Application of cost-utility analysis in oral health: A preference-based quality of life measure for dental caries among adolescents

Ruvini Manjula Hettiarachchi BDS, MSc, MD
Centre for Applied Health Economics
School of Medicine
Griffith University

A thesis submitted in fulfilment of the requirement for the degree of Doctor of Philosophy

Griffith University, Queensland, Australia.

July 2020

Abstract

Oral diseases are considered a significant public health problem due to their high prevalence and impact on people's day-to-day activities and quality of life (QoL). Further, treatment of oral diseases imposes a substantial burden on health care systems, families and individuals. Cost-utility analysis (CUA) is a type of full economic evaluation preferred by most health technology assessment agencies to prioritise health care interventions in the context of limited resources. CUA compares the effects of interventions, in the form of a summary of health outcomes that incorporates both quantity and quality of life years. CUA is an important approach to evaluate oral heath interventions, as oral diseases have considerable impact on people's QoL. Hence, the application of CUA in oral health research is worthy of exploration.

Two systematic literature reviews were conducted to evaluate the application of CUA in oral health interventions and the paediatric QoL instruments used in oral health research. The first systematic review was performed to evaluate the usage of CUA in oral health interventions, the methods used and the reporting quality of CUA in publications on oral health interventions. This review identified an increasing trend of CUA use in oral health research over time, especially from 2011 to 2016. The majority of CUA publications were of good reporting quality and provided conclusions concerning the most cost-effective intervention among the different options compared; hence, these will assist in healthcare decision-making. However, among 23 CUAs published in dentistry, only four were conducted among paediatric populations and all four were related to dental caries interventions. As children are the main target group of the public health care system, the second systematic review was focused to identify the generic or disease-specific paediatric QoL instruments used in oral health research. This review provided an overview of 11 oral health-specific QoL instruments and five generic instruments used in oral health research among children and adolescents. Of these 11 oral health-specific QoL instruments, none were preference-based QoL measures (PBMs), whereas, two (CHU 9D and EQ-5D-Y) of the five generic instruments were PBMs.

The background analysis identified only limited CUAs conducted among paediatric populations on oral health interventions and found no condition-specific PBMs in oral health to be used in CUA in this population. The limited number of CUAs identified in the first review was likely due to the fact that there is no oral health-specific paediatric PBM for use in economic evaluations. PBMs are important to assist estimation

of more-accurate utility values for economic evaluations. Existing oral health-related QoL instruments are non-PBM and hence, cannot be used to generate utility values. The background analysis identified the necessity of paediatric utility measures to quantify outcomes in terms of the quality-adjusted life years (QALYs) and to promote the economic evaluation of oral health care interventions using CUA among children and adolescents. Therefore, the main aim of this study was to develop an oral health-specific preference-based QoL measure to facilitate the identification of high-value oral health interventions in adolescent populations.

All four paediatric CUAs identified in the first systematic review were all related to interventions to prevent dental caries, the most prevalent childhood oral disease. This indicates that the economic evaluations of paediatric oral health studies were mainly concentrated on dental caries; thus, the availability of a paediatric CSPBM for dental caries would facilitate better evaluation of oral healthcare interventions among paediatric populations. Therefore, dental caries was the focus of the PBM.

Preference-based measures consist of two components: a health state classification system and a set of utility weights that enable the generation of utility values for the health states defined by the classification system. The classification system for the PBM was developed as a de novo measure using literature reviews, qualitative interviews with adolescents and expert opinion using the Delphi technique. A systematic literature search of paediatric oral health-related QoL instruments was used to identify possible domains and items to be included in the classification system. Studies eliciting utility values for oral health outcomes and clinical dentistry references were also reviewed. Based on the findings, a draft classification system was developed and refined using semi-structured interviews with a convenience sample of 15 12–17-year-old adolescents who had active caries or previous experience with dental caries. The draft classification system was further refined and validated by a group of dental experts, using a modified Delphi technique. The classification system consists of five items (pain/discomfort, difficulty in eating food/drinking, worried, ability to participate in activities, and appearance) and each item has a four-level severity-based response scale. The resulting preference-based measure is named Dental Caries Utility Index (DCUI). The target group of this instrument was set as children above 12 years of age, considering that mixed dentition ends at 12 years and given the cognitive burden required for younger children to understand the concept and wordings of PBMs. Further, the Flesch Kincaid reading score of the finalised

instrument was 64.6, indicating that adolescents aged 12–13 years can easily understand the classification system.

The next stage was to generate a utility algorithm for the DCUI classification system. Discrete choice experiments (DCEs) were selected as the preference elicitation technique to value health states generated by the DCUI classification system. DCEs produce utility values on a latent scale; therefore, it is important to anchor them onto a full health-dead scale to calculate QALYs. As there were no previous studies to identify the most suitable method for dental caries health state valuation, two pilot studies were conducted prior to the main survey with two anchoring approaches: DCE with duration (DCE_{TTO}) and DCE with visual analogue scale (DCE_{VAS}). The pilot surveys were conducted online among a sample from the Australian general population. Two separate DCE designs were created using Ngene software. The DCE_{TTO} design included duration as an additional attribute, whereas DCE_{VAS} included DCE choice tasks with five attributes from the classification system and a separate visual analogue scale (VAS) task for the purpose of anchoring. Conditional logit was used to model the DCE data. Modelled DCE_{TTO} coefficients were anchored using the coefficient for the duration. Modelled DCE_{VAS} data were anchored based on two methods: using worst heath state of VAS and mapping DCE onto VAS. A total of 200 participants completed the DCE_{TTO} survey and 191 participants completed the DCE_{VAS} survey; there was no statistically significant difference between the two samples in relation to their sociodemographic characteristics. Further, there was no statistically significant difference between the participants' self-reported difficulty in understanding and completing the valuation tasks between two approaches.

The coefficient estimates from the unadjusted DCE_{TTO} model showed that the duration coefficient was in the expected direction and significant. Of the 15 coefficients estimated for each level in five dimensions, five were non-significant and one was not in the expected direction (level 2 of 'difficulty in eating food/drinking'). The coefficients of all dimensions except 'difficulty in eating food/drinking' and 'ability to participate activities' were ordered as expected (i.e., higher utility decrements were associated with increasing severity levels). Therefore, an adjusted model was estimated by combining both levels 1 and 2 of 'difficulty in eating food/drinking' and levels 3 and 4 of 'ability to participate activities'. All coefficients estimated from this adjusted model were in the expected direction and were logically consistent. Of the 14 coefficients estimated, 10 including the coefficient for duration were significant. Therefore, anchoring of the

coefficients onto the full health-dead scale was performed based on the adjusted DCE_{TTO} model. In the DCE_{VAS} approach, all coefficients estimated from the unadjusted model were in the expected direction, except that of level 2 of 'appearance', and the magnitude of the coefficients increased with the severity level of each dimension. Of the 15 coefficients estimated, four were non-significant. Therefore, an adjusted model was estimated by combining levels 1 and 2 of the dimension 'appearance'. The anchoring of DCE data onto the full health-dead scale was completed based on adjusted DCE_{VAS}, as all coefficients were in the expected direction, logically consistent and, of the 14 coefficients estimated, 11 were significant.

The resultant utility values from these two valuation approaches were compared. Rescaled coefficients from the DCE_{VAS} were in the expected order and significant compared to the rescaled coefficients from the DCE_{TTO} approach. Further, DCE_{TTO} produced more disperse utility decrements; as a result, the severe health states were valued as worse than death. This is unreliable for a condition like dental caries, in which participants may be reluctant to trade life years to avoid being in a severe dental caries health state. As the pilot data revealed that the DCE_{VAS} model performed optimally and produced more-reliable utility values for dental caries health states compared to the DCE_{TTO} approach, the DCE_{VAS} approach was utilised for the main survey.

The main valuation survey was conducted as an online survey of an age and sex representative sample of the adult Australian general population. A total of 995 adults completed the survey. The survey included a set of DCE tasks and VAS tasks, basic social-demographic questions, the DCUI, a generic preference-based measure (EQ-5D-5L) and an oral health QoL instrument (OHIP-14). DCE data were modelled using conditional logit. All estimated coefficients from the DCE data were in the expected direction and order for the five dimensions. All coefficients were statistically significant except for the second levels of the 'worried' and 'appearance' domains. The estimated coefficients were rescaled onto full health-dead scale using two methods: VAS worst heath state and mapping DCE onto the VAS. Both of these methods produce largely similar utility values for the DCUI health states. The mean absolute error value for the DCE estimates based on the mapping approach was lower compared to anchoring based on VAS worst health state. Therefore, the final utility algorithm was generated based on the rescaled coefficients from mapping DCE onto VAS. The Australian-specific tariff of DCUI ranges from 0.1681 to 1.0000. The utility algorithm will enable the calculation of utility values from the participants' responses for DCUI in economic evaluations.

This study has certain limitations. Due to resource constraints and feasibility concerns, the DCUI was developed for dental caries economic evaluations among Australian adolescents. However, there is great potential for the DCUI to be used in adolescents in other countries, as an adult measure or to evaluate oral health conditions other than dental caries. Therefore, future research is recommended in these areas. Further, the DCUI should be validated rigorously with target group adolescents at dental clinic settings. Due to the methodological constraints, health state valuation was conducted using an adult sample, a common method to elicit preferences for paediatric PBMs. Therefore, future studies are recommended to assess whether there is any significant difference between the health state utility values for DCUI derived from an adult sample and the preferences of an adolescent sample.

A new health state classification system and utility algorithm completed the new Preference-based QoL measure for dental caries: Dental Caries Utility Index (DCUI). The DCUI will facilitate the assessment of oral health interventions using a CUA framework and will aid resource allocation through economic evaluations for dental caries, the most prevalent childhood disease among Australian adolescents.

STATEMENT OF ORIGINALITY

This work has not previously been submitted for a degree or diploma in any university. To the best of my knowledge and belief, the thesis contains no material previously published or written by another person except where due reference is made in the thesis itself.

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Ruvini Manjula Hettiarachchi

July 2020

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List of Abbreviations

BWS	best-worst scaling
CBA	cost-benefit analysis
CC	conventional crown
CDB	conventional dental bridge
CEA	cost-effectiveness analysis
CFDP	conventional fixed dental prosthesis
CHEERS	Consolidated Health Economic Evaluation Reporting Standards
CHQ	Child Health Questionnaire
CHU9D	Child Health Utility 9D index
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CMA	cost-minimisation analysis
COHIP	Child Oral Health Impact Profile
COHIP-SF 19	Child Oral Health Impact Profile-Reduced
COHQoL	Child Oral Health Quality of Life Questionnaire
C-OIDP	Child Oral Impacts on Daily Performances Questionnaire
CPQ 11-14	Child Perceptions Questionnaire for 11 to 14 years
CPQ 8-10	Child Perceptions Questionnaire for 8 to 10 years
CSPBMs	condition-specific PBMs
CUA	cost-utility analysis
DALYs	disability-adjusted life years
DCEs	discrete choice experiments
DCUI	Dental Caries Utility Index
DFTO	dental free-time trade-off
DMFS	decayed, missing and filled permanent teeth surfaces
DVAS	dental visual analogue scale
ECOHIS	Early Childhood Oral Health Impact Scale
EQ -5D -Y	EuroQoL-5D youth version
EQ-5D-5L	EuroQoL-5D- 5 level version
FIS	Family Impact Scale
GUHREC	Griffith University Human Research Ethics Committee
HTA	health technology assessment
HYEs	healthy-years equivalents

ICER	Incremental cost-effectiveness ratio
ITQOL	Infant and Toddler Child Quality of Life Questionnaire
MAE	mean absolute error
MAUIs	multi-attribute utility instruments
MIQ	Malocclusion Impact Questionnaire
MNL	multinomial model
OHIP	Oral Health Impact Profile
OHRQoL	oral health-related quality of life
PBMs	preference-based quality of life measures
P-CPQ	Parental-Caregiver Perceptions Questionnaire
PedsQL	Paediatric Quality of Life Inventory
POQL	Paediatric Oral Health-related Quality of life Questionnaire
QALYs	quality-adjusted life years
QAPY	quality-adjusted prosthesis year
QATY	quality-adjusted tooth-years
QLTY	quality-of-tooth-years
QoL	quality of life
RPD	partial removable denture
RUT	random utility theory
SG	standard gamble
SOHO-5	Scale of Oral Health Outcomes for 5-year-old children
STI	single tooth implant
TOQOL	Teen Oral Health-Related Quality of Life instrument
TTO	time trade off
VAS	visual analogue scale
WHO	World Health Organization

Acknowledgements

This doctoral thesis was carried out under the primary supervision of Prof. Paul Scuffham with the assistance of A. Prof Joshua Byrnes and Dr. Sanjeewa Kularatna. I acknowledge their cooperation, time and contributions throughout the thesis project. I wish to express my gratitude to them for their constant motivation and encouragement. Particularly I would like to express my sincere gratitude to Prof. Paul Scuffham for his guidance and support given me to complete my thesis. I wish to thank A. Prof Joshua Byrnes for his support and providing me feedback on my research work. I am also grateful to Dr. Sanjeewa Kularatna for encouraging me and being supportive during the stressful phase of thesis completion.

I would like to thank A.Prof. Gang Chen and Dr. Brendarn Mulhern for their continuous support as experts in the field especially for the designing and analysis of the health state valuation study. I would like to mention A.Prof. Gang Chen for his guidance on methodological aspects of the research, statistical analysis and always being available despite his busy schedule.

I am very grateful to Griffith University for providing me scholarships, support and guidance to complete my thesis. I wish to thank Centre for Applied Health Economics for providing me necessary support to complete my Thesis.

I am very thankful to my loving husband, Deepal who has lovingly and without complaining helped me with the housework and looking after my daughter. He has been the strength for me throughout this journey. Thank you my lovely daughter Dilenya for understanding me and encourage me with lots of love and cuddles when I was in hard times. I would not have been able to complete this thesis without my family support and understanding. I will be failing in my duties if I do not express my gratitude to parents for their sacrifices, support and love without which I would not have been what I am today. Also I would like to thank my friends and family members who have supported me along the way.

A very special gratitude goes out to study participants and expert panel members for devoting their time for my research. Without them, this project would not be a success. My special thanks to Alex Parkman and SurveyEngine team for their technical support and hosting online survey especially the technical team for applying their skills to design advanced slider tasks for my online survey. I would like to thank Dr. Megan Cross for

helping me with proof reading, according to the guidelines laid out in the university-endorsed national *Guidelines for Editing Research Thesis*.

I also would like to acknowledge and grateful to my examiners for the time and effort they have dedicated to reading this thesis and for providing me with valuable feedback to improve my work as a researcher.

List of publications and conference abstracts from Doctoral research

Publications and submitted manuscripts

- Hettiarachchi RM, Kularatna S, Downes MJ, Byrnes J, Laloo R, Kroon J, Johnson N, Scuffham PA. The cost-effectiveness of oral health interventions: A systematic review of cost-utility analyses. Community Dent Oral Epidemiol. 2018;46(2):118–124. https://doi.org/10.1111/cdoe.12336

- Hettiarachchi RM, Kularatna S, Byrnes J, Scuffham PA. Pediatric Quality of Life Instruments in Oral Health Research: A Systematic Review. Value Health. 2019;22(1):129-35. https://doi.org/10.1016/j.jval.2018.06.019

- Hettiarachchi RM, Kularatna S, Byrnes J, Mulhern B, Chen G, Scuffham PA. Valuation study for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents - Study Protocol. BMJ Open 2020; 10:e038626. https://10.1136/bmjopen-2020-038626

- Hettiarachchi RM, Byrnes J, Kularatna S, Chen G, Mulhern B, Scuffham PA. Development of a classification (descriptive) system for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among adolescents- Submitted to Journal of Public health Dentistry Manuscript ID-JPHD-OA-05-20-0212

Conference Abstracts

- Hettiarachchi RM, Kularatna S, Byrnes J, Scuffham PA. (2017, December 5). *Children and Adolescents quality of life instruments in oral health research: A systematic review.* Poster presented at the Griffith Health HDR research day. Gold Coast, Australia. Conference proceedings poster presentation.

