Economic implications of intellectual disability in childhood: a trial-based evaluation

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Certificate of original authorship

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 part of the collaborative doctoral degree and/or 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.

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Table of contents

List of figures

List of tables

List of abbreviations

2SLS	two-stage least squares
ADHD	attention-deficit/hyperactivity disorder
AIC	Akaike information criterion
ASC	alternative specific constant
ASD	autism spectrum disorder
ATC	anatomical therapeutical chemical
AUD	Australian dollar
BIC	Bayesian information criterion
CADTH	Canadian Agency for Drugs and Technologies in Health
CAPES	Child Adjustment and Parent Efficacy Scale
CBA	cost-benefit analysis
CBCL	Child Behaviour Checklist
CCA	cost-consequence analysis
CEA	cost-effectiveness analysis
CHERE	Centre for Health Economics Research and Evaluation
CI	confidence interval
CLAD	censored least absolute deviations model
CSRI	Client Services Receipt Inventory
CSRI-C	Client Services Receipt Inventory for Childhood
CUA	cost-utility analysis
CV	contingent valuation
DASS21	Depression Anxiety Stress Scale 21
DBC	Developmental Behaviour Checklist
DBC-P	Developmental Behaviour Checklist - Parent Version
DBC-P24	Developmental Behaviour Checklist - Parent Version Short Form
DBC-U4	Developmental Behaviour Checklist - Under 4
DCE	discrete choice experiment

DD	developmental disability
ECBI	Eyberg Child Behaviour Inventory
GMNL	generalised multinomial logit
GP	general practitioner
HREC	Human Research Ethics Committee
HRQoL	health-related quality of life
HTA	health technology assessment
HUI	Health Utilities Index
ID	intellectual disability
IIA	independence of irrelevant alternatives
IID	identically and independently distributed
IQ	intellectual quotient
IRR	incidence rate ratio
IV	instrumental variable
LSAC	Longitudinal Study of Australian Children
MAUI	multi-attribute utility instrument
MBS	Medicare Benefits Schedule
MNL	multinomial logit
MSAC	Medical Services Advisory Committee
MXL	mixed logit
NDA	National Disability Agreement
NDIS	National Disability Insurance Scheme
NICE	National Institute for Health Care Excellence
NR	not reported
NSW	New South Wales
OECD	Organisation for Economic Cooperation and Development
OLS	ordinary least squares
PAFAS	Parent and Family Adjustment Scale

PBAC	Pharmaceutical Benefits Advisory Committee
PBS	Pharmaceutical Benefits Schedule
PDD-NOS	Pervasive Developmental Disorder-Not Otherwise Specified
PDR	Parent Daily Report
PPC	Parent Problem Checklist
PS	Parenting Scale
PSOC	Parenting Sense of Competency Scale
QALY	quality adjusted life year
QLD	Queensland
RCT	randomised controlled trial
RUM	random utility model
SDQ	Strengths and Difficulties Questionnaire
SE	standard error
SF-36	36-Item Short Form Health Survey
SMC	Scottish Medicines Consortium
SSTP	Stepping Stones Triple P
TBPS	Total Behaviour Problem Score
TPI	Triple P International
US	United States
UTS	University of Technology Sydney
VIC	Victoria
WHO	World Health Organisation
WTA	willingness to accept
WTP	willingness to pay

Abstract

Childhood intellectual disability (ID) has a significant impact on individuals, families, society and the healthcare system. This thesis explores the economic implications of an early behavioural intervention designed to lessen these impacts, Stepping Stones Triple P (SSTP). Innovative methods are used to contribute the body of knowledge that can guide economic evaluations in this population. Particular attention is given to the inclusion of caregiver effects in economic evaluations.

SSTP is a parenting program that is designed for families of children that have a disability. It adopts a public health approach and aims to prevent and treat behavioural and emotional problems in children. It is a multilevel program with varying levels of intensity available depending on the needs of the child and the family. Data collected alongside a trial evaluating the public health roll-out of SSTP were utilised throughout this thesis. An overview of SSTP and the trial evaluating SSTP are described in Chapter 2.

This thesis comprises five related studies. The first study, presented in Chapter 3, describes the health-related quality of life (HRQoL) of caregivers who care for a child with ID. This study demonstrated that caregivers have a lower HRQoL than the age-equivalent general population. This study provides utility values of caregivers of children with ID across five comorbid disability groups (autism spectrum disorders, genetic disabilities, physical disabilities, sensory disabilities and language disabilities) that can be used to inform comparative cost-effectiveness analyses.

In Chapter 4 a discrete choice experiment was developed to estimate a monetary value of informal care tasks provided by caregivers of children with ID. The values generated by this methodology were lower than would be expected using traditional approaches, and may reflect the satisfaction and pleasure derived by an individual from providing care for a family member. The values generated in this study can be used directly in economic evaluations to estimate the cost of informal care

Chapter 5 compares the use of a time-use diary and recall questionnaire to measure the time spent providing informal care. The results demonstrated the considerable caregiver burden of childhood ID, with an average of five to six hours of care provided due to the child's disability per day. Analysis of the two methodologies showed that the recall

questionnaire overestimated the time spent providing informal care, which may be a result of recall bias, an inability to adjust for joint production or double counting of normal caregiver tasks. This chapter highlighted that the methods chosen to measure informal care can have important implications when considering the relative costs and benefits of competing interventions.

Chapter 6 presents analyses which measure the costs associated with caring for a child with ID. Trial-based data and linked administrative data were used to measure medical service use and pharmaceutical use in children and caregivers, and the productivity of caregivers. Estimates from Chapters 5 and 6 were used to calculate the total societal cost of childhood ID. The total cost to society of childhood ID was estimated to be between $9.126 billion and $14.159 billion per year. The cost of time spent caregiving accounts for the largest proportion of the total cost.

The final component of this research, presents the results of a cost-consequence analysis of SSTP (Chapter 7). The costs associated with implementing SSTP are compared to benefits derived from SSTP and other cost savings estimated in the preceding chapters. The cost of SSTP ranged from $2,661-$10,800 per family depending on the level of intervention delivered and method of implementation. Cost savings realised included productivity gains due to a lower frequency of caregiver absenteeism and reduced costs of informal care doing household errands. SSTP also resulted in an increase in costs to government, families and society due to increased service use by caregivers. Other benefits were also realised, including a reduction in child behavioural and emotional problems, improvements in parenting practices, improvements in family practices and reduced caregiver stress. The findings show that SSTP can represent excellent value for money, improving outcomes at low cost.

This thesis demonstrates the considerable burden associated with ID and the need for evidence-based interventions that can improve outcomes for this group. It demonstrates the significant role provided by caregivers and provides innovative methods for measuring and valuing informal care that can be applied to economic evaluations of complex interventions.