement provided, have been verbally informed about the study, have had a chance to ask questions, and consent to completing the questionnaires.

_____   _____
Signature of parent   Signature(s) of investigator(s)

_____   _____
Please PRINT name   Please PRINT name

_____   _____
Date   Date

_____
Telephone/Mobile

_____
Please print your Child's name: First Name   Last Name

REVOCATION OF CONSENT

I hereby wish to WITHDRAW my consent to participate in the research proposal described above and understand that such withdrawal WILL NOT make any difference to my medical care or my relationship with the Hospital or my medical attendants.

_____   _____
Signature   Date

_____   _____
Please PRINT name   [Date Received – Office use only]

If you no longer wish to take part in this research, please complete and send the Revocation of Consent form to:
>   Dr Narelle Smith, Co-Investigator,
>   Department of Mathematical Sciences,
>   Faculty of Science,
>   University of Technology, Sydney (UTS)
>   PO Box 123, Broadway 2007, Sydney, NSW.

[PLEASE KEEP THIS COPY OF THE FORM FOR YOUR RECORDS]

UTS in conjunction with South Eastern Sydney Area Health Service - Eastern Section, CCCBD, Sydney Children's & Prince of Wales Hospitals, and the Royal Hospital for Women

Parent Proxy Code:_____

REVOCATION OF CONSENT
(Use this form if you no longer wish to take part in this study)

Support Services for Young People with Cancer or a Blood Disorder

I hereby wish to WITHDRAW my consent to participate in the research proposal described above and understand that such withdrawal WILL NOT make any difference to my medical care or my relationship with the Hospital or my medical attendants.

_____   _____
Signature   Date

_____   _____
Please PRINT name   [Date Received – Office use only]

If you no longer wish to take part in this research, please complete and send this form to:

        Dr Narelle Smith, Co-Investigator,
        Department of Mathematical Sciences,
        Faculty of Science,
        University of Technology,
        Sydney (UTS)
        PO Box 123 Broadway 2007,
        Sydney, NSW.

UTS in conjunction with South Eastern Sydney Area Health Service - Eastern Section, CCCBD, Sydney Children's & Prince of Wales Hospitals, and the Royal Hospital for Women

Parent Proxy Code:_____

SUBJECT CONSENT FORM

Support Services for Young People with Cancer or a Blood Disorder

You are making a decision to voluntarily participate in this research project. Your signature indicates that you have read and understood the information statement provided, have been verbally informed about the study, have had a chance to ask questions, and consent to completing the questionnaires.

_____

Signature of parent   Signature(s) of investigator(s)

_____

Please PRINT name   Please PRINT name

_____

Date   Date

_____

Telephone/Mobile

Please print your Child/Charge's name:

_____

First Name   Last Name

[PLEASE SEND OR GIVE THIS FORM BACK TO JANE EWING, THE RESEARCHER]

Date: ☐☐☐☐☐☐

Date Returned: ☐☐☐☐☐☐

Support Services for Young People with Cancer or a Blood Disorder

Questionnaires for Parents/Caregivers

All the information provided in this survey is strictly confidential and only the researchers with direct involvement in the study will have access to this information.

UTS and CHERE in conjunction with the South East Sydney Area Health Service (Eastern Section), the Centre for Children's Cancer and Blood Disorders, and Sydney Children's Hospital.

How to complete this survey

Thank you for taking time to complete this important survey. The instructions for filling it out are provided with each questionnaire. Please answer every question. If you are unsure about how to answer a question, mark the response for the closest answer to how you feel, or the answer that is the best you can remember.

Please do not discuss with others the answers you should give because it is <u>what</u> <u>you think</u> that is important to us. However, once you have completed the booklet please feel free to discuss your answers with others if you wish, but please leave your answers unaltered. Thank you.

The questions ask you to think about different time periods. Some ask about the last month, some the last week, and some don't give any particular time. Please check the time period carefully for each question.

Please remember that participation is voluntary and all information is confidential to the researcher and her supervisors at UTS.

To fill in the questions either mark a box or write your answer in the space provided.