Acknowledgement of published papers

Included in this thesis are papers in *Chapters 2,3,4,5 and 6* which are co-authored with other researchers. My contribution to each co-authored paper is outlined at the front of the relevant chapter. The bibliographic details/status for these papers including all authors, are:

Chapter	Paper
Chapter 2	Hettiarachchi RM, Kularatna S, Downes MJ, Byrnes J, Laloo R, Kroon J, Johnson N, Scuffham PA. The cost-effectiveness of oral health interventions: A systematic review of cost-utility analyses. Community Dent Oral Epidemiol. 2018;46(2):118–124. https://doi.org/10.1111/cdoe.12336 Hettiarachchi RM, Kularatna S, Byrnes J, Scuffham PA. Pediatric Quality of Life Instruments in Oral Health Research: A Systematic Review. Value Health. 2019;22(1):129-35. https://doi.org/10.1016/j.jval.2018.06.019
Chapter 3	Hettiarachchi RM, Byrnes J, Kularatna S, Chen G, Mulhern B, Scuffham PA. Development of a classification (descriptive) system for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among adolescents- Submitted to Journal of Public health Dentistry Manuscript ID-JPHD-OA-05-20-0212
Chapter 4	Hettiarachchi RM, Kularatna S, Byrnes J, Mulhern B, Chen G, Scuffham PA. Valuation study for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents - Study Protocol. BMJ Open 2020; 10:e038626. https://10.1136/bmjopen-2020-038626
Chapter 5	Hettiarachchi RM, Byrnes J, Kularatna S, Chen G, Mulhern B, Scuffham PA. Comparison of two approaches to value health states derived by preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents Prepared for submission to the European Journal of health Economics

Chapter 6	Hettiarachchi RM, Kularatna S, Byrnes J, Chen G, Mulhern B, Scuffham PA. Valuing a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI): Using DCE and VAS to develop an Australian utility algorithm Prepared for submission to the journal Value in Health

Appropriate acknowledgements of those who contributed to the research but did not qualify as authors are included in each paper.

(Signed) _____ (Date)_____

Ruvini Manjula Hettiarachchi

(Countersigned) _____ (Date)_____

Supervisor: Prof. Paul Scuffham

Chapter 1. Introduction

1.1 Background

1.1.1 Oral diseases

Oral diseases are considered a significant public health problem (1). Being one of the most prevalent chronic conditions, oral disease affects more than 3.5 billion people worldwide (2), impacting their day-to-day activities and quality of life (QoL). Oral diseases are one of the most expensive diseases to treat and have a substantial effect on the economy of an individual, their family and the health care system (3). Oral diseases comprise a wide range of conditions, including dental caries, periodontal (gum) disease, oro-dental trauma, oral cancer, oral manifestations of HIV infection and developmental defects such as dental fluorosis (2). Oral diseases are highly prevalent among poor and disadvantaged socio-economic groups. Unhealthy diets with high free sugar, tobacco and alcohol consumption are the major risk factors for oral diseases, which share these modifiable risk factors with other leading non-communicable diseases such as diabetes and cardiovascular conditions (2).

Dental caries, also known as tooth cavities or decay, is a breakdown of teeth due to acidic by-products from bacterial fermentation of dietary carbohydrates (4). The presence of early cariogenic bacteria, poor oral hygiene and frequent consumption of a sugary diet are the risk factors for dental caries. According to the Global Burden of Disease study 2017, untreated dental caries in permanent teeth was the most common disease condition and it was estimated that 2.3 billion people were suffering from dental caries in their permanent teeth (5). Moreover, dental caries is the most prevalent oral disease among children and adolescents. It is estimated that dental caries in primary teeth affects more than 530 million children (2). In Australia, data from the National Child Oral Health Study 2012–14 (6) indicated that 38.2% of 12–14-year-old children experienced dental caries in their permanent teeth. Approximately 15.4% of 12-14 year old Australian children had a permanent tooth with untreated dental caries; 1.6% had at least one permanent tooth missing due to dental decay and 28.3% had at least one permanent tooth filled because of dental decay (6). The average number of decayed, missing and filled permanent teeth surfaces (mean DMFS) for 12–14-year-old children was 0.9 (6). The Australian National Survey of Adult Oral Health 2017–2018reported aggregated data for people 15–34 years old (7); thus, national data for 15–19 years olds adolescents are not

available. However, a study conducted in New South Wales, Australia, found that 44.4% of 14–15-year-old adolescents had dental caries (8).

1.1.2 Impact of oral diseases among children and adolescents

Oral health is an integral aspect of general health and wellbeing. It influences how people enjoy life, speak, eat and socialise (1). Good oral health can have a positive impact on an individual's overall health, emotional and social well-being, and self-esteem (9). In contrast, oral diseases are the most common chronic disease and oral health problems can disturb daily activities, positive social interactions and emotional wellbeing. As oral disorders have a significant impact on an individual's physical, social and emotional well-being, the terms QoL and oral health-related QoL have become widely popular in the oral health research arena in recent years and more research has focused on the effect of oral health on QoL (10, 11). Oral health-related quality of life (OHRQoL) is a multidimensional construct that includes a subjective evaluation of oral health, functional well-being, emotional and social wellbeing, and satisfaction with care (11). It reflects an individual's satisfaction with their day-to-day functionalities, such as eating, sleeping and engaging in social interaction, and their self-esteem with respect to their oral health (10). Locker and Allen defined OHRQoL as the 'impact of oral disorders on aspects of everyday life that are important to patients and persons, with those impacts being of sufficient magnitude, whether in terms of severity, frequency or duration, to affect an individual's perception of their life overall (12)'.

Multiple studies have demonstrated that oral health problems negatively impact the QoL and daily activities of children (13, 14) and adolescents (15). Dental caries, the most common chronic childhood disease, has a negative impact on eating, sleep, school performance, smiling patterns and social interactions (14). Children with traumatic injuries to teeth are more likely to report negative effects on eating and enjoying food, smiling and laughing, and interactions with other people compared to children without any traumatic injury (15). Oral disorders such as malocclusion (16) and dental fluorosis (17) have also been proven to have negative impacts on children's QoL and their day-to-day living. Moreover, adolescence is the age when many children have a greater concern for the appearance of their face and teeth. Positive social interaction and greater self-esteem are critical for development from adolescence into adulthood (9). Thus, the psychological impact of oral diseases can disrupt QoL, self-esteem and positive social interaction, and has negative impacts on adulthood as well (1).

1.1.3 Economic burden of oral diseases

The current treatment options available for oral diseases are generally expensive and the costs of dental treatment impose a significant burden on individuals, families and health care systems. The provision of oral health care accounts for around 5% of the total health budget of most high-income countries, whereas the provision of oral health care is beyond the budgetary capacity of most low- and middle-income countries (2). In Australia, 6% of the total health expenditure (AU$10.2 billion) was recurrent expenditure on dental services and per capita dental expenditure was AU$416 in 2016–2017 (18). In addition to direct health care cost, oral diseases incur inevitable indirect costs due to loss of productivity at work and school and reduce QoL. As money and resources are limited, health care planners require trustworthy information to select oral health care interventions that provide the best value for money by maximising the QoL and reducing the associated burden to the individual, family and health care system (19). Health technology assessment (HTA) is a multidisciplinary process that refers to the systematic evaluation of the properties and effects of health interventions or technologies (20). The main purpose of HTA evaluations is to inform health policy decision-making (20). Economic evaluation is one of the tools in the HTA process that assists in prioritising health care interventions, which provide maximum health impact in a resource scarce setting (19, 21, 22).

In Australia, the health care system is funded through a combination of public and private sectors: federal and state governments, private health insurers, and individual out-of-pocket payments by patients. The Australian Government HTA process is conducted through HTA agencies: the Therapeutic Goods Administration (TGA), the Medical Services Advisory Committee (MSAC), the Pharmaceutical Benefits Advisory Committee (PBAC) and the Prostheses List Advisory Committee (PLAC) (23). Each HTA agency supports the HTA framework through:

- Evaluation of the safety and efficacy of health technologies
  The approval of therapeutic goods prior to marketing is granted through the Australian Register of Therapeutic Goods (ARTG). All dental and surgical instruments, dental fillings and devices (e.g., orthodontic braces) approved to use in Australia are listed under the ARTG (24).
- Appraisal of the comparative effectiveness of health technologies. HTA agencies assist public funding decisions through the Medicare Benefits

Schedule (MBS), the Pharmaceutical Benefits Scheme (PBS) and the National Immunisation Programme (NIP), the private insurance reimbursement of prosthetic devices through the Prostheses List and conducting post market surveillance (23).

Commonly used drugs in dentistry such as antibiotics are listed under the PBS drugs list (25). Currently, Medicare under the MBS covers some surgical procedures performed by approved dentists such as inpatient oro-maxilofacial surgeries and specified items under the Cleft Lip and Palate Scheme (26). Also, Medicare covers specified dental care services for eligible children under the Child Dental Benefits Schedule and eligible adults. However, eligibility for public dental services and the dental procedures covered through public funding varies widely across different states and territories (26). Recently, proposals have been suggested to expand Medicare funding for dental services (27). Further, public funds are allocated to health research and public health interventions. The National Health and Medical Research Council recommended improving funding for research related to public health interventions (28). Australia has launched the National Oral Health Plan 2015–2024, in which targets have been set to achieve better oral health among the Australian population by the year 2025 (29). It has been identified that the expansion of oral health care services and the establishment of public health interventions to improve the oral health are important to achieve these targets (30). The formal HTA processes have the potential both to guide funding decisions on the expansion of health care services and to select the public health interventions with the best value for money (31).

1.1.4 Health economic evaluation of oral health care interventions

Economic evaluation of a health intervention can be defined as the 'comparative analysis of alternative courses of action in terms of both their costs and consequences' (32). Different types of economic evaluations are used to assess health care interventions. The measurement of cost is similar across most economic evaluations; however, they differ in the nature of outcomes measured (32). Cost-minimisation analysis (CMA) refers to a situation where the outcomes of two or more health interventions are considered equivalent; thus, value for money is solely dependent on the intervention that has the lower cost (32). Cost-effectiveness analysis (CEA) measures the effectiveness of competing interventions in terms of natural units such as number of decayed teeth. Cost-utility analysis (CUA) measures effect in terms of health-related utility, in the form of a summary health outcome that considers both life years and the quality of those life years

(32). CUA is considered a variant of CEA (32). In cost-benefit analysis (CBA), both cost and outcomes are measured in monetary units (32).

Stephen and Campbell published the first economic evaluation in dentistry in 1978 (22, 33) and since then, several systematic reviews reported an increasing number of publications related to economic evaluations of oral health care interventions (3, 22). The majority of the economic evaluations examined dental caries prevention; however, it is a noteworthy trend that there are economic evaluations related to highly expensive dental treatments such as implants (34). Further, there have been improvements in the reporting quality of the published economic evaluations in dentistry. Nevertheless, the absence of some important analysis, insufficient reporting of outcomes and cost measurement, and misuse of terminologies were observed when these economic evaluations were compared with standard check-list criteria (3, 22, 35). Rogers et al reported a lack of high-quality economic evaluations in oral health research among paediatric populations (36).

1.1.5 Cost-utility analysis (CUA)

CUA has recently become increasingly popular in health economic evaluation. Here, costs are measured in monetary units and the outcomes are measured as a summary health measure, in terms of both quantity and quality of life (32). This approach allows the comparison of cost-effectiveness across various disease conditions as well as different health care interventions (19, 21). Hence, CUA has become the most common type of economic evaluation used to assess health interventions and assist health care decision-making processes (37). CUA is the form of health economic evaluation accepted by the health technology assessment authorities of many developed countries, such as the United Kingdom, Canada and Australia (37-40).

1.1.5.1 CUA and oral health

Over the past decades, health care provision has concentrated on the patient with a growing attention on patient-reported outcomes (11) and patient preferences to assess the effectiveness of health care interventions, understand disease burden and health inequalities, and prioritise health care resource allocation (41). Patient-reported outcome measures (e.g., health-related quality of life) are those that patients report about their own health (42). Their preferences explain the desirability of a specific health state of an individual; in health economics, utility is the measure of preference or value that is placed upon a particular health state (32). Since CUA incorporates both the quantity and quality

of life as a summary health measure to quantify health outcomes (32), CUA is now recognised as an important form of economic evaluation in the context of patient-centred health care provision in every area of health care, including oral health. Moreover, oral diseases have considerable impact on QoL (43); thus, CUA is an important approach to evaluate oral health interventions. However, a recent systematic review of economic evaluations in dentistry reported that there were only eight CUAs out of 79 published full economic evaluations from 1975 to 2013 (22). A critical and comprehensive review of the use of CUA in oral health has not been undertaken. Therefore, the use of CUAs of oral health interventions is worth exploring and will facilitate better adoption of CUA within the field of dentistry.

1.1.5.2 CUA and QALYs

Quality adjusted life years (QALYs) are the most common summary outcome measure used in CUA (32, 44). QALYs combine both the quality (morbidity) and quantity (mortality) gain into a single summary measure, thus enabling comparisons of health gain or loss across different disease areas (32). QALYs are commonly used in assessments of health care technologies and public health interventions (45). The quality of life component of QALYs is measured based on preferences for different states of health rather than with respect to any particular disease or disability (45).

Disability-adjusted life years (DALYs) and healthy-years equivalents (HYEs) are the other common alternatives to the QALY (32). The DALY is a summary measure of disability and mortality in the population to estimate the burden of diseases. It is expressed as the number of years lost due to ill-health, disability or early death (44). The DALY approach is commonly used for international comparisons of disease burden (5). Both the QALY and the DALY are anchored onto a 0–to–1 health scale with interval scale properties. QALY is a measure of health expectancy; thus, 1 represents full health and 0 represents death on the QALY scale. In contrast, the DALY is a measure of a health gap and the 1 and 0 values of the DALY scale represent death (full disability) and full health (no disability), respectively (45). Unlike the QALY, the DALY incorporates QoL specific to particular diseases, rather than to health states. Thus, DALYs do not simultaneously take into account all illnesses, nor comorbid conditions within the same population or individual (45). HYEs measure the preferences over the individual's entire path of health states rather than for each state alone and are based on a two-stage standard gamble

procedure to elicit preferences (32). Therefore, the HYE approach comparatively imposes more practical difficulties in implementation; thus, it is not as popular as QALY (32).

Both QALY and DALY have been used as outcome measures in oral health economic evaluations (46). Moreover, although the common outcome measure in CUA is QALY, CUAs of oral health interventions have used different derivatives of QALY (47), such as quality adjusted tooth years (QATY) (48), quality of tooth years (QLTY) (49) and quality adjusted prosthesis years (QAPY) (50), as the outcome measure. These oral health-specific utility measures would be more sensitive when comparing the cost-effectiveness of different oral health interventions. However, unlike the QALY, these cannot be used to compare the cost-effectiveness of oral health interventions with interventions in other areas of health (51). Therefore, the use of oral health-specific utility measures may hold back the main strength of the CUA: comparison of health interventions across all areas of health to optimise the health care decision-making process.

1.1.5.3 Methods to estimates QALYs

To estimate QALYs, utility weights that represent the QoL of the health state under consideration are needed, as well as the duration of that heath state. QALYs are calculated by multiplying the time spent in each health state by the utility weight associated with that health state (52). These utility weights or health state values represent individuals' preferences for different health states and are usually presented on a full health-dead scale (53). The measurement of utility weights involves two steps: defining and valuing the health state of interest (52). Individuals' preferences for health states are obtained using a preference elicitation technique (54).

Preference elicitation techniques

Preference elicitation methods can be broadly categorised as either cardinal or ordinal. Cardinal preferences methods generate preferences in a quantitative form, that is, they provide direct estimates of the degree to which one health state is preferred over another (55). The time trade-off (TTO), standard gamble (SG) and rating scales are the commonly used cardinal preference elicitation techniques (56). In TTO experiments, individuals are given a choice between two scenarios: living in full health for a short period and living in an impaired health state for a longer time. The period living in full health is adjusted until the individual indicates no difference between the two choices

(32). In the SG method, individuals are given a choice between living in a health state of interest for a period of time with 100% certainty and living in full health for the same period of time but with a risk of immediate death. The probability of immediate death in the second alternative is modified until the participant considers there to be no difference between the two alternatives (32). Rating scales for the health state evaluation studies are usually in the form of a visual analogue scale (VAS), where the top of the scale indicates the 'best imaginable health' and the bottom indicates 'the worst imaginable health'. Individuals are asked to place the health state of interest on this scale (32). It is the simplest form, easy to understand and better than the SG and TTO methods in terms of response rate (47). The main concerns over VAS preference elicitation are that the VAS does not involve the time spent in each health state and does not explicitly involve choices or trade-offs (55).

Ordinal preference elicitation methods involve ordering preferences for two or more alternatives (55). Discrete choice experiments (DCEs) and ranking exercises such as best-worst scaling are the commonly used ordinal preference elicitation methods (55). A DCE asks respondents to simply choose which health state they prefer from a selection of alternative health states (55). Best-worst scaling (BWS) is a ranking approach. In BWS, participants are required to state their best and worst preferences, typically for three or more sets of items or profiles (57). Compared to the SG and TTO methods, ordinal preference elicitation methods such as DCE and BWS are considered easier for respondents to understand (55). In earlier studies, ordinal methods were generally used to evaluate healthcare services, treatment interventions, health policies and practices; however, they became popular in health state valuation due to reduced measurement error and ease of administration (55, 58).

Several preference elicitation studies in oral health research (42) have used different elicitation techniques, such as SG (63), TTO (64), VAS (65), DCE (66-68) and WTP (69). However, these studies were focused on health states defined by a single characteristic such as a healthy tooth, a painful tooth or preferences for different oral health treatments such as crowns, bridges and prosthetic appliances (44) rather than oral health states defined by a classification system. An interesting feature of preference elicitation tasks in dentistry is that the existing elicitation techniques were adapted to oral health. A study conducted to assess utility values using dental free-time trade-off (DFTO) and dental visual analogue scale (DVAS) for 12 dental health states among adolescents found that the DVAS is comparatively better in terms of test-retest reliability and ease of

use, and the resultant utility values were in the expected order for 12 health states compared to DFTO (64).

Direct and indirect approaches

Both direct and indirect approaches could be used in health state valuation studies (53, 59). In direct approaches, study participants rate their own health state or a hypothetical health state using one or more of the preference elicitation techniques described above (53, 55). Direct elicitation ensures that the health state values provide an accurate estimation of an individual's own preferences; however, they are time-consuming and complex to administer (52). Further, ethical issues may arise when a trial participant with severe diseases answers an instrument in which they must consider their own health in the trade-off (59).

The indirect approach involves the use of pre-scored preference-based QoL measures (PBMs) (52). These include multi-attribute classification systems (and hence, are also known as multi-attribute utility instruments: MAUIs) to define health states and predetermined utility weights are available for any combination of attribute levels that describe a particular health state (52). Study participants self-report their health states using a PBM and the utility weights for each participant's health state are calculated based on the predetermined scores. Usually these predetermined scores are generated from a separate study with direct preference elicitation administered typically to a sample of general population (54). PBMs are becoming a more prevalent method of estimating utility weights to calculate QALYs, as they are easy to use and less time-consuming, and their tariffs or scoring algorithms are readily available (47, 51, 52).

1.1.5.4 Preference-based quality of life measures (PBMs)

There are two types of PBMs: generic and condition-specific. Generic PBMs can be used among populations with any disease or condition (48), whereas condition-specific PBMs (CSPBMs) are designed for specific diseases or conditions. To date, several different commonly used generic preference-based measures have been developed, including the EuroQoL-5D, SF-6D and CHU9D (47). Patients can complete these instruments within a few minutes and a utility score is produced using an algorithm generated often from a sample of the general population. In addition, disease-specific PBMs have been developed for certain disease conditions because generic PBMs may not include all of the important domains for each and every disease; therefore, generic PBMs

may not be sensitive to clinically meaningful changes in a patient's health (50). PBMs consist of two components: a health state classification system (also known as a descriptive system) and a set of utility weights that enable the generation of utility values for the health states defined by the classification system (60, 61).

Developing a classification system for a Preference-based QoL measure

A classification system is composed of a set of dimensions or attributes with response levels designed to measure health. The classification system of a PBM can be developed *de novo* or by either using an existing non-preference-based QoL measure (62). The development of CSPBMs from existing non-preference-based QoL measures is often subject to certain methodological constraints related to the underlying non-preference-based QoL measure. These non-preference-based QoL measures are often complex and lack a clear multidimensional structure (62). Researchers have used different approaches, such as literature reviews, qualitative research and expert opinion, either alone or in combination, to develop a PBM as a *de novo* measure. Brazier et al (62) revealed that 10 of 26 studies developed a classification system *de novo*.

In oral health research, there has been a tremendous increase in the assessment of OHRQoL among paediatric populations (11) over the past decades and several non-PBM OHRQoL instruments have been developed and validated. However, a comprehensive review of available OHRQoL instruments for paediatric populations has not taken place. Gilchrist et al assessed the methodological quality of paediatric OHRQoL instruments but included three most frequently used OHRQoL instruments only (63). Therefore, a comprehensive review of paediatric quality of life instruments used for oral health research will facilitate an overview of the use of PBMs in oral health research and identify the most suitable process to develop a classification system for an oral health preference-based QoL measure.

Utility scoring algorithms

Scoring algorithms generate utility values for all health states defined by the classification system based on elicitation results for a select number of health states. Once these values are obtained, statistical modelling techniques are used to generate an algorithm and provide estimated utility values for all possible health states derived from the classification system. Random utility theory (RUT) is used as the theoretical basis for the modelling of health states values. The RUT assumes that the utility value attached to

alternatives in a choice scenario can be summarised by an explainable (or systematic) component and an unexplainable (or random) component (55).

Previously, conventional valuation methods such as SG or TTO were commonly used to value health states to develop utility algorithms. In addition to the decreased respondent burden, DCEs and BWS are typically conducted without an interviewer and are compatible with online surveys, which expedites the data collection process (64). Moreover, in DCE health state design, the choice tasks consist of attributes and severity levels for each attribute; it is compatible with the structure of PBM classification systems. Therefore, ordinal methods such as DCEs and BWS become more popular in recent health state valuation studies (65).

Anchoring

Ordinal methods such as DCE and ranking tasks produce utility values on a latent scale. Therefore, it is important to anchor the utilities generated from the DCE design to the full health-dead scale to calculate the QALY (66). Several anchoring methods have been proposed by recent studies. Most are based on the TTO method and common practice is to conduct a separate TTO study (53) with a smaller sample and then rescale the DCE data onto the full health-dead scale using different statistical procedures such as anchoring the worst state using TTO (67), mapping DCE into TTO (66, 67), and DCE/TTO hybrid models (66). Bansback et al (64) proposed a method (DCE_{TTO}) that incorporates duration as an additional attribute of the DCE choice task defined by the classification system to be valued (68). This method has been widely used in recent research, as it overcomes the necessity of conducting a separate study in addition to the DCE for the purpose of anchoring. In addition, anchoring using the coefficient for 'dead' (66) and willingness-to-pay (WTP) to avoid the mild and severe health states (69) has also been used as anchoring approaches.

1.1.5.5 Whose preference should be valued?

Researchers continue to argue over whose preference should be valued. Some argue that the health states of interest should be valued by the patients because they know them better than the general population. Conversely, the general population will provide relatively unbiased valuations, as they do not have a special interest in a particular treatment (generally). Simultaneously, public money will often be allocated to fund a new treatment and therefore, it is the general public who should value the health states.

Preference elicitation for health states from children and adolescents is considered still more challenging and there is a continuous debate as to whether the parents/guardians or adult general population could or should be used as proxy for the child or adolescent. For infants, toddlers and pre-schoolers, researchers must depend on a proxy respondent, usually a parent, to value health states (54). Montgomery and Kusel (70) emphasised that children are not 'mini adults' and that preferences related to paediatric health care provision should be elicited from children. Several paediatric preference-based QoL measures have recently been introduced (53). However, valuing health states directly from paediatric populations is associated with methodological constraints, as identifying a suitable valuation technique for this group is relatively challenging (53). Health state valuations may impose a great cognitive burden on children and adolescents. Further, certain techniques consider 'immediate death' during the valuation tasks; asking children to value 'immediate death' is generally ethically inappropriate. Therefore, it is fairly common to conduct valuation studies for paediatric PBMs using an adult general population sample and to develop utility algorithms based on general population weights (70). DCEs are easier for paediatric populations to understand than other traditional methods; however, methodological constraints require specially designed anchoring methods to rescale the DCE values to a full health-dead scale. For instruments such as the CHU9D, valuation tasks were conducted among children using BWS and, for the purpose of anchoring, a separate TTO study was conducted with a young adult sample (67).

The preference elicitation studies in oral health among children and adolescents were conducted using samples of children (71), parents or the adult general population (72, 73). Barber et al (74) developed a DCE study to elicit preferences for adolescent and parent preferences for hypodontia treatment and emphasised the importance of including adolescents during the development of DCE health states to ensure that a valid and reliable health state survey results. A study conducted in the UK (75) used a DCE to elicit utility weights for dental health states to inform an economic analysis of oral health promotion approaches for child dental caries; however, this study used proxy respondents from an online sample of the general adult population.

Valuing health state preferences for a prevalent condition like dental caries using a general population sample is justifiable due to several reasons. In Australia, only 9.9% of the population aged 15 years or over had no experience of dental decay in their permanent teeth (7), indicating that the vast majority have experienced dental caries in their lifetime. Further, more than 30% of those aged 15 years and over had untreated

coronal decay and an average of 12.8 decayed, missing and/or filled teeth (7). Thus, members of the general population would be able to value the dental caries health states through the patients' perspective as well. Public taxes are used for funding the federal and state public dental services, implementing population-based preventive interventions, providing incentives for the purchase of private health insurance (an indirect taxpayer contribution) and funding universities that educate dental health professionals and conduct oral health research (7). Current oral health care provision in Australia offers eligible children aged 2–17 years dental care through the Australian Government-funded Child Dental Benefits Schedule. In 2016–2018, 50% of Australian children in this age range accessed government-funded oral health programmes (29). Dental care for children who are not eligible for Child Dental Benefits Schedule is paid by their parents as direct out-of-pocket payments or as an insurance premium. In 2017-18, 58% of the total expenditure on dental services comprised out-of-pocket costs paid directly by individuals and around 19% was from health insurance funds (18). Thus, parents' preferences are important for deciding which treatment or preventive options give the best value for money. Considering all these facts, valuing health state preferences for dental caries using a general population sample is justifiable, since this sample could provide reliable information, as around 90% of the population have experience of dental caries and also made a greater contribution to dental care expenditure through: tax, out-of-pocket expenses and insurance premiums.

Background analysis of the application of CUA in oral health revealed that a comprehensive review of the subject has not been undertaken. Further, a comprehensive review of paediatric QoL instruments and the use of PBMs in oral health research form a gap in the literature. Therefore, based on this background analysis, the following objectives were proposed for the present study.

1.2 Objectives

General objective

To develop an oral health-specific preference-based quality of life measure to facilitate the identification of high-value oral health interventions in adolescent populations.

Specific Objectives

1. To conduct a comprehensive systematic literature review to assess CUAs of oral health interventions
2. To conduct a comprehensive systematic literature review to identify the generic or disease-specific and preference- or non-preference-based paediatric QoL instruments used in oral health research
3. To develop a classification system for a PBM for dental caries among adolescents in Australia
4. To generate a preference-based algorithm of utility weights for the health states defined by the new classification system

1.3 Thesis Structure

This thesis consists of seven chapters. Chapter 1 (this chapter) provides a general introduction and overview of the current gap in research. Chapter 2 presents two published systematic review papers corresponding to Objectives 1 and 2 of this thesis. Chapter 3, which is presented in the form of manuscript, corresponds to the third objective of this thesis (i.e., to develop a classification system). Chapters 4, 5 and 6 correspond to the Objective 4 of the study and are also presented as manuscripts prepared for publication. Chapter 4 is a protocol paper that describes the methodological aspect of the health state valuation study. Chapter 5 presents the comparison of two approaches to value dental caries health states. Chapter 6 reports the health state valuation study. The concluding chapter (Chapter 7) includes a general discussion of the study methods, findings, conclusions and recommendations. An overview of the structure and content of the thesis is provided in Figure 1.1.

Overall, this thesis includes five articles formatted to meet the requirements of the peer-reviewed academic journals where they have been published, are currently under review or will be submitted. As most of the content in this thesis is in the form of journal articles, there may be some unavoidable repetition of the details in the introduction, descriptions of the study methods and discussion. All journal articles are presented in an identical format with tables and figures included within the text to ensure consistency throughout the thesis. Tables and figures, included as supplementary materials in the published journal articles, are also incorporated within the text. A single standard referencing format (Vancouver) has been used throughout the thesis and the references are provided at the end of the thesis to avoid duplication.

This thesis is prepared in accordance with Griffith University policies (griffith.edu.au/hdr/thesis preparation).

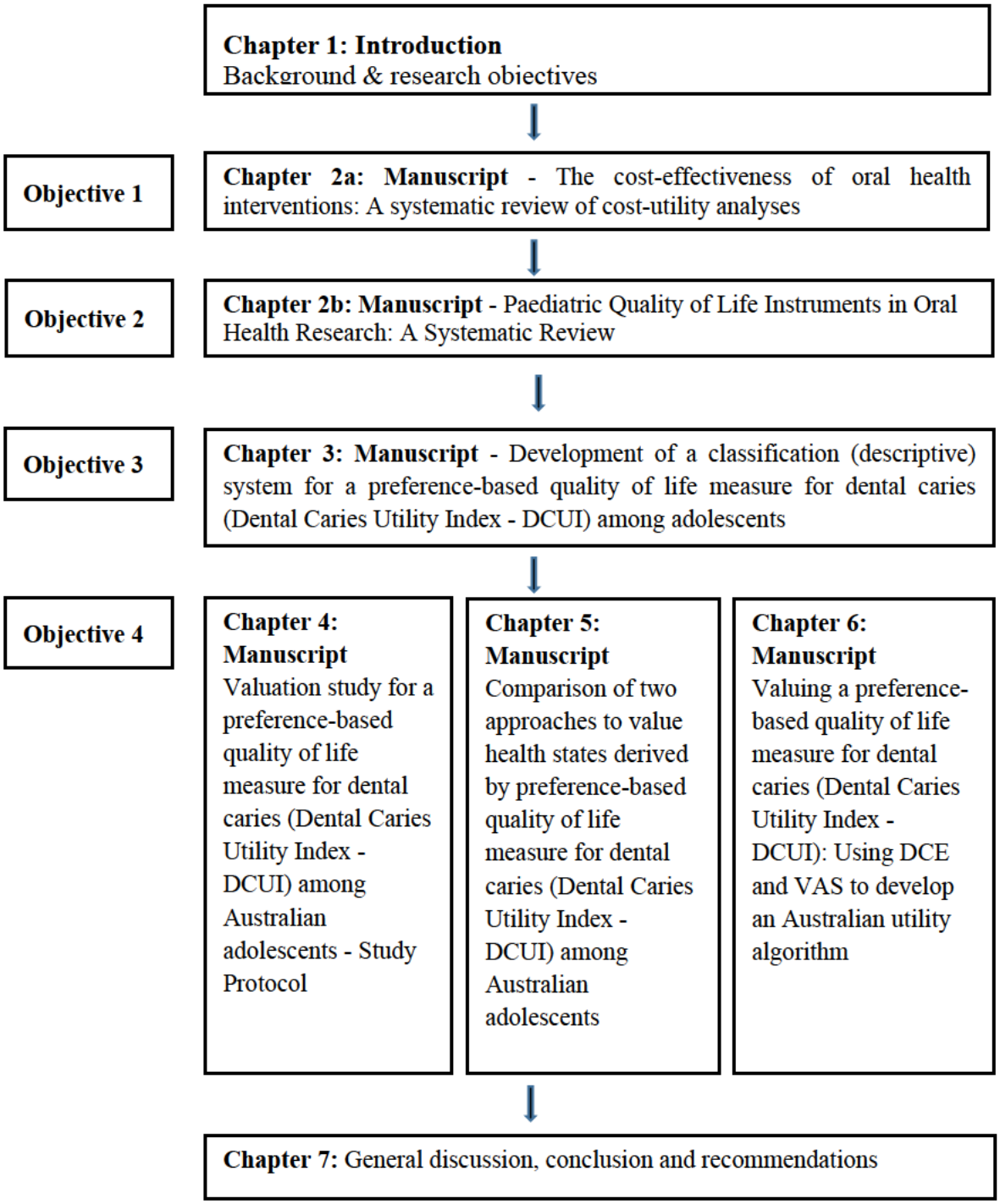

Figure 1.1: Schematic overview of the structure and content of the thesis

Chapter 2. Literature review

This chapter presents the two published manuscripts based on the two systematic reviews undertaken to achieve the objectives 1 and 2 of the study.

Manuscript 2.1 presents an overview of the usage of cost-utility analysis in oral health interventions, the methods used and the reporting quality of CUA in publications on oral health interventions. As the main aim of this review was to get an overview of use of CUA in oral health research, this was not confined to CUA conducted among paediatric populations.

Manuscript 2.2 presents the findings from a comprehensive systematic literature review to identify the generic or disease specific and preference- or non-preference-based paediatric quality of life instruments used in oral health research.

2.1 The cost-effectiveness of oral health interventions: A systematic review of cost-utility analyses

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a published co-authored paper. The bibliographic details of the co-authored paper, including all authors, are:
Hettiarachchi RM, Kularatna S, Downes MJ, Byrnes J, Laloo R, Kroon J, Johnson N, Scuffham PA. The cost-effectiveness of oral health interventions: A systematic review of cost-utility analyses. Community Dent Oral Epidemiol.2018; 46 (2):118–124. https://doi.org/10.1111/cdoe.12336

Copyright statement:
The article has been published in a peer-reviewed journal with an international readership. The Impact Factor (2018) for this journal is 2.278. The journal 'Community Dentistry and Oral Epidemiology' allows the authors to archive post-print (accepted peer-reviewed) version after an embargo period of 12 months. Further details are available here: https://authorservices.wiley.com/author-resources/Journal-Authors/licensing/self-archiving.html

My contribution to the paper involved the designing of the study, literature search, quality assessment of the included studies, data analysis and drafting the article.