If you need help to answer any questions, please phone
Jane Ewing on the number
9514 2240

Date:_____

PedsQL ™

Pediatric Quality of Life Inventory

Version 4.0

PARENT REPORT for **Young People** (ages **16-24**)

DIRECTIONS

On the following page is a list of things that might be a problem for **your child.**
. Please tell us **how much of a problem** each one has been for **your child/charge** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

*In the past **ONE month,** how much of a **problem** has your charge had with …*

Physical Functioning *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than one block	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activity or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

Emotional Functioning *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or blue	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other young people	0	1	2	3	4
2. Other young people not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other young people	0	1	2	3	4
4. Not able to do things that other young people his or her age can do	0	1	2	3	4
5. Keeping up with other young people	0	1	2	3	4

STUDY/WORK FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class/at work	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with study/work	0	1	2	3	4
4. Missing class/work because of not feeling well	0	1	2	3	4
5. Missing class/work to go to the doctor or hospital	0	1	2	3	4

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Date:_____

PedsQL ™

Cancer Module

Version 3.0

PARENT REPORT for **Young People** (ages **16-24**)

DIRECTIONS

Young people with cancer sometimes have special problems. On the following page is a list of things that might be a problem for **your child/charge.**. Please tell us **how much of a problem** each one has been for **your child/charge** during the **past one month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.

If you do not understand a question, please ask for help.

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*In the past **month,** how much of a **problem** has your charge/child had with …*

Pain and Hurt (PROBLEMS WITH…)	Never	Almost Never	Some-times	Often	Almost Always
1. Aches in joints and/or muscles	0	1	2	3	4
2. Having a lot of pain	0	1	2	3	4

Nausea (PROBLEMS WITH…)	Never	Almost Never	Some-times	Often	Almost Always
1. Becoming nauseated during medical treatments	0	1	2	3	4
2. Food not tasting very good to him/her	0	1	2	3	4
3. Becoming nauseated while thinking about medical treatments	0	1	2	3	4
4. Feeling too nauseous to eat	0	1	2	3	4
5. Some foods and smells making him/her nauseous	0	1	2	3	4

Procedural Anxiety (PROBLEMS WITH…)	Never	Almost Never	Some-times	Often	Almost Always
1. Needle sticks (i.e. injections, blood tests, IV's) causing him/her pain	0	1	2	3	4
2. Getting anxious about having blood drawn	0	1	2	3	4
3. Getting anxious about having needle sticks (i.e. injections, blood tests, IV's)	0	1	2	3	4

Treatment Anxiety (PROBLEMS WITH…)	Never	Almost Never	Some-times	Often	Almost Always
1. Getting anxious when waiting to see the doctor	0	1	2	3	4
2. Getting anxious about going to the doctor	0	1	2	3	4
3. Getting anxious about going to the hospital	0	1	2	3	4

Worry (PROBLEMS WITH…)	Never	Almost Never	Some-times	Often	Almost Always
1. Worrying about side effects from medical treatments	0	1	2	3	4
2. Worrying about whether or not his/her medical	0	1	2	3	4
3. Worrying that the cancer will reoccur or relapse	0	1	2	3	4

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*In the past **one month,** how much of a **problem** has your charge/child had with …*

Cognitive Problems *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Difficulty figuring out what to do when something bothers him/her	0	1	2	3	4
2. Trouble solving math problems	0	1	2	3	4
3. Trouble writing study papers or reports	0	1	2	3	4
4. Difficulty paying attention to things	0	1	2	3	4
5. Difficulty remembering what he/she reads	0	1	2	3	4

Perceived Physical Appearance *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling that he/she is not good looking	0	1	2	3	4
2. Not liking other people to see his/her scars	0	1	2	3	4
3. Being embarrassed about others seeing his/her	0	1	2	3	4

Communication *(PROBLEMS WITH…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Difficulty telling the doctors and nurses how he/she feels	0	1	2	3	4
2. Difficulty asking the doctors or nurses questions	0	1	2	3	4
3. Difficulty explaining his/her illness to other people	0	1	2	3	4

Satisfaction Survey:

Parents/Guardians/Caregivers of Young People aged 16-24

In answering this questionnaire please think about your responses in terms of <u>the last two to three years</u> only.

Please rate your level of satisfaction with each of the following possible concerns or issues, by marking one box in each row nearest to what you think.