(Signed) _____ (Date)_____

Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____
Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____
Supervisor: Prof. Paul Scuffham

The cost-effectiveness of oral health interventions: A systematic review of cost-utility analyses

Hettiarachchi RM, Kularatna S, Downes MJ, Byrnes J, Laloo R, Kroon J, Johnson N, Scuffham PA.

2.1.1 Abstract

Objectives

To assess the usage of cost-utility analysis (CUA) in oral health interventions and to evaluate the methods used and the reporting quality of CUA in publications on oral health interventions.

Methods

A systematic review was performed on literature published between 2000 and 2016 where cost-utility analyses of oral health interventions were included. The reporting quality of these oral health CUAs was assessed against the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) checklist.

Results

Of the 6637 publications identified initially, 23 met the inclusion criteria. Of these, 14 (61%) had been published in the last 6 years. Included studies were on oral cancer (n = 6), provision of dental prosthesis (n = 6), dental caries (n = 4), periodontal diseases (n = 3), antibiotic prophylaxis (n = 2), dento-facial anomalies (n = 1) and dental service provision (n = 1). Twenty-one studies were able to identify the most cost-effective intervention among the different options compared. Of the 23 studies identified, 15 (65%) used quality-adjusted life years (QALY) as the outcome measure, and 18 (78%) reported an incremental cost-effectiveness ratio. The economic perspective was clearly stated in 13 articles (57%). Twenty studies (87%) reported the discount rate, and 22 (96%) undertook sensitivity analysis. The reporting quality of studies, appraised by the CHEERS checklist, varied from 75% to 100% (median 92%).

Conclusion

The use of CUAs in evaluation of oral health interventions has been increasing recently, especially from 2011 to 2016. The majority of CUA articles were of good reporting

quality as assessed by the CHEERS checklist and were able to provide conclusions regarding the most cost-effective intervention among the different options compared: this will assist in healthcare decision-making and resource allocation. These positive outcomes of our study encourage wider use of CUAs within the dental and oral health professions.

Keywords

Cost-utility analysis, economic evaluations, oral health, systematic review

2.1.2 Introduction

Oral diseases are among the most common forms of chronic disease and are one of the most expensive to treat (1). The World Health Organization (WHO) has estimated that 60-90% of school children and the majority of adults suffer from dental caries in most industrialized countries (76). Moreover, oral cancer was ranked the 15th most common cancer in the world in 2012 (77). The economic burden of oral diseases is considerable, and the provision of oral health care represents a substantial proportion of the total health budget in many countries (1). For example, dental care expenditure in the USA was $113.5 billion US dollars (per capita dental expenditure was 351 US dollars) in 2014 (78), whereas, in Australia, recurrent expenditure on dental services was $8.7 billion Australian dollars (per capita dental expenditure was 380 Australian dollars) in 2012–13 (79).

Many new treatment options and preventive strategies have been introduced to combat oral diseases, but these are usually associated with high costs and health planners require valid information to make informed decisions to allocate scarce resources for these competing strategies (19). Economic evaluation is one of the tools that allows health planners to identify interventions which provide the best value for money (19, 21, 22). They provide a valuable contribution in the optimal selection among competing options and assist in deciding how to allocate health care resources to gain maximum health impact (19).

Cost-utility analysis (CUA) is a type of full economic evaluation in which cost is measured in monetary units and the outcomes are measured as a summary measure of health gain, in terms of both quantity and quality of life (32). The primary measure used in CUA is Quality-Adjusted Life Years (QALY) (32). The expression of health outcomes as a combination of both length and quality of life in a CUA allows the comparison of the cost-effectiveness across different interventions (such as drugs or procedures) and

different health domains (such as dental caries, cancer, myocardial infarction) (19, 21, 80). Hence, CUA has recently been used extensively to prioritise health care interventions and inform decision-making processes, especially in high-income countries (78). Many health advisory agencies in Europe, Canada and Australia recommend using CUA in health economic evaluations (37-40).

Despite the growing demand for economic evaluations in oral health care provision, the use of CUA is limited. Cost-effectiveness analysis (CEA), which incorporates different health outcomes in 'naturally occurring' units (such as cost of preventing a case), is the most common form of full economic evaluation in dentistry (19, 22). CUA is a special type of CEA which measures a health outcome in terms of health-related quality of life. This is an important approach for evaluating oral heath interventions because oral diseases have considerable impact on people's quality of life (QoL) (81). Nonetheless, limited use of CUA in oral health is evident from a recent systematic review (22), in which the authors identified only eight CUA analyses out of 79 full economic evaluations published in dentistry during 1975 to 2013. A critical review of the use of CUA in oral health has not been undertaken to date, although its application to other health conditions such as HIV/AIDS, diabetes and cancer (82-84) has been reviewed comprehensively. Accordingly, we conducted this systematic review to assess the usage of CUA in oral health interventions, to evaluate the methods used and to critically appraise the reporting quality of relevant studies. This review will facilitate better use of CUA in dentistry and help to improve decision-making.

2.1.3 Methods

The review followed the Preferred Reporting System for Systematic Reviews and Meta-Analysis (PRISMA) strategy (Figure 1) (85). The protocol was registered in the international database of prospectively registered systematic reviews in health and social care (PROSPERO), Centre for Reviews and Dissemination, University of York (No: CRD42016035530). The PICO for this review was: Population: people with oral health issues; Intervention/comparator: oral health interventions; and Outcome: Cost-utility analysis.

A comprehensive literature search was undertaken using multiple databases: MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL Plus) and Dentistry & Oral Sciences Source through EBSCOhost, Cochrane Database of Systematic Reviews (Issue 12 of 12, December 2016), and the Centre for Reviews and

Dissemination (University of York, United Kingdom). The search was conducted using the terms 'oral health' AND 'economic evaluation*' with appropriate truncation and adjacency settings (**Text box 1**). Our search was designed to capture all full economic evaluations related to oral health without time limitation first, then to limit studies to those with a utility based health outcome measure, and then to further limit the analysis to studies published after the year 2000 at the title and abstract screening phases, in order to ensure currency of the findings. Inclusion was limited to articles written in English and to human studies. In vitro and laboratory-based studies, letters, editorials, unpublished grey literature, guidelines, conference proceedings, case reports, methodology papers and literature reviews were excluded.

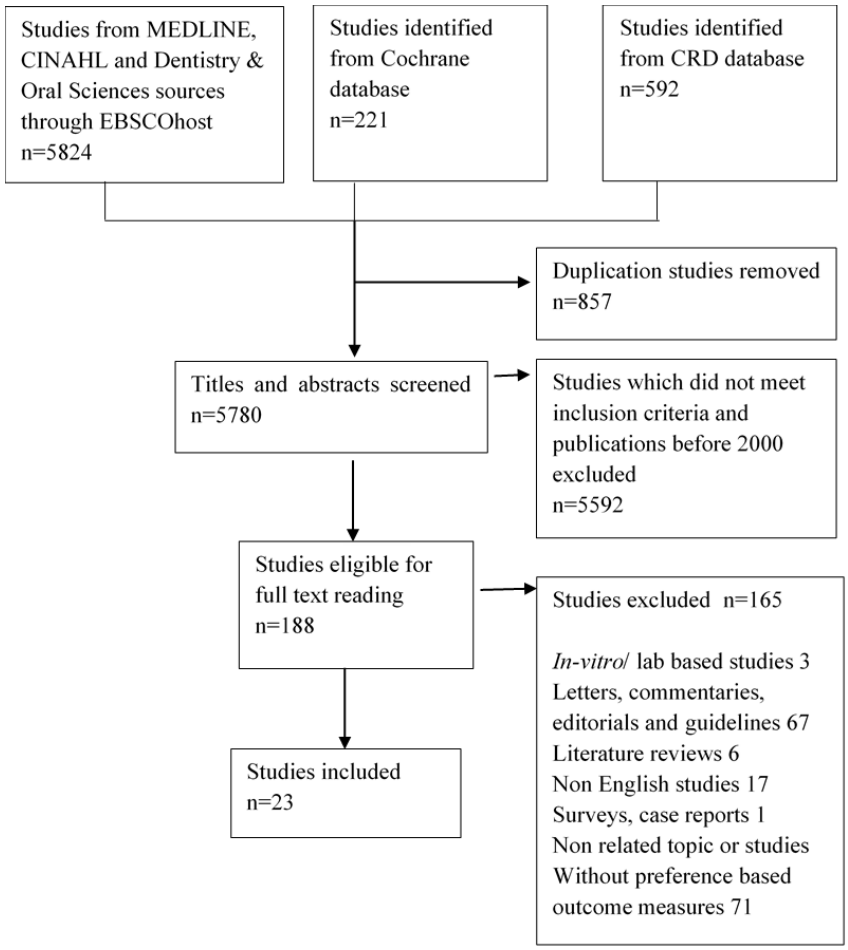

Figure 2.1: PRISMA flow diagram of the different phases of the systematic review

1. ‘Dentistry+’

2. ‘Caries preventive’ or ‘Caries Prevention’ or ‘dental’ or ‘tooth decay’ or carious or ‘periodontal’ or ‘dentistry’ or ‘oral cancer’ or ‘oral neoplas*’ or ‘children dental care’ or ‘Fluoride mouth rins*’ or ‘water fluoridation’ or ‘fissure sealant’ or ‘Oral Health’ or ‘oral hygiene’

3. ‘Health Care Costs+’ or ‘Health Expenditures+’ or ‘Economics, Dental+’

4. ‘economic evaluation*’ or ‘Cost Analysis’ or ‘Cost effectiveness’ or ‘Cost Benefit*’ or ‘cost minimization*’ or ‘cost-utility’

The search terms were combined by using the Boolean terms ‘AND’ ‘OR’ (#1 OR #2) AND (#3 OR #4) to achieve the final result.

Text Box 2.1: Search terms

Data were extracted from the selected articles using summary tables. These were also used for data synthesis. The reporting quality of the selected economic evaluations was assessed using the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) checklist (86). This provides guidelines for 24 items to be included when reporting economic evaluations. The selected articles were appraised against each item by two independent reviewers (RH and SK) and discrepancies were resolved by discussion. Each item in the CHEERS checklist was scored as having met the criteria in full (‘1’), not at all (‘0’) or not applicable (NA). When items partially met the criteria, they were scored as ‘0’: no partial scores were assigned to avoid introducing subjectivity. Study quality was expressed as a proportion of the items fully met for each article.

2.1.4 Results

Twenty-three articles were included in the systematic review (Figure 1). The number of published CUAs in oral health interventions has significantly (p=0.019) increased over time from four publications (17%) during the 2000-2005 period to 14 CUAs (61%) in 2011 to 2016 (**Table 2.1**). Most of the studies (n=7; 30%) were conducted in the USA (48, 87-92), followed (n=4; 17%) by the UK (81, 93-95). Three studies (13%) from each were conducted in Australia (51, 96, 97) and Netherlands (98-100) whereas only 2 studies (9%) were reported from the Asian region (101, 102) **(Table 2.1)**. Among the selected articles, the most frequent were on oral cancer (n=6; 26%) (88, 89, 92, 94, 98, 100), prosthetic management (n=6; 26%)(34, 49, 50, 93, 99, 102) and prevention of

dental caries (n=4; 17%) (48, 51, 96, 97). Detailed descriptions of the included studies are provided in **Table 2.2**.

The majority of the studies (n=15; 65%) used QALY as the summary health outcome measure. Disability-adjusted Life Years (DALY) (n=2; 9%), Quality-Adjusted Tooth-Years (QATY) (n=2; 9%), Quality-Adjusted Prosthesis Years (QAPY) (n=2; 9%) and Quality-of-Tooth-Years (QLTY) (n=1; 4%) were other reported outcome measures **(Table 2.1)**. A study assessing the cost-utility of periodontal maintenance therapy used net tooth years and prosthetic adjusted net tooth years to measure the treatment outcome (103). Of the 15 studies which used QALY as the outcome measure, eight studies (87-92, 94, 98) obtained utility values provided in the literature to calculate the outcome measure, whereas one study (81) obtained utility values from the patients using the time trade-off method. The EuroQol-EQ-5D-3L questionnaire was used by four studies (93, 95, 100, 101) and one study (97) used the Child Health Utility 9D (CHU9D) questionnaire with utility values based on proxy population weights. One study (99) converted patient's SF-36 scores to QALYs using the SF-6D excel scoring programme **(Table 2.3)**. The two studies (51, 96) which used DALY, incorporated disability weights from the literature to calculate DALYs. Studies which used QAPY (50, 102) and QLTY (49) obtained utility values directly from the patients through using a standard gamble, willingness-to-pay experiment or a visual analogue scale. Of the two studies which used QATY, one (48) obtained utility values provided in the literature whereas the other (34) obtained utility values from a patient's survey.

Of the 23 studies, perspectives included societal measures (48, 87-89, 97, 99, 101), the health care systems (34, 51, 93-95, 100), and patient or third-party payer (50, 90-92, 95, 99), whereas 6 studies did not explicitly state the perspective used in their analysis (49, 81, 96, 98, 102, 103) **(Table 2.1)**. The costs of interventions included in the analyses showed marked differences in their scope and categorisation and were reported in substantially differing forms such as direct costs, indirect costs, recurrent and capital costs, professional charges, treatment and total cost. All but 2 studies (91%) clearly mentioned the data sources and publications from which the cost data were extracted. The most frequent currency presented was US Dollars (n=9; 39%). Seven studies (49, 81, 89, 90, 95, 98, 99) did not report the base year and/or conversion of the currencies used **(Table 2.4)**. Most of the studies (n=12) used 3% to discount both costs and benefits. Three studies did not report the discount rate (49, 93, 98) **(Table 2.1)**. Seven studies out of 23 included the discount rate in the sensitivity analysis, of which four (50, 87, 91, 96)

identified the discount rate as an important factor impacting on the incremental cost-effectiveness ratio (ICER)

Table 2.1: Characteristics of the cost-utility analysis in oral health interventions (n, %)

Characteristic	Studies (n=23)
Year of publication	
2000-2005	4 (17%)
2006-2010	5 (22%)
2011-2015	14 (61%)
p- value	0.019*
Study country	
United States	7 (30%)
United Kingdom	4 (17%)
Australia	3 (13%)
Netherlands	3 (13%)
Asian Countries	2 (9%)
Other	4 (17%)
Dental condition	
Oral cancer	6 (26%)
Dental prostheses	6 (26%)
Dental caries prevention	4 (17%)
Periodontal diseases	3 (13%)
Pre-dental antibiotic prophylaxis	2 (9%)
Dento-facial anomalies	1 (4%)
Dental service delivery	1 (4%)
Type of of the intervention	
Treatment procedures	9 (39%)
Prevention intervention	4 (17%)
Screening test or programmes	5 (22%)
Dental material or pharmaceuticals	3 (13%)
Programme evaluation	2 (9%)
Health outcome	
QALY	15 (65%)
DALY	2 (9%)
QATY	2 (9%)
QAPY	2 (9%)
QLTY	1 (4%)
Net tooth years and prosthetic-adjusted net tooth years	1 (4%)
Study perspective**	
Health sector	6 (26%)
Societal	7 (30%)
Patient or third-party payer	6 (26%)
Not explicitly stated	6 (26%)
Discounting	
3%	12 (52%)
5%	3 (13%)
Other	4 (17%)
Justify why not discounted	1 (4%)
Not mentioned	3 (13%)

Reporting of cost-effectiveness ratio	
Incremental cost-effectiveness ratio (ICER) reported	18 (78%)
Not reported but provide values	4 (17%)
Other	1 (4%)
Presentation of	
Time horizon	22 (96%)
Intervention and comparator	23 (100%)
Sensitivity analysis used	22 (96%)

**p value Z test for proportions was used.* ***Two studies used more than one perspective*

QALY: Quality-adjusted life years; DALY: Disability- adjusted life years; QATY: Quality-adjusted tooth years: QAPY: Quality- adjusted prosthesis years; QLTY: Quality- of- tooth years

Table 2.2: Details of the studies included

Author, year	Dental condition	Type of study	Country	Setting	Study population/ sample	Intervention/experimental group	Comparator	Health outcome	Time frame/ analytical horizon
Acevedo et al, 2016	Oral cancer	Markov model	USA	Clinic	Patients with early stage clinically node negative oral cancer	Elective neck dissection at the time of primary tumour resection	Watchful waiting	1. Quality-adjusted life years (QALY)	30-year time horizon and 1-year cycle length
Agha et al, 2005	Pre-dental antibiotic prophylaxis	Cohort decision model and 4-state Markov process	USA	Clinic	Ten million Hypothetical cohort of 40 years US adults either a high or moderate risk for developing endocarditis	Seven prophylaxis strategies and no prophylaxis 1. Oral Amoxicillin 2g, 1h before the procedure (BTP) 2. Oral Clarithromycin 500mg, 1 h BTP 3. Oral Clindamycin 600 mg, 1 h BTP 4. Oral Cephalexin 2 gm, 1 h BTP 5. Intravenous (IV) or intramuscular (IM) Ampicillin 2 gm, 30min BTP; 6. IV or IM Cefazolin 1 gm, 30 min BTP; 7. IV Clindamycin 600 mg, 30 min BTP 8. No antibiotic prophylaxis		1. Cases of endocarditis prevented 2. Lives saved 3. QALY	1-time need for a prophylactic antibiotic prior to a dental procedure 55 years Markov cycle length 1 year
Balevi et al, 2007	Prosthetic management	Decision tree analysis	Canada	School	Middle-aged adult patients convenience sample of school teachers n = 40	4 Treatment options: conventional crown (CC), single tooth implant (STI), conventional dental bridge (CDB) and partial removable denture (RPD)		1. Patient preferences, standard gamble and willingness to pay utilities 2. QLTY	5-years
Bhuridej et al, 2007	Dental caries	Retrospective cohort	USA	Clinic	6 years old Medicaid enrolled children n = 2132	Children with sealed first permanent molars	Children with non-sealed first permanent molars	1. Caries experience from the claim data 2. QATY	4 years
Chun et al, 2016	Prosthetic management	Markov model	Korea	Clinic	Not clearly stated	Implant and conventional fixed dental prosthesis		Quality-adjusted prosthesis year (QAPY)	50 years

Author, year	Dental condition	Type of study	Country	Setting	Study population/ sample	Intervention/experimental group	Comparator	Health outcome	Time frame/ analytical horizon
Ciketic et al, 2010	Dental caries	Cohort life table modelling (risk type interventio n model)	Australia	Comm unity	All children under 15 years of age within South East Queensland region n = initial cohort of 36,322 newborns	Fluoridation of drinking water supplies for South East Queensland	Current practice of not fluoridating the water supplies of South East Queensland Population	DALY	100 years of life table
Cobiac & Vos, 2012	Dental caries	Life table modelling	Australia	Comm unity	Australia's population	Population health impacts and cost-effectiveness of extending public water fluoridation: 1. To all Australian communities with a population of at least 1000 people (89% coverage) 2. To all communities in Australia (100% coverage)	To baseline water fluoridation coverage of 69% in 2003	1. Number of averted caries in children and adults 2. DALY	Average 15-year lifespan of a treatment plant
Cunningha m et al, 2003	Dento-facial anomalies	Not mentioned	UK	Orthod ontic/su rgical unit	Patients received orthognathic treatment for dentofacial disharmony n = 21	Patients received orthognathic treatment	No treatment approach (only based on assumptions)	1. Patients utility values 2. QALY	At the start and end of treatment and three times during treatment
De Almeida et al, 2016	Oral cancer	Decision tree model and Markov model	USA	Hospita l	Hypothetical patients with early oropharyngeal cancer	patients with oropharyngeal cancer treated with transoral robotic surgery	Patients with oropharyngeal cancer treated with(chemo) radiotherapy	QALY	3 months' cycle Markov model 10 year
Dedhia et al, 2011	Oral cancer	Cohort state-transition	USA	commu nity	Simulated cohort of males more than 40 years regularly	Yearly community screening for all high-risk males over age 40 years	No yearly community screening	1. Prevalence of precancer	1-year cycle 40 years

Author, year	Dental condition	Type of study	Country	Setting	Study population/ sample	Intervention/experimental group	Comparator	Health outcome	Time frame/ analytical horizon
		Markov model			using tobacco and/or alcohol			2. Yearly death rate from oral cancer 3. QALY	
Fardal & Grytten, 2014	Periodontal diseases	Not mentioned	Norway	Clinic	Patients with periodontal diseases referred to the Periodontology specialist in South-West Norway	Patients fully complied with the periodontal maintenance programme for a minimum of 16 years n = 80	Patients non - compliant with the periodontal maintenance programme for at least 3 years n = 25	1. Patients' expectations and changes in oral health 2. Net tooth years and prosthetic adjusted net tooth years	20 years
Higashi et al, 2002	Periodontal diseases	Cohort decision analytic and Markov models	USA	Clinic	Hypothetical cohort of Caucasian males and females (age 35) who were referred to a periodontist with mild periodontitis	Sub cohorts based on the 1. Treatment or no treatment 2. Smoker or non-smoker 3. Possessing an interleukin -1 IL-1 (+) or IL-1(-) genotype		QALY	30 years
Hulme et al, 2014	Prosthetic management	Double blind, controlled, crossover clinical trial	UK	Clinic	Edentulous patients n = 71	Patients got silicone impressions (n=34) and alginate impressions (n = 37) for complete dentures in period 1 and reverse the impression material in period 2		1.Oral health-related QOL using OHIP-EDENT 2. QALY	Time until the second denture adjustment
Hulme et al, 2016	Dental care provision	Non-randomised comparative study	UK	Clinic	New patients recruited at six dental practices three incentive practices and three traditional practices	Blended/incentive-driven model of dental service provision with the traditional dental service provision		1. Oral health-related QOL using OHIP 14 2. QALY	2 years

Author, year	Dental condition	Type of study	Country	Setting	Study population/ sample	Intervention/experimental group	Comparator	Health outcome	Time frame/ analytical horizon
Jensen et al, 2016	Prosthetic management	clinical trial	Netherlands	Clinic	Patients with an edentulous maxilla and a bilateral free-ending in the mandible n = 30	Conventional removable partial dentures (RPDs) and implant-supported RPDs (ISRPDs)		1. Oral health related quality of life using OHIP 2. Chewing ability 3. QALY	3 months each in both arms
Koh et al, 2015	Dental caries prevention	Cohort Markov model	Australia	Home based and clinic	Children age 6 months to 6 years	1. Home visit intervention for children (n = 188) by oral health therapists 2. Telephone intervention for children (n = 58) dental care instructions through telephone calls by oral health therapists	Usual care group (n = 40) children registered in the public school dental programme	1. Number of carious teeth prevented 2. QALY	5 1/2 years 6 months to 6 years
Mohd-Dom et al, 2014	Periodontal diseases	multicentre, time motion, prospective study	Malaysia	Clinic	Patients with moderate to severe periodontitis	Patients attending specialist periodontal clinics, Malaysia n = 165	Hypothetical group of patients attend biannual dental visits only for regular dental check-up and scaling	1. Clinical attachment levels of the periodontium 2. QALY	1 year
Skaar et al, 2015	Pre-dental antibiotic prophylaxis	Cohort Markov decision model	USA	Clinic	Hypothetical cohort of 65-year-old patients undergone total hip arthroplasty (THA)	1. Having antibiotic prophylaxis for the first 2 years after undergoing THA 2. Having antibiotic prophylaxis for the patient's lifetime	Patients with THA having no antibiotic prophylaxis before dental visits	QALY	Over the simulated lifetimes of hypothetical cohort of patients with THA. 6months cycles
Speight et al, 2006	Oral cancer	Decision-analytic model	UK	Primary care	Hypothetical population over the age of 40 years	Eight hypothetical screening programmes 1. No screen 2. Invitational screen – general medical practice (GMP)		QALY	60-year lifetime

Author, year	Dental condition	Type of study	Country	Setting	Study population/ sample	Intervention/experimental group	Comparator	Health outcome	Time frame/ analytical horizon
						3. Invitational screen – general dental practice (GDP) 4. Opportunistic screen – GMP 5. Opportunistic screen – GDP 6. Opportunistic high-risk screen –GMP 7. Opportunistic high-risk screen – GDP 8. Invitational screen – specialist			
Van der Linden, 2016	Oral cancer	Decision tree and Markov model	Netherlands	Clinic	Patients with T1–T2 oral cancer	1.USgFNAC (ultrasound guided fine needle aspiration cytology) 2. SLNB (sentinel lymph node biopsy) 3. USgFNAC and, if negative, SLNB 4. END (elective neck dissection)		1.QALY	5 years, 10 years or life time
Van der Meij et al, 2002	Oral cancer	Decision model	Netherland	Clinic	Hypothetical cohort patients with oral lichen planus (OLP) n = 10,000,000	1. Screening programme for oral cancer in OLP patients 2. Screening by a specialist 3. Screening by a dentist	No screening	1.Equivalent lives saved (ELS) 2.QALY	For a period of 1 year
Zitzmann et al, 2006	Prosthetic management	Clinical trial	Switzerland	Clinic	Edentulous patients n = 60	Edentulous patients treated with implant retained over denture (n = 20) and implant supported over denture (n = 20)	Edentulous patients treated with complete denture (n = 20)	1. Patients' perception questionnaire and visual analogue scale 2. QAPY	Up to 10 years
Zitzmann et al, 2013	Prosthetic management	Prospective preference trial probabilistic model	Switzerland	Clinics	Patients need tooth replacement in the maxillary anterior region, including first premolars	Patients with implant supported single crowns ISCs (n = 15)	Patients with fixed dental prosthesis FDPs (n = 11)	1. Patients' perception questionnaire and visual analogue scale 2. QATY	3, 5 and 10 years

QALY: Quality-adjusted life years; DALY: Disability-adjusted life years; QATY: Quality-adjusted tooth years; QAPY: Quality-adjusted prosthesis years; QLTY: Quality-of-tooth year

Table 2.3: Preference-based outcome used in the CUA in oral health interventions

Study	Preference-based outcome type	Measurement of Preference-based outcome	Method/instrument used to obtain utility values
Acevedo et al, 2016	QALY	Model estimate	Utility values from the literature
Agha et al, 2005	QALY	Model estimate QALYs calculated based on the utility value for each health state and the number of years spent in that health state. Adjustments were done for decrements in quality of life and short term health states were performed.	Utility value for each long term health state derived from literature (Fryback D. et al, 1993).
Balevi et al, 2007	QLTY	Model estimate QLTY = Expected utility values × [years of survival]	Utility values for each treatment method calculated using weighted average of standard gamble utilities and willingness to pay utilities from sample of school teachers, Canada as a part of the same study
Bhuridej et al, 2007	QATY	Total average QATYs of four intervals = Σ TiUi / N Ti = value of tooth-year of each tooth in each interval Ui = value of health state of teeth in the interval N = total number of at-risk tooth-years in each interval	Utility values from literature (Fyffe and Kay, 1992)
Chun et al, 2016	QAPY	Patients survey	Calculated from the patients willingness to pay per 1 QAPY for the implant compared to the CFDP over the years after treatment
Ciketic et al,2010	DALY	Model estimate- DALY calculated using the existing data on caries episodes and adjusted for the age cohorts of 2,7,12 and gender.	0.057 (Begg S. et al, 2007) incorporated
Cobiac & Vos, 2012	DALY	Model estimate-DALY calculated based on time spent with the caries symptoms from clinic data and proportion of people with caries who are symptomatic from literature and apply disability weight of 0.057.	Disability weight 0.057 (Australian burden of disease disability weight for symptomatic caries derived using EQ 5D+ disability weight regression model)
Cunningham et al, 2003	QALY	QALYs gained = (change in patient's utility between the start and the end of treatment) X patient's future life expectancy. Any short term QALY lost due to process of treatment were subtracted.	Patients utility values using time trade off method
De Almeida et al, 2016	QALY	Model estimate QALY for each treatment= utility weight X time spent in each health state and weighted according to the probability of each health state	Health state utilities for all relevant treatment, complication, remission and recurrences using standard gamble method derived from Adelstein DJ et al, 2012
Dedhia et al, 2011	QALY	Model estimate	Age specific utility data from the data developed by National Centre for Health Statistics (Gold MR et al, 1998) and utility data for precancer and cancer (Downer et al, 1997)
Fardal & Grytten, 2014	Net tooth years and prosthetic adjusted net tooth years	Mean and SD for Anxiety and Discomfort- last elements of the EQ-5D: measured using Visual Analogue Scales (VAS)	Reference values from patients during the initial periodontal treatment

Higashi et al, 2002	QALY	QALY=life expectancy in the certain health states x utility for the health state	Utility for mild periodontitis and severe periodontitis from literature reviews and expert clinical opinion
Hulme et al, 2014	QALY	EQ-5D-3L	Utility values - UK tariff value
Hulme et al, 2016	QALY	EQ-5D-3L and mapping of patient data from OHIP-14	Utility values for EQ-5D - UK tariff value OHIP-14 scores mapped using regression techniques to the baseline EQ-5D-3L scores
Jensen et al, 2016	QALY	Patients SF-36 scores were converted in to QALY	Scores converted using SF-6D excel scoring programme of the University of Sheffield
Koh et al, 2015	QALY	Model estimate	Utilities for caries and healthy from utility survey using CHU9D (parents visited community dental clinics with children younger than 5 years)
Mohd-Dom et al, 2014	QALY	EQ-5D-3L	A proxy Thai population weights
Skaar et al, 2015	QALY	Model estimate QALY estimated based on the assigned utility weight for each period according to the health event experienced	Health state utilities for primary total hip arthroplasty, aseptic revision and septic revision from literature
Speight et al, 2006	QALY	Model estimate	Health state utility scores for oral precancer and cancer from literature (study based on a convenience sample of UK general public n=100)
Van der Linden, 2016	QALY	EQ5D	Utility values from a clinical trial data
Van der Meij et al, 2002	QALY	Calculated based on the health state utilities for oral cancer and life expectancy	Health state utilities for oral cancer assessed by Downer et al, 1997
Zitzmann et al, 2006	QAPY	Calculated by adjustment of the duration of a dental state by the patient's preference for that state	Utility values based on the patient preferences using visual analogue scale scores before and after treatment
Zitzmann et al, 2013	QATY	QATY estimated by considering the type of reconstruction used to replace the missing tooth and its effect on the adjacent teeth.	Utility values based on the visual analogue scale scores before and after treatment

QALY: Quality-adjusted life years; DALY: Disability- adjusted life years; QATY: Quality-adjusted tooth years: QAPY: Quality- adjusted prosthesis years; QLTY: Quality- of-tooth year

Table 2.4: Details of the economic data and related analysis

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
Acevedo et al, 2016	Third-party payer's	Costs of all aspects of treatment and follow-up	1. Cost of treatment and cost of end-of-life care- from literature 2. Cost of chemotherapy and radiation- from wholesale drug pricing and literature	US dollars	2015	3% utilities and costs	Yes One-way and probabilistic sensitivity analysis	TreeAge
Agha et al, 2005	Societal	Direct medical cost for antibiotics Hospital cost Indirect cost of patient or caregiver time lost	1. Cost for antibiotic - Drug Topics Red Book 2000 and dispensing cost 2. Hospital costs - estimated using the Medicare cost from the nationwide inpatient sample for specific Medicare diagnosis-related groups (DRGs) in 1997 and prior estimates 3. Indirect cost of patient or caregiver time lost- report by the Bureau of Labor Statistics	US dollars	2003	3% benefits and costs	Yes One-way sensitivity analyses	TreeAge and Microsoft Excel
Balevi et al, 2007	Not mentioned clearly	Cost of each treatment using the direct out of pocket costs that the patient or insurer pays to the dentist	Sources of the cost data not clearly mentioned	Canadian dollars ($CDN)	NM	NM	Yes	SPSS and TreeAge
Bhuridej et al, 2007	Societal	Total cost of each yearly interval for each tooth = the total number of each type of treatment x its fee	1. Total number and type of treatment - from eligibility and dental claims files for children enrolled in the Iowa Medicaid programme from 1996 through 2000 2. Fee- American Dental Association (ADA) 2001 Survey of Dentists Fees	US dollars 2001 value	Child's sixth birthday	3% costs and outcome	Yes One-way sensitivity analysis	SAS
Chun et al, 2016	Not mentioned clearly	Direct and indirect cost.	The data was investigated from Statistics Korea.	Korean won	2013	3%	Yes	TreeAge
Ciketic et al, 2010	Not mentioned clearly	Costs of fluoridation Costs of dental treatment	1. Costs of fluoridation-from a scoping report on fluoridation, the Queensland Government in 2002 (cost of a fluoridation plant + the annual cost of consumables +annual maintenance costs)	Australian dollars (AUD)	2003	3%	Yes One way and two way	Risk 4.5