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Satisfaction Survey: Proxies of Adolescent & Young Adults aged 16-24

Please rate your level of satisfaction with each of the following issues by ticking one box in each row:

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
Scale 1: General Satisfaction					
1. The overall care your child is receiving.	☐	☐	☐	☐	☐
2. How friendly and helpful the staff is.	☐	☐	☐	☐	☐
3. The way your child is treated at the hospital.	☐	☐	☐	☐	☐
Scale 2: Information					
4. How much information was provided to you about your child's diagnosis.	☐	☐	☐	☐	☐
5. How much information was provided to you about the treatment and course of your child's disease.	☐	☐	☐	☐	☐
6. How much information was given to you about the side effects of your child's treatment.	☐	☐	☐	☐	☐
7. How soon information was given to you about your child's test results.	☐	☐	☐	☐	☐
8. How often you are updated about your child's disease and health.	☐	☐	☐	☐	☐
Scale 3: Inclusion of Family					
9. The sensitivity shown to your family during your child's treatment.	☐	☐	☐	☐	☐
10. The willingness to answer questions that you and your family may have.	☐	☐	☐	☐	☐
11. The effort to include your family in discussion of your child's care and other information about your child's disease.	☐	☐	☐	☐	☐
12. How much time the staff gave you to ask any questions you may have had about your child's disease and treatment.	☐	☐	☐	☐	☐

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied

Scale 4: Communication

13. How well the staff explained your child's disease and treatment to your child in a way that she/he could understand. ☐ ☐ ☐ ☐ ☐

14. The time taken to explain your child's disease and treatment to you in a way you could understand. ☐ ☐ ☐ ☐ ☐

15. How well the staff listens to you and your concerns. ☐ ☐ ☐ ☐ ☐

16. The preparation provided for you about what to expect during tests and procedures. ☐ ☐ ☐ ☐ ☐

17. The preparation provided for your child about what to expect during tests and procedures. ☐ ☐ ☐ ☐ ☐

Scale 5: Technical skills

18. How well the staff responds to your child's needs. ☐ ☐ ☐ ☐ ☐

19. Efforts to keep your child comfortable and as pain-free as possible. ☐ ☐ ☐ ☐ ☐

20. How quickly the staff responds to your child's nausea. ☐ ☐ ☐ ☐ ☐

21. How much time the staff took to help you with your child coming back home. ☐ ☐ ☐ ☐ ☐

Scale 6: Emotional Needs

22. The amount of time given to your child to play, talk about her/his feelings, and ask any questions she/he may have. ☐ ☐ ☐ ☐ ☐

23. The amount of time spent helping your child with going back to school/study/work. ☐ ☐ ☐ ☐ ☐

24. The amount of time spent attending to your child's emotional needs. ☐ ☐ ☐ ☐ ☐

25. The amount of time spent attending to your emotional needs. ☐ ☐ ☐ ☐ ☐

Scale 1A: General Satisfaction	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
1. Staff efficiency in referring you and your child to the appropriate support services available.	☐	☐	☐	☐	☐
2. How staff relate to your child.	☐	☐	☐	☐	☐
3. Suitability of the ward for people of my child's age.	☐	☐	☐	☐	☐

Scale 2A: Information

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
4. Information given to you about therapies such as dietary, herbal, massage, relaxation to complement the main treatment.	☐	☐	☐	☐	☐
5. Information given to you about accessing services for emotional/psychological support.	☐	☐	☐	☐	☐
6. Information given to you about accessing financial support services.	☐	☐	☐	☐	☐

Scale 3A: Inclusion of family

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
7. The level of cultural sensitivity shown to your child and your family.	☐	☐	☐	☐	☐
8. The level of involvement of your family.	☐	☐	☐	☐	☐

Scale 4A:Communication

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
9. How well the support staff explained the support services available to you and your child.	☐	☐	☐	☐	☐

Scale 5A: Technical skills

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
10. How efficiently staff respond to your child's unplanned admissions caused by his/her illness or the prescribed treatment.	☐	☐	☐	☐	☐

Scale 6A: Emotional needs

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
11. The amount of staff effort in linking your family with other families facing similar health situations.	☐	☐	☐	☐	☐

Scale 7A: Financial Needs

	very dissatisfied	dissatisfied	undecided	satisfied	very satisfied
12. The amount of attention initiated by staff in helping you address any financial needs caused by your child's illness.	☐	☐	☐	☐	☐

Scale 8A: Current state of well-being

	Very Poorly	Poor	Average	Good	Very Good
13. Please circle one of the faces to show how you are feeling in yourself today.	☹	☹	😐	☺	😀

Scale 9A: Comments

You are half way through!