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
			2. Costs of dental treatment – data from the Australian Dental Association					
Cobiac & Vos, 2012	Health sector	Costs of public water fluoridation Costs of caries treatment	1. Costs of public water fluoridation (Urban/rural) – literature 2. Costs of caries treatment – from the standard Australian Dental Association costs for service	AUD	2003	3%	Yes	R
Cunningham et al, 2003	Not mentioned clearly	Costs incurred during treatment	Key unit cost and time for treatment – based on standard NHS salary scales and patient data	British pounds	NM	6% cost 2% benefits	Yes	NM
De Almeida et al, 2016	Societal	Institutional cost Professional fees Direct patient cost	1. Institutional costs – mean costs from 88 patients with T1 and T2 oropharyngeal cancers treated with TORS or (chemo) radiotherapy in Mount Sinai Hospital, New York from 2007 to 2010. 2. Professional fees – based on Medicare reimbursement for Current Procedural Terminology (CPT) codes billed from the above cohort of patients 3. Loss of wages – duration of lost work (from disability claims or from expert estimates) and the average wage and employment rate for the cohort (from the Bureau of Labour and Statistics). 4. Out-of-pocket costs – literature 5. Costs for surveillance – Medicare reimbursement for routine surveillance examinations and investigations	US dollars	Cost inflated to present day value	3% Cost and benefit	Yes Deterministic one-way, two-way and probabilistic sensitivity analysis	TreeAge
Dedhia et al, 2011	Societal	Costs for outpatient evaluation and office biopsies Cost of preoperative investigations	1. Costs for outpatient evaluation and office biopsies and Professional surgical fees – Medicare payment to Pennsylvania Area 99 based on 2009 Current Procedural Terminology Codes	US dollars	NM	3%Cost and utilities	Yes One-way sensitivity analysis	TreeAge

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
		Hospital costs for inpatient treatment Professional surgical fees	2. Cost of preoperative investigations – Federal Register, Volume 71, No. 226, November 24, 2006 3. Hospital costs for inpatient treatment – 2007 median cost, Agency for Healthcare Research and Quality's (AHRQ) Healthcare Cost and Utilization Project 4. Chemotherapy costs – published report for outpatient chemotherapy for advanced head and neck cancer 5. Radiation therapy – 2004 Medicare reimbursement for intensity-modulated radiation therapy (IMRT) 6. Total Stage I/II and stage III/IV Treatment costs year 1 7. WTP threshold – Braithwaite RS et al, 2008					
Fardal & Grytten, 2014	Not mentioned clearly	Cost of treatments	1. Cost of treatments – periodontal treatment and maintenance cost based on an hourly rate of non-surgical and surgical fees 2. Prosthetic replacement costs – based on a three-unit bridge or a single implant replacement at €2500 from literature	€ Euro	2013	5%	No	NM
Higashi et al, 2002	Third party payers	Direct medical cost data for the 4 health states Cost of the genetic test	1. Direct medical cost – Washington dental services reimbursement rates 2. Cost of the genetic test from the manufacturer	US dollars	NM	3%	Yes Best case/ worst case and one-way	NM
Hulme et al, 2014	Health care sector	Cost of resources used to construct the dentures	From literature, pay circulars and based on cost of materials	British pounds	2012	NM	Yes	STATA 12

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
		Costs of adjustments made for both dentures and other health service Costs resulting from problem with the dentures (ex.GP visits)						
Hulme et al, 2016	Commissioner and health care provider	Payment for units of dental activity (UDAs) Costs to the dental provider	1.NHS England financial value of UDAs 2. Material costs from the Kent Express Catalogue and MGill price list 3. Staff cost from pay circular and NHS agenda for change pay scales	British pounds	NM	3.5%	Yes	STATA and Excel
Jensen et al, 2016	Societal and payer perspective	Opportunity cost of the procedures Healthcare costs	1. Opportunity cost – Dutch costing manual and the standard salary scales of the Collective Labour Agreement (CAO) 2. Healthcare costs – market prices using tariffs for the Dutch situation	Euro	NM	Justify why not discounted	Bootstrapping	Excel
Koh et al, 2015	Societal	Programme cost Treatment costs Indirect costs	1. Programme cost per person – Programme data 2. Treatment (unit costs) – Hospital GA data, ADA schedule of fees and PBS code 3302T/3348F 3. Indirect costs – Utility Survey (part of the study) data	US dollars	2014 Converted from AUD 2013	5% costs and effects	Yes One way and probabilistic	TreeAge
Mohd-Dom et al, 2014	Societal	Capital building/asset cost Recurrent cost Patients cost	1. Capital step down/activity-based costing 2. Recurrent – step-down/activity-based costing 3. Patients cost – patient's diary using human capital approach. Sources of cost data reported.	Malaysian ringgit (MYR)	2012	5% cost	Yes One-way scenario-based	SPSS

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
							sensitivity analysis	
Skaar et al, 2015	Payers	Cost for aseptic revision Cost for Septic revision Cost for Dental Visit Cost for nonfatal adverse events Cost for fatal adverse events	1. Cost for aseptic revision – national average Medicare reimbursements and Health Care Utilization Project 2. Cost for septic revision – national average Medicare reimbursements and Health Care Utilization Project 3. Cost for Dental Visit – medical expenditure panel survey 4. Cost for Nonfatal Adverse Events-national average Medicare reimbursements and RED BOOK, Thomson Reuters 5. Cost for Fatal Adverse Events – national average Medicare reimbursements and RED BOOK, Thomson Reuters	US dollars	2013	3% Cost and QALY	Yes	TreeAge
Speight et al, 2006	Health care	Health state costs Unit costs assigned to the screening programmes	Cost data from a case note review at two large hospitals: University College London Hospitals (Maxillofacial Unit) and Barnet and Chase Farm District General Hospital. (Department of Maxillofacial Surgery), published costs, systematic review and by expert opinion using the Trial Roulette approach	British pounds	2002-2003	3.5% Cost and benefits	Yes	Microsoft Excel
Van der Linden et al, 2016	Health care	Direct medical costs	From the Dutch cost manual 2015, VU University Medical Centre or Dutch tariffs	Euro	2015	4% costs and 1.5% effects	Yes Probabilistic sensitivity analyses	NM
Van der Meij et al, 2002	Not mentioned clearly	Cost screening Cost biopsy by specialist	Cost calculations based on the estimates. Not mention the source	US$	NM	NM	Yes	NM

Author, year	Economic Perspective	Calculation of cost/ input data cost	Economic data source	Currency	Base year/ conversion	Discount rate	Sensitivity analysis	Software
		Cost per oral cancer treatment per case						
Zitzmann et al, 2006	Patients	Initial dental health care cost Treatment time and health care resource consumption Time costs of patients (travel and treatment time)	1. Initial dental health care cost – official national dental tariff structure 2. Treatment time and health care resource consumption – records from maintenance dental care during 3 years 3. Time costs of patients (travel and treatment time) – assuming opportunity cost of wage rate of unskilled worker	Swiss francs	2000 Swiss francs (CHF 100 = US$61)	3%	Yes	S-Plus
Zitzmann et al, 2013	Health care	Treatment costs Follow-up and maintenance costs, resource use	1. Treatment costs – Swiss tariff for dental treatments (surgical and reconstructive) and laboratory fees 2. Follow-up and maintenance costs, resource use – taking into account an annual recall examination with sensitivity testing, periodontal examination, periapical radiographs, occlusal control, professional cleaning and oral hygiene instructions	Swiss francs	2008 Swiss francs (CHF 100 = US$93)	3%	Yes	STATA and TreeAge

NM: Not mentioned; QALY: Quality-adjusted life years

Eighteen articles reported an ICER. Four studies (34, 81, 92, 98) did not use the term ICER; however, they provided values for the incremental cost and outcome gained **(Table 2.1).** Different threshold limits (by currency type and amount) have been used by authors to assess the cost-effectiveness of the interventions compared: a US$50,000 threshold was most commonly used **(Table 2.5)**. Of the 23 studies, 21 were able to identify the most cost-effective procedure among the different options compared **(Table 2.5)**. However, in six studies (89, 90, 94, 98, 99, 102), the authors reported uncertainties of their conclusion depending on the factors included in the analysis. Studies that assessed the cost-effectiveness of pre-dental antibiotic prophylaxis (87, 91) showed findings more compatible with the latest guidelines and recommendations (104) in this area. The studies that assessed the dental prosthetic material (93) and dental service model (95) were inconclusive about the cost-effectiveness of the intervention being studied **(Table 2.5)**.

The assessment of reporting quality of each study using the CHEERS checklist is provided in **Table 2.6**. The reporting quality of the included studies varied from 75 to 100% (median 92%) and there is no marked variation in reporting quality when compared with the year and country of publication. The items that least complied with the CHEERS were on characterising heterogeneity (item 21) (43% compliant among 23 articles) and statement of any conflict of interest (item 24) (48% compliant among 23 articles).

Table 2.5: Reporting of incremental cost effectiveness ratio of CUA in oral health interventions

Study	Dental condition	Intervention and comparator	Base case analysis	WTP used	Incremental cost effectiveness ratio (ICER)	Value of ICER	Conclusion of cost effectiveness by the authors
Acevedo et al, 2016	Oral cancer	Elective neck dissection vs watchful waiting at the time of primary tumour resection	Cost and QALY for each strategy were reported.	$100,000/ QALY	Not mentioned as ICER, but incremental cost and QALY compared with the watchful waiting reported.	Over a lifetime, elective neck dissection was $6,000 less cost and 0.42 QALYs more effective compared with watchful waiting.	'Addition of elective neck dissection is a cost-effective strategy for patients with early-stage oral cavity cancer'(92).
Agha et al, 2005	Predental antibiotic prophylaxis	7 pre-dental antibiotic prophylaxis regimens as in American Heart Association guidelines vs no prophylaxis	Endocarditis cases prevented, death and QALY per 10million patients were reported.	$50,000	Yes. Incremental cost-utility ratio (ICUR) calculated as cost per QALY gained in each antibiotic strategy compared with the no prophylaxis strategy.	Cost/ QALY gained Oral Clarithromycin $88,007 Oral Cephalexin $99,373 Oral Clindamycin$101142 Cefazolin (parenteral) $199430 Clindamycin -$411093 Oral Amoxicillin and Ampicillin (parenteral) - Not effective	'Pre-dental antibiotic prophylaxis is cost-effective only for patients with moderate or high risk of endocarditis. Clarithromycin should be the drug of choice and cephalexin is an alternative.'(87).
Balevi et al, 2007	Prosthetic management	4 treatment options for the management of endodontically abscessed tooth	Cost and QLTY (mentioned as QATY in the results section of this study) for each option for molars and incisors were reported.	NM	Not reported. Only calculated cost per QLTY in each option (Conventional crown- CC, single tooth implant- STI, conventional dental bridge- CDB and partial removable denture- RPD).	Cost-utility (CND$ per QLTY) Molar - CC 6.90 STI 9.51 CDB 7.73 RPD 3.85 Incisor - CC 5.27 STI 8.35 CDB 6.13 RPD 2.74 Not comparing each option or with no treatment approach.	'The removable partial denture (RPD) was the favoured treatment based on a cost-utility'(49)
Bhuridej et al, 2007	Dental caries prevention	Sealing vs non-sealing first permanent molars	Cost and QATY for each first permanent molar were reported.	NM	Yes. Cost per each QATY gained from sealing each molar compared with non-sealing and cost per 0.19 QATY ratio reported.	Maxillary 1st permanent molars #3= $439.6 #14=$327.43 Mandibular 1st permanent molars #19 =$193.0 #30= $202.3	'Sealants improved overall utility of first permanent molars after 4 years'(48).
Chun et al, 2016	Prosthetic management	Implant and conventional fixed dental prosthesis (CFDP)	Direct cost, total cost and QAPY from 0-50 years were reported.	10000 to 80000 Korean won/ QAPY	Yes	ICER for direct cost and total cost from 0- 50 years were reported.	'The CFDP was more cost-effective unless the WTP was more than 75,000 won at the 10th year after prosthodontic treatment. But changed to implant as time passed'(102).
Ciketic et al, 2010	Dental caries prevention	Fluoridation vs non-fluoridation of drinking water in South East	Mean total intervention cost, net costs, cost and DALY savings if fluoridation	$50,000 /DALY	Yes: Calculated from cost per DALY saved	ICER $64,127/DALY ICER without cost offset $3,608/ DALY	'Fluoridation remains a very cost-effective for reducing dental decay'(96).

Study	Dental condition	Intervention and comparator	Base case analysis	WTP used	Incremental cost effectiveness ratio (ICER)	Value of ICER	Conclusion of cost effectiveness by the authors
		Queensland, Australia.	implemented were reported.				
Cobiac & Vos, 2012	Dental caries prevention	Extending public water fluoridation vs baseline coverage of 69% in Australia	Treatment cost offsets, intervention cost, DALYs averted and probability of cost saving were reported.	$50,000/DALY	Yes Median cost effectiveness ratio	Communities >1000 people Median cost-effectiveness ratio ($/DALY) dominant All communities Median cost-effectiveness ratio ($/DALY) $92 000	'Extending coverage of fluoridation to all communities in Australia of at least 1000 people is cost effective'(51).
Cunningham et al, 2003	Dento-facial anomalies	Orthognathic treatment vs no treatment	Mean discounted cost and mean QALYs gained for combined group, single jaw and bimaxillary group were reported.	£825/ QALY	Not mentioned as ICER, but incremental cost for each additional QALY compared with the no treatment reported.	Incremental cost for each additional QALY compared with the no treatment: Combined group £561 Single jaw group £617 Bimaxillary group £546	'Orthognathic treatment is cost effective compared to no treatment'(81).
De Almeida et al, 2016	Oral cancer	Trans-oral robotic surgery vs (chemo)radiotherapy for the oropharyngeal cancer	Cost and QALY for each method were reported.	$50,000/ QALY	Yes Incremental cost-utility ratio calculated.	Trans-oral robotic surgery dominated with cost savings of $1366 and an increase of 0.25 QALY per patient treated.	'Trans-oral robotic surgery is cost effective to treat early oropharyngeal cancers'(88).
Dedhia et al, 2011	Oral cancer	community screening vs no screening for oral cancer in high-risk males	<u>Screen</u> Cost $640.20 QALY 15.467 <u>No-Screen</u> Cost $898.50 QALY 15.4257	$75,000/ QALY	Yes	No-screen is more expensive and less effective with incremental cost of $258 and incremental effectiveness of -0.0414 QALYs than screening. Screening is the dominant strategy.	'A community-based screening program targeting high-risk males is likely to be cost-effective'(89).
Fardal & Grytten, 2014	Periodontal diseases	Patients fully complied with the periodontal maintenance programme vs patients non–compliant/ partial compliant	<u>Compliance group</u> Total cost €5842 Tooth loss years 17.7 <u>Partial compliant group</u> Total cost €6395 Tooth loss years 17.7x2.55	NM	Yes ICER reported as cost of buying an extra tooth year	Cost of buying one tooth year ICER €6395- €5842/ 27.44= €20.2	'The cost of buying an extra tooth year was €20.2'(103).
Higashi et al, 2002	Periodontal diseases	Patient tested interleukin 1 (IL-1) genetic test for	Test strategy showed additional costs of $147,114	NM	Yes Cost per QALY gained	Test strategy resulted $32633 per QALY gained compared to no test strategy.	'Genetic testing of mild Periodontitis patients is

Study	Dental condition	Intervention and comparator	Base case analysis	WTP used	Incremental cost effectiveness ratio (ICER)	Value of ICER	Conclusion of cost effectiveness by the authors
		periodontal diseases vs not tested	per 1000 patients over a 30 years', reduction of 0.61% absolute severe periodontitis cases and 0.45% absolute increase of QALY				Cost-effective if treatment effectiveness is independent of IL-1 genotype. Additional data are needed for the more accurate analysis'(90).
Hulme et al, 2014	Prosthetic management	Silicone vs alginate impressions for complete dentures	Silicone Cost £388.57 QALY 0.05 OHIP 28.64 Alginate Cost £363.18 QALY 0.06 OHIP 36.17	£20,000/ QALY	Yes Incremental cost per QALY and OHIP-EDENT point	Cost/QALY -5.758 (alginate dominates) QALY gain negligible between groups Cost/change one OHIP point £3.41 compared to alginate	'Silicone was more costly and negligible QALY gain, but improved OHIP-DENT. Difficult to make robust Claims about the comparative cost-effectiveness'(93).
Hulme et al, 2016	Dental care provision	Blended/incentive-driven model of dental service provision with the traditional dental service provision	Mean cost, QALY and OHIP-14 for commissioner's and health provider perspectives were reported.	£20,000/ QALY	Yes cost per QALY and OHIP-14 point	Commissioner's perspective ICER (£199.22 /OHIP-14 point) £/QALY incentive dominated Service provider's perspective –104.03£/OHIP-14 incentive dominates 122,089.48£/QALY	'The economic analysis showed the INCENTIVE arm attract a higher cost for the dental commissioner but to be financially attractive for the service provider'(95).
Jensen et al, 2016	Prosthetic management	Removable partial dentures (RPDS) and implant-supported removable partial dentures (ISRPDS)	RPD Opportunity cost €985 Tariff cost €850 QALY 0.8 OHIP 41.1 ISRPD Opportunity €2475 Tariff cost €2610 QALY 0.8 OHIP 22.4	NM	Yes incremental cost per QALY/OHIP/Mixed Ability Index (MAI) point	Cost/QALY negligible Opportunity Cost/OHIP €80 Opportunity Cost/MAI €786 Tariff cost/OHIP €94 Tariff cost/MAI €921	'Depending on the choice of outcome measure and monetary threshold, ISRPD is cost-effective when payers are willing to pay more than €80 per OHIP point gained'(99).
Koh et al, 2015	Dental caries prevention	Home-visit intervention conducted by oral health therapists vs telephone-intervention and usual	For 100 children over 5.5 years, Usual care Cost $348903 Caries prevented 258 QALY 540 Telephone	NM	Yes Cost per number of carious lesions prevented and QALY gained	Home visits and telephone save $167032 and $144709, prevent 113 and 100 carious teeth and gain 7 and 6 QALY compared to no intervention.	'Both the home visits and telephone-based community interventions conducted by oral health therapists were highly cost-effective than no intervention'(97).

Study	Dental condition	Intervention and comparator	Base case analysis	WTP used	Incremental cost effectiveness ratio (ICER)	Value of ICER	Conclusion of cost effectiveness by the authors
		care in preventing early childhood caries	Cost $204193 Caries prevented 158 QALY 546 Home visits Cost $181870 Caries prevented 145 QALY 547				
Mohd-Dom et al, 2014	Periodontal diseases	Specialist periodontal programme vs biannual dental visits at the general dental practice	Specialist treatment Cost/patient 2524MYR QALY gain 3.8 CAL (mm) 0.3 Biannual visits Cost /patient 810 MYR QALY gain 3.8 CAL (mm) 0.3	NM	Yes Cost per additional QALY gained and per millimetre clinical attachment level gained	Specialist treatment 5713MYR per additional QALY 451 MYR per millimetre CAL gained compared to hypothetical biannual visits	'Specialist periodontal care for patients with periodontitis is very cost-effective compared to biannual dental treatment'(101).
Skaar et al, 2015	Pre dental antibiotic prophylaxis	Pre dental antibiotic prophylaxis for the first 2 years /life time after undergoing total hip arthroplasty vs no antibiotic prophylaxis	No prophylaxis Cost US$ 11,909 QALY 12.3745 For 2 years Cost US$11,941 QALY 12.3741 For life time Cost US$12,209 QALY 12.3734	$50,000 to $100,000 per QALY	Yes Incremental cost per QALY gained	For 2 years Incremental cost 33 US$ Incremental QALY -0.0004 For life time Incremental cost 301 US$ Incremental QALY -0.0011 Compared to the no prophylaxis, both are more costly and less effective	'No-antibiotic prophylaxis strategy was cost-effective for dental patients who had undergone total hip arthroplasty'(91).
Speight et al, 2006	Oral cancer	Screening for oral cancer in primary care	Cost and QALY for each screen strategy were reported.	£20,000-£30000 per QALY	Yes Cost per additional QALY	Opportunistic high-risk screening General Dental Practice (OHS-GDP) £22,850/ QALY compared with no screening. Opportunistic high-risk screening General Practitioner (OHS-GP) £23,728/ QALY compared with OHS–GDP. Opportunistic screening-GP £25,961/ QALY compared with the OHS-GP.	'Opportunistic high-risk screening, particularly in general dental practice, may be cost- effective. There is considerable uncertainty in the parameters used in the model'(94).
Van der Linden et al, 2016	Oral cancer	4 strategies for the detection of occult lymph node	Cost and QALY for each strategy on 5 years, 10 years and	€28.000/ QALY	Yes, Incremental cost-utility ratio	5- or 10-years. Sentinel lymph node biopsy (SLNB) resulted highest additional QALY (0.12	'SLNB is the preferred strategy in a 5- or 10-year time horizon. From a lifetime

Study	Dental condition	Intervention and comparator	Base case analysis	WTP used	Incremental cost effectiveness ratio (ICER)	Value of ICER	Conclusion of cost effectiveness by the authors
		metastases in oral cancer	lifetime horizon were reported.			and 0.26) for the smallest additional costs (€56 and €74) and in lifetime, Elective neck dissection (END) results highest additional QALYs (0.55) for an additional €1.626 compared to ultrasound guided fine needle aspiration cytology.	horizon, END may be preferred'(100).
Van der Meij et al, 2002	Oral cancer	Screening for the oral cancer in patients with oral lichen planus	Screening compared to no screening Extra cost $ 1265229 Health gain 592 QALY Equivalent of lives saved 23.68	NM	Cost per additional QALY not reported. But can be calculated from the values provided.	Cost per one ELS saved= 53430$ Cost effectiveness of screening by dentist compared to the no screening not reported. But can be calculated from the figure.	'Screening for oral cancer in OLP patients seems attractive. However varying the variables in the decision model has significant impact on the final cost and effectiveness'(98).
Zitzmann et al, 2006	Prosthetic management	Implant-supported overdenture (ISD), implant-retained (IRD) and complete dentures for the edentulous patients	3 years ,3% discount Complete denture Cost CHF 3672 QAPY 0.82 IRD Cost CHF 8859 QAPY 1.39 ISD Cost CHF 17,822 QAPY 1.50	Swiss francs 0-5000 ceiling ratio	Yes Cost per QAPY gained	ICER for Implant retained overdenture compared to complete denture and Implant supported overdenture compared to implant retained overdenture for 3, 5 and 10 years with discount rate of 0% and 3% were reported.	'Implant-supported overdenture becomes cost-effective at a threshold ratio of CHF 19,800 (base-case) and CHF 7100 (10-year horizon) /QAPY gained'(50).
Zitzmann et al, 2013	Prosthetic management	Implant supported single crowns (ISCS) vs fixed dental prosthesis (FDP) for the restoration of an anterior missing teeth	ISCS Cost CHF 4498 ± 632, QATY 2.82 ± 0.16 FDP Cost CHF 5082 ± 432, QATY 2.80 ± 0.24 Both additional costs of CHF 200/ patient	Swiss francs 0-5000 ceiling ratio	Yes	Implant treatment CHF 584 (incremental costs), an increase in expected QATY of 0.01 over a time horizon of 3 years and 0.04 QATY over a 10 years compared to the fixed dental prosthesis.	'Implant supported single crowns (ISCS) is cost-effective compared to the fixed dental prostheses (FDP)'(34).

Table 2.6: Appraisal of the studies against CHEERS checklist

Item	Study																						
	Acevedo et al, 2016	Agha et al, 2005	Balevi et al, 2007	Bhuridej et al, 2007	Chun et al, 2016	Ciketic et al, 2010	Cobiac & Vos, 2012	Cunningham et al, 2003	de Almeida et al, 2016	Dedhia et al, 2011	Fardal & Grytten, 2014	Higashi et al, 2002	Hulme et al, 2014	Hulme et al, 2016	Jensen et al, 2016	Koh et al, 2015	Mohd-Dom et al, 2014	Skaar et al, 2015	Speight et al, 2006	van der Linden et al, 2016	van der Meij et al, 2002	Zitzmann et al, 2006	Zitzmann et al, 2013
Title and abstract																							
Title	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1
Abstract	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	0
Introduction																							
Background and objectives	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Methods																							
Target population and subgroups	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Setting and location	0	1	1	1	1	1	1	0	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1
Study perspective	1	1	0	1	0	0	1	0	1	1	0	1	1	1	1	1	1	1	1	1	0	1	1
Comparators	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Time horizon	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Discount rate	1	1	0	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	0	1	1
Choice of health outcomes	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Measurement of effectiveness	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Measurement and valuation of Preference-based outcomes	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Estimating resources and costs	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	1
Currency, price, date and conversion	1	1	0	1	1	1	1	0	1	0	1	0	1	0	0	1	1	1	1	1	0	1	1
Choice of model	1	1	1	NA	1	1	1	NA	1	1	NA	1	NA	NA	NA	1	NA	1	1	1	1	NA	1

Assumptions	1	1	1	NA	1	1	1	NA	1	1	NA	1	NA	NA	NA	1	NA	1	1	1	1	NA	1
Analytical methods	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Results																							
Study parameters	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Incremental costs and outcomes	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0
Characterizing uncertainty	1	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1
Characterizing heterogeneity	0	1	0	1	0	1	1	1	1	0	0	1	0	1	0	0	0	0	1	0	1	0	0
Discussion																							
Study findings, limitations, generalizability, and current knowledge	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1
Other																							
Source of funding	1	0	1	0	1	1	1	1	0	1	1	1	1	1	1	1	1	0	1	0	0	1	1
Conflicts of interest	1	0	1	0	0	0	1	0	0	1	1	0	0	1	1	0	1	1	1	1	0	0	0
Total	**22**	**22**	**18**	**20***	**18**	**22**	**24**	**18***	**22**	**22**	**19***	**22**	**19***	**20***	**19***	**22**	**21***	**22**	**24**	**22**	**18**	**19***	**20**
Percentage (%)	**92**	**92**	**75**	**91**	**75**	**92**	**100**	**75**	**92**	**92**	**86**	**92**	**86**	**91**	**86**	**92**	**95**	**92**	**100**	**92**	**75**	**86**	**83**

**Out of 22 criteria*

2.1.5 Discussion

Oral health interventions can have a considerable impact on the improvement of patients' QoL. CUA can be considered an important technique to be used to evaluate oral heath interventions (81). A recent systematic review of economic evaluations in dentistry identified eight CUAs in oral health from 1975 to 2013, of which seven were published since 2000 (22). The present review was able to identify an increasing number of publications describing CUAs over time. This is a notable trend indicating growing attention being paid to the use of CUA in dentistry. However, there were only two studies from Asian countries where resources and money are scarcer than in developed countries: none were found from Africa, Latin America or South America. Even though it was evident from a systematic review (80) that the number of CUAs in health care interventions in Asia has increased gradually, it seems that CUA is not commonly used to prioritize oral health interventions in most low and middle income countries. In these developing countries, prioritization of oral health interventions may be based on research conducted in developed countries and lean more towards ad hoc policy decisions dependent on the political and economic situations of the particular country (105). In contrast, CUA is the most preferred or required method by health technology assessment (HTA) agencies (37-40) in most developed countries to evaluate interventions in all health systems including oral health. This is evidenced by the increasing number of CUA particularly from the developed countries over time.

Most publications were able to demonstrate that particular interventions were indeed cost-effective, as confirmed by this review. This could be partially due to publication bias, where a cost-effective intervention has a higher probability of being published than interventions that are not cost-effective. The QALY has been the preferred method used to measure health outcomes in CUAs published during the past 20 years (44), again as confirmed by the present study. Interestingly, a few studies used different derivatives of quality-adjusted outcomes specific to dentistry (such as QATY, QAPY and QLTY). It is inevitable that oral health-specific utility measures will be more sensitive in capturing the effectiveness of oral health interventions; and more appropriate when comparing the effectiveness of different oral health interventions. However, these cannot be used to compare cost-effectiveness across different areas of health, as is the case with QALY (51). Comparison of incremental gains in QALY across a range of health care interventions through league tables allows evaluation of the cost-effectiveness of oral

health interventions in comparison to those in other areas of health: such information is of great potential value to decision makers in provision of care (32). The assessment of comparative effectiveness of oral health interventions with interventions in different areas of health is not simple and straight forward as evidence suggests that the different health utility instruments would result in different values based on the descriptive systems (106) and the methods used to elicit preferences especially in dentistry (107). It is prudent, therefore, to consider whether the use of oral health-specific utility measures hinders the main strength of the CUA when debates on optimisation of the total health budget across all areas of health are held.

Detailed descriptions of the sources and estimates of cost and outcome data (as well as adherence to standard economic evaluation methods in research) can provide better guidance for policy makers and practitioners in health care decisions. Economic evaluations can be conducted from different perspectives such as those of a health-care provider, a health system, patients, third parties and/or society (21). This affects the types of costs relevant to the analysis. It is impressive that most studies included in the present review explicitly stated the economic perspective. However, wide variation in the reporting of cost data was observed; this has an impact on the decision-making process where comparisons of all available interventions are made with the intent of a 'best' option being recommended for implementation in real practice settings.

The CHEERS checklist provides guidelines for the reporting of health economic evaluations. The present review revealed high-quality reporting associated with CUAs for oral health interventions published during the years 2000 to 2016. However, providing a statement on conflict of interest and the characterising of heterogeneity should be improved. A recent systematic review that included all types of economic evaluations in dentistry (22) concluded that the quality should be improved, especially in the areas of discounting and sensitivity analysis. However, the majority of the articles included in the present review undertook discounting and sensitivity analyses. Although the earlier systematic review (22) did not report the quality of studies based on the type of economic evaluation, it appears as though non-CUA evaluations may suffer from poorer reporting than CUA evaluations.

This systematic review has some limitations. We included only English-language articles with original cost-utility estimates. CUAs published in non-English languages were not reviewed, and this may have limited the number of articles identified from non-English speaking countries and (in particular) poorer countries. Also, direct comparison

across all oral health interventions was not possible due to differences in methods used in the included studies, such as different populations, perspectives, costs and outcome measures.

In conclusion, this study provides an overview of usage of CUA in oral health interventions, the methods used and the reporting quality of these. The use of CUA in evaluation of oral health interventions has been increasing over time. Except for 2, all publications included in the present review were able to provide conclusions regarding the most cost-effective intervention among the different options compared, which will assist in resource allocation and in health care decision-making. The majority of the articles followed the standard methods of reporting health economic evaluations although the provision of a statement on conflict of interest and characterisation of heterogeneity are areas which need improvement.

2.2 Pediatric Quality of Life Instruments in Oral Health Research: A Systematic Review

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a published co-authored paper. The bibliographic details of the co-authored paper, including all authors, are:
Hettiarachchi RM, Kularatna S, Byrnes J, Scuffham PA. Pediatric Quality of Life Instruments in Oral Health Research: A Systematic Review. Value Health. 2019; 22(1):129-35. https://doi.org/10.1016/j.jval.2018.06.019

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The article has been published in a peer-reviewed journal with an international readership. The Impact Factor (2018) for this journal is 5.037. The journal 'Value in Health' allows the authors to archive post-print (accepted peer-reviewed) version after an embargo period of 12 months. Further details are available here: https://www.elsevier.com/about/policies/sharing

My contribution to the paper involved: designing of the study, literature search, quality assessment of the included studies, data analysis and drafting the article.

(Signed) _____ (Date) _____

Ruvini Hettiarachchi

(Countersigned) _____ (Date) _____

Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date) _____

Supervisor: Prof. Paul Scuffham

Pediatric Quality of Life Instruments in Oral Health Research: A Systematic Review

Hettiarachchi RM, Kularatna S, Byrnes J, Scuffham PA.

2.2.1 Abstract

Objectives

To identify the generic or disease-specific paediatric quality of life (QoL) instruments used in oral health research among children and adolescents; and to provide an overview of these QoL instruments.

Methods

A systematic literature search was performed with multiple databases to identify the paediatric QoL instruments used in oral health research.

Results

The literature search yielded 872 records and from these 16 paediatric QoL instruments were identified that had been used among children and adolescents in oral health research. Of these, 11 were oral health-specific QoL instruments and five were generic instruments. Of the 11 oral health-specific QoL instruments, none were multi-attribute utility instruments (MAUI) whereas out of five generic instruments, two (CHU 9D and EQ-5D-Y) were classified as a MAUI. Except for one, all paediatric QoL instruments were published after the year 2000 and the majority originated from the USA (n=8). Of the 11 oral health-specific QoL instruments, five instruments are designed for the respondent to be a child (i.e. self-report), one uses proxy responses from a parent/guardian and five instruments have both self and proxy versions. Of the five generic QoL instruments, one uses proxy responses and the other four instruments have both self and proxy versions.

Conclusion

This review identified a wide variety of paediatric oral health-specific and generic QoL instruments used in oral health research among children and adolescents. The availability of these QoL instruments provides researchers with the opportunity to select the instrument most suited to address their research question.

Highlights

- Oral health related Quality of Life (OHRQoL) among children and adolescents is an important area of research with growing attention within both research and clinical practice and as a result, a considerable number of QoL instruments have been developed for paediatric populations over the recent past.
- The present review provides an overview of 16 paediatric quality of life (QoL) instruments used among children in oral health research. Of these 16 instruments, 11 were oral health-specific QoL instruments and five were generic instruments.
- The availability of these QoL instruments for children and adolescents provides researchers with the opportunity to select the instrument most suited to address their research question.