Your information is important. Please keep going.

Preferences for Support Services

Proxy/Caregiver's views

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Preferences for Support Services

This questionnaire is about support services (in addition to your child/charge's medical and nursing care). It describes some hypothetical (or imaginary) support services.

We want you to imagine which support services would help you and your child/charge, and which mix you would choose (column "Mix A" or "Mix B" – see example).

We have included some definitions or meaning of terms on the next page that you might find useful.

Remember that your answers <u>will not alter</u> your child/charge's current use of support services.

Example:

Support Services:	**Mix A:**	**Mix B:**
Cultural/Ethnic support	No	Yes
Spiritual support	**No**	**No**
A person who can advise you and your child/charge about available financial support and how to access it	**No**	**Yes**
Support to help your child/charge return to his/her studies and/or work	**Yes**	**No**
Emotional support **for your child/charge**	**Yes, provided by a counsellor and peer support**	**Yes, provided by a counsellor only**
Emotional support for **you and your family**	**Yes, provided by a peer support only**	**No**

Which mix of support services would you choose?
(Please mark one box only)

	Mix A
X	**Mix B**

DEFINITIONS and TERMS

1. Cultural/Ethnic support
Whether staff discuss options with you and your child/charge regarding your cultural needs and those of your family, in providing suitable support from appropriate cultural and ethnic groups that you and your child/charge might find helpful.

2. Spiritual support
Whether staff discuss options with you and your child/charge regarding your spiritual needs and those of your child/charge and family, in providing suitable support from appropriate spiritual or religious groups that might be helpful.

3. Financial support
Whether the hypothetical support service includes a person who can advise you and your child/charge of your eligibility for financial assistance (either through Government or private funding). Would you prefer a service where you were informed about the available financial support, and how to access it?

4. Rehabilitation
Whether the staff discussed with you and your child/charge, and provided appropriate referral for rehabilitative support, to achieve a smooth return to study and/or work for your child/charge. For example, it could be you are referred for assistance regarding mobility issues or for other equipment; or perhaps in helping your child/charge's peers or work colleagues to understand his/her medical problem (if he/she wishes); or providing referral for specialist subject teacher(s) to assist him/her in catching up with his/her study; or any other help to make his/her transition back to work or study that much smoother.

5. Emotional support for your child/charge
Whether emotional support is offered to your child/charge, by way of a counsellor/psychologist only, peer support only, a combination of a counsellor/psychologist and peer support, or none of these. (In this context a peer is considered as a person with similar experience and is a non-professional, whereas a counsellor/psychologist is a professional adviser)

6. Emotional support for your family
Whether emotional support is offered to you and your family, by way of a counsellor/psychologist only, peer support only, a counsellor/psychologist and peer support, or none of these.

- **Cultural** means, the ideas, customs and art of a particular society or group.
- **Ethnic** means, "of or relating to a human group with racial, religious, and linguistic characteristics in common"
- **Spiritual** means, "relating to a person's beliefs as opposed to his or her physical or material needs" or "relating to religious beliefs"
- **Religious** means 'of religion', which is "belief in or worship of a supernatural power or powers considered to be divine or to have control over human destiny", or "any formal expression of such belief: the Christian religion"
- **Peer** means, "a person of equal social standing, rank, age, etc"
- **Counsellor** means, "an adviser, a person giving professional guidance on personal problems"

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	No	Yes, provided by a counsellor
Emotional support for you and your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A	☐
Mix B	☐

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	No	Yes, provided by a counsellor
Emotional support for you and your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A	☐
Mix B	☐

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for you and your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A   ⬚.

Mix B   ⬚.

Scenario 4

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for you and your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A   ⬚.

Mix B   ⬚.

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Scenario 5
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Support Services:	**Mix A**	**Mix B**
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	No	Yes, provided by a counsellor
Emotional support for you and your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ▢

Mix B ▢

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Scenario 6
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Support Services:	**Mix A**	**Mix B**
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	No	Yes, provided by a counsellor
Emotional support for you and your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ▢

Mix B ▢

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for you and your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Scenario 8

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a peer	Yes, provided by a counsellor and a peer
Emotional support for you and your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a counsellor and a peer	No
Emotional support for you and your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a counsellor and a peer	No
Emotional support for you and your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐

Mix B ☐

Scenario 11

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for you and your family	No	Yes, provided by a counsellor

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Scenario 12

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for you and your family	Yes, provided by a peer	Yes, provided by a counsellor and a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a counsellor and a peer	No
Emotional support for you and your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ⬚

Mix B ⬚

Scenario 14

Support Services:	Mix A	Mix B
Cultural/Ethnic support	Yes	No
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	No	Yes
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a counsellor and a peer	No
Emotional support for you and your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ⬚

Mix B ⬚

Scenario 15

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	No	Yes
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	No	Yes
Emotional support for your child/charge	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for you and your family	Yes, provided by a counsellor and a peer	No

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

Scenario 16

Support Services:	Mix A	Mix B
Cultural/Ethnic support	No	Yes
Spiritual support	Yes	No
A person who can advise you and your child/charge about available financial support and how to access it.	Yes	No
Support to help your child/charge return to his/her studies and/or work	Yes	No
Emotional support for your child/charge	Yes, provided by a counsellor	Yes, provided by a peer
Emotional support for you and your family	Yes, provided by counsellor	Yes, provided by a peer

Which mix of support services would you choose?

(Please tick one box only)

Mix A ☐.

Mix B ☐.

You are nearly at the end
of the survey!

Parent, Guardian or Caregiver

Family Information Form

The following personal information enables us to analyse the data appropriately and will be stored securely and separately from the questionnaire booklets. Please remember, individual participants will not be identifiable in any reports resulting from this study.

Thank you, please complete the last page.

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Parent -Family Information Form

As the nominated parent, please print your name:

_____

First Name   Last Name

What is your relationship to the young adult patient (please check or circle)?

☐ Mother, Step Mother, Foster Mother ☐ Grandmother ☐ Guardian ☐ Spouse ☐ Partner
☐ Father, Step Father, Foster Father ☐ Grandfather ☐ Uncle ☐ Aunt ☐ Other _____

INFORMATION ABOUT THE NOMINATED PARENT, GUARDIAN OR CAREGIVER

Gender: ☐ male   *Post Code:* |___|___|___|___|
          ☐ female

Ethnic Group ☐ Aboriginal/Torres Strait ☐ Maori/Cook Islands ☐ Pacific Islander
or Race: ☐ Asian   ☐ European/Pakeha
☐ Other_____

Current Age Bracket: | 18 to 19 | 20 to 29 | 30 to 39 | 40 to 49 | 50 to 59 | 60 to 69 | 70 + |

Marital Status: ☐ Single   ☐ Separated   ☐ Living with someone
                   ☐ Married   ☐ Divorced   ☐ Widowed

Highest Level ☐ 6th grade or less   ☐ High School Certificate (HSC)
of Education: ☐ 7th-9th grade or less ☐ Some tertiary certification course
                 ☐ 9th-12th grade or less ☐ Graduate or Professional Degree

Occupation
Or Job Title: |_|_|_|_|_|_|_|_|_|_|_|_|_|_|_|_|_|_|_|

IMPACT SCALE

To what extent has your child's illness and treatment:

(a) disrupted your ability to:

   (i) Pursue your goals?

   (ii) Keep up with the extra bills?

(b) interfered with:

   (i) Family life and relationships?

   (ii) Personal relationships?

*In the past **30 days**,* how many days have you missed from work, due to your child's or young person's physical or mental health?   ☐

In the past **30 days**, has your child's or young person's health interfered with…	Never	Almost Never	Some-times	Often	Almost Always
Your daily routine at work	0	1	2	3	4
Your ability to concentrate at work	0	1	2	3	4

Thank you for taking the time to fill in this survey.
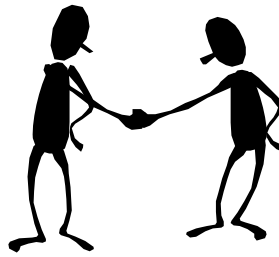
Please check that you have answered every question before
you hand it back to Jane or post it to her. If posting please use the
pre-addressed reply-paid envelope provided.

We appreciate your help with this important research.

If you need help to answer any questions, please phone Jane Ewing on the number

9514 2240

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