2.2.2 Introduction

Oral health related Quality of Life (OHRQoL) among children and adolescents is an important area of research with growing attention both within research and clinical practice (11). Sicho and Border reported a marked increase in OHRQoL research in paediatric and orthodontics during 2006-2010 compared to other disease areas such as geriatrics, oral medicine and surgery (11). Oral health problems have considerable impact on Quality of Life (QoL) of the children (13, 14) and adolescents (15). Dental caries is the most common chronic childhood disease and has negative impacts on eating, sleep, school performance, smiling patterns and social interactions (14). Children with traumatic injuries to teeth (15), malocclusion (i.e. imperfect positioning of the teeth when the jaws are closed (16)) and dental fluorosis (i.e. developmental defect of tooth enamel (17) caused by excessive intake of fluorides, characterized by discoloration and pitting of teeth) also have negative impacts on children's QoL and their day-to-day living. As oral disorders cause significant impact on an individual's physical, social and emotional well-being, the psychological impact of oral diseases can disrupt the quality of life (9), self-esteem (9) and positive social interactions (1).

Children and adolescents have a different perception about QoL issues compared to adults. Young children are the prime target group of oral health care services in many countries (108) and assessment of QoL of children and adolescents provides useful information on the impact of oral health (109). In clinical settings, measuring QoL will facilitate prioritizing health problems for individual patients and monitoring responses to

treatment (41). These will also guide policymakers, researchers, programme evaluators and clinicians to assess health care interventions and the prioritization of health care resource allocation (41). As a result of the growing research interest in QoL among children and adolescents, a considerable number of paediatric QoL instruments have now been developed (63) despite the great difficulties associated with development and validation of paediatric QoL instruments.

There are different types of QoL instruments. Generic QoL instruments are designed for different types of disease and different patient populations (32). These are comprehensive measures of QoL that are widely used and have established validity and reliability across different disease conditions and patient populations. Disease-specific QoL instruments are designed to assess the QoL concerning specific diseases, medical conditions or patient populations (32, 110). The generic and disease-specific QoL instruments that are developed based on classification system and preferences weights are known as preference-based or multi-attribute utility instruments (MAUI) (32). These preference-based instruments are extensively used in cost-utility analysis (32).

In many areas of health, QoL instruments specifically developed for children and adolescents have been comprehensively reviewed (111-113). Nevertheless, in oral health there is a great paucity of research. The assessment of the methodological quality of child oral health related QoL measures reported by Gilchrist et al. (63) was confined to the most frequently used three child oral health related QoL measures Child Perceptions Questionnaire (CPQ), the Child Oral Impacts on Daily Performances (C-OIDP) and the Child Oral Health Impact Profile (COHIP). Hence, a comprehensive review of the QoL instruments used among children and adolescents in oral health is warranted and will facilitate researchers to use instruments that are more appropriate in oral health research. Therefore, the present review was conducted to identify generic or disease specific and preference- or non-preference-based paediatric QoL instruments used in oral health research among children and adolescents and to provide an overview of these QoL instruments.

2.2.3 Methods

The review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) strategy **(Figure 2.2),** which allows systematic selection of articles (85). The Population, Intervention, Comparator and Outcome (PICO) for this review was as follows: Population: children and adolescents with oral health issues;

Intervention/comparator: any oral health condition; and Outcome: QoL using any paediatric QoL instrument. To identify the QoL instruments used in oral health studies among children and adolescents, a systematic literature search was performed with multiple databases including MEDLINE, Dentistry & Oral Sciences Source, Cumulative Index to Nursing and Allied Health Literature (CINAHL Plus), and Econlit through EBSCohost, Cochrane Database of Systematic Reviews and PubMed. Literature was searched up to February 2017 with search terms built around 'oral health' AND 'Children or adolescents' AND 'quality of life' with appropriate truncation and adjacency settings. Full search terms are available in the **Text Box 2.2.**

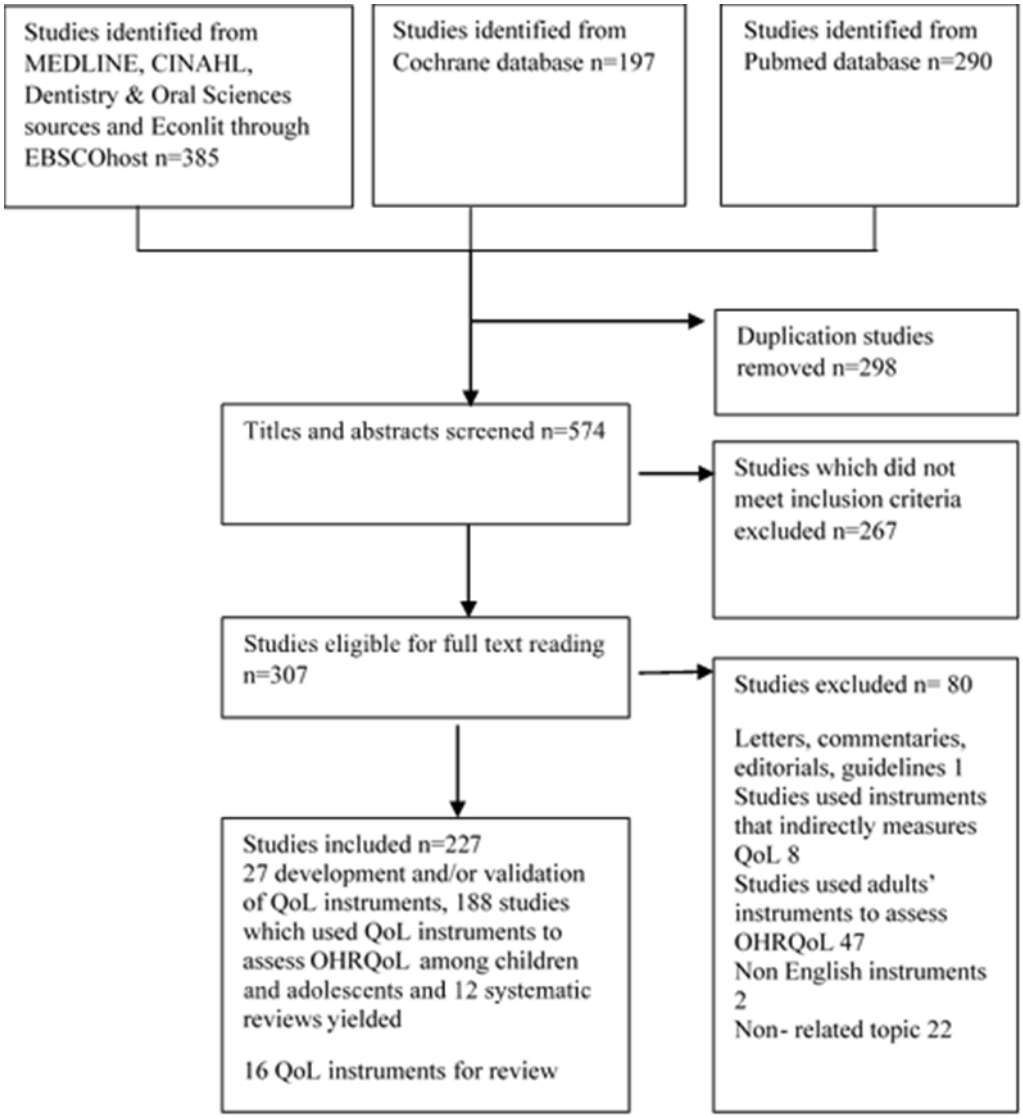


Figure 2.2: PRISMA flow diagram of the different phases of the systematic review

S1: (**MH** "Dentistry+")

S2: **Abstract, title and full text** "dental" or "dentistry" or "Oral Health" or "oral hygiene" or "tooth decay" or "carious" or "dental caries" or "periodontal" or "oral precancer*" or "oral neoplas*" or "children dental care" or "Fluoride mouth rins*" or "Fluoride mouth wash" or "water fluoridation" or "fissure sealant" or "dental prosthe*" or "orthodontic" or OPMD or TMJ

S3: (**MH** "child") OR (**MH** "adolescent") OR (**MH** "infant")

S4: **Abstract, title and full text** child* OR adolescen* OR teenage* OR kid* OR pediatr*

S5: **Abstract, title and full text** "Quality of life" or "QOL" or "Quality of life instruments" or "MAUI" or "multi attributable utility instruments" or "Generic" or "condition specific" or "oral health related quality of life" or OHRQoL or "HRQoL" or "preference based instrument"

S6: **Abstract, title and full text** "instrument*" or "questionnaire"

S7: **Abstract, title and full text** "validation" or "validity" or "reliability" or "adaptation" or "valid" or "reliable" or "psychometric" or "dimensions"

Search (S1 OR S2) AND (S3 OR S4) AND S5 AND S6 AND S7

Text box 2.2: Search terms

Of the publications retrieved through database searches, any study or systematic review that included a paediatric QoL instrument/questionnaire to measure QoL related to any oral health condition in children or adolescents was included in the review. Inclusion was restricted to the studies published in English language and human studies. Letters, commentaries, editorials, conference abstracts and proceedings, guidelines, surveys and case reports were excluded from the review. Studies that used instruments which indirectly measured QoL (e.g. Shame and stigma scale, pain scales, disease symptom inventories, jaw function limitation scale), did not use separate QoL instruments, but instead incorporated a few QoL based questions into knowledge and practices questionnaires, used QoL instruments in languages other than English where an English version was not available, used QoL instruments specifically designed for a defined geographical region or specific ethnic group (e.g., COHRQoL-25 for Indonesia

and OH-ECQoL for North Indian population, since these instruments could be used only for very restricted populations), and used QoL instruments developed originally for adults older than 18 years to assess OHRQoL among children and adolescents were also excluded from the review.

A list of QoL instruments used in oral health studies among children and adolescents was prepared from the information on the included studies and systematic reviews. After this, a specific search was done to obtain the information regarding each instrument in the list by searching the specific web page for the instrument or identifying the original development and validation study of the instrument. Characteristics of each instrument were then extracted into an excel table including generic or disease specific instrument, country of origin, specified age group, number of items and domains, proxy or self-reported, time to complete the instrument and whether validity and reliability were established or not.

2.2.4 Results

The literature search yielded 872 records and after removing duplicates 574 records were selected for the title and abstract review. In the next step, 267 studies were excluded based on the exclusion criteria and 307 were eligible for the full text reading. This resulted in a total of 228 articles included in the review (27 development and/or validation of QoL instruments, 188 studies that used QoL instruments to assess OHRQoL among children and adolescents and 12 systematic reviews) with 16 paediatric QoL instruments used in oral health research. Of these 16 instruments, 11 were oral health-specific QoL instruments and five were generic instruments. Of the 11 oral health-specific QoL instruments, none were MAUIs, whereas out of five generic instruments, two (Child Health Utility-9D index [CHU9D] and Euro QoL-5D youth [EQ-5D-Y]), were classified as MAUI.

Oral health-specific QoL instruments for children and adolescents

All oral health-specific QoL instruments were published after the year 2000. Child Oral Health Impact Profile (COHIP) (114), Child Oral Health Quality of Life Questionnaire (COHQoL), Child Oral Impacts on Daily Performances (C-OIDP) (108) and Early Childhood Oral Health Impact Scale (ECOHIS) (115) were the most common instruments used among the assessment of OHRQoL among children and adolescents. Of these 11 oral health-specific QoL instruments, the majority originated from the USA

(n=6), followed by the UK (n=3), Canada (n=1) and Thailand (n=1). Among these instruments, the COHIP and the C-OIDP have corresponding adult measures. Development of the C-OIDP (108)was based on the original adult OIDP index whereas others were developed based on the items derived from the literature review and focus group discussions with experts, parents, children and adolescents. The applicable age groups for these instruments ranged from 3 to 18 years.

COHIP, COHQoL, C-OIDP, Paediatric Oral Health-related Quality of life Questionnaire (POQL) (116) and Teen Oral Health-Related Quality of Life instrument (TOQOL) (117) were developed to assess OHRQoL across various oral health problems. COHIP (114, 118), COHQoL (119-121) and POQL (116) are available in different formats such as long and short versions and self-reported and proxy versions. Moreover, COHQoL consists of different measures namely, Child Perceptions Questionnaire for 11-14 years (CPQ 11-14), Child Perceptions Questionnaire for 8-10 years (CPQ 8-10), Parental perceptions questionnaire (PPQ) and family impact scale. Paediatric Quality of Life Inventory (PedsQL) Oral Health Scale was designed to be used in combination with the PedsQL 4.0 Generic Core Scales and/or disease-specific modules, to measure child's general oral health status in patients with acute and chronic health conditions and healthy children, which can then be related to QOL assessed Generic Core Scale or disease-specific modules of PedsQL (122, 123). The Scale of Oral Health Outcomes for 5-year-old children (SOHO-5) (124), Early Childhood Oral Health Impact Scale (ECOHIS) (115) and Michigan Oral Health-Related QoL (MOHRQoL) Scale (125) were designed for the assessment of OHRQoL mainly in early childhood dental caries. Malocclusion Impact Questionnaire (MIQ) (126, 127), and OHRQoL Hypodontia (128) are the other condition- specific OHRQoL instruments developed for children and adolescents**.**

Of the 11 oral health-specific QoL instruments, five instruments were self-report, one used proxy responses and five instruments had both self and proxy versions. The number of domains included in the instrument ranged from 1 to 5 and the number of items included in the instruments ranged from 7 to 37. Except for PedsQL Oral Health Scale, all oral health-specific QoL instruments included non-oral health domains and/or items such as functional, social and emotional well-being. These non-oral health domains and items meant to capture the impact of conditions of teeth, jaws and face on the child's overall health and emotional and psychosocial consequences of the oral diseases. The recall period for the instruments ranged from 'at the moment' to the 'entire life span'.

Table 2.7 provides a detailed description of the oral health-specific QoL instruments identified.

Except for the Michigan Oral Health-Related QoL scale (125), all oral health QoL instruments reported the assessment of psychometric properties, at the initial development and evaluation process. The authors reported face validity and reliability only for the original child version of the Michigan Oral Health-Related QoL scale (125). Among the instruments that reported psychometric properties, the C-OIDP showed evidence for the concurrent validity and reliability, whereas the other instruments mainly reported evidence for the construct validity and reliability. Nevertheless, authors of the CPQ 8-10 (120) and the Family impact scale (129) of the COHQoL suggested that the discriminative validity of these scales needed to be further evaluated. A detailed description of validity and reliability of the oral health-specific quality of life instruments at the initial development and evaluation process are provided in the **Table 2.8.**

Table 2.7: Characteristics of the oral health-specific QoL instruments among children and adolescents

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of scores	Scoring method	Respondent	Avg. time to complete	Recall period
1. COHIP	USA	COHIP (114, 130)	2007	8-15 years	5	1.Oral health 2.Functional well-being 3.Social-emotional well-being 4.School environment 5. Self-image	34	0-136	5-point scale (0–4)	Self/ proxy	NM	3 months
		COHIP-SF19 (118)	2011	7-17 years	3	1.Oral Health 2.Functional well-being 3.Socio-emotional well-being	19	0-76	5-point scale (0–4)	Self/ proxy	Less than 10 minutes	3 months
2. COHQoL	Canada	CPQ 8-10 (120)	2004	8-10 years	4	1.Oral symptoms 2.Functional limitations 3.Emotional well-being 4.Social well-being	25	0-100	5-point scale (0–4)	Self	NM	4 weeks
		CPQ 11-14 (119)	2002	11-14 years	4	1.Oral symptoms 2. Functional limitations 3. Emotional well-being 4.Social well-being	37	0-148	5-point scale (0–4)	Self	NM	3 months

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of scores	Scoring method	Respondent	Avg. time to complete	Recall period
		CPQ11-14 short (16 items) (121)	2004	11-14 years	4	1.Oral symptoms 2.Functional limitations 3.Emotional well-being 4.Social well-being	16	0-64	5-point scale (0–4)	Self	NM	3 months
		CPQ11-14 short (8 items) (121)	2004	11-14 years	4	1.Oral symptoms 2.Functional limitations 3.Emotional well-being 4.Social well-being	8	0-32	5-point scale (0–4)	Self	NM	3 months
		P-CPQ (131)	2003	Parents / caregiver of 6-14 years	4	1.Oral symptoms 2.Functional limitations 3.Emotional well-being 4.Social well-being	31	**NS**	5-point scale (0–4)	Proxy	NM	3 months
		FIS (129)	2001	Parents / caregiver of 6-14 years		1.Oral symptoms 2.Functional limitations 3.Emotional well-being 4.Social well-being	14	0-33	5-point scale (0–4)	Proxy	NM	3 months
3. C-OIDP (108)	Thailand		2003	11-12 years	1	-	8 performances	0-72	4 point scale (0–3)	Self Interview with pictures	10 minutes	3 months
4. ECOHIS (115)	USA		2006	3-5 years	6	1.Symptoms 2.Function	13	Child section	6 point scale	Proxy	NM	Entire life span

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of scores	Scoring method	Respondent	Avg. time to complete	Recall period
						3. Psychological 4. Self-image/ social interaction 5. Parent distress 6.Family function		0-36 family section 0-16				
5. Michigan OHRQoL scale (125)	USA	Orginal child version	2002	4 years and above	3	1. Pain/Discomfort 2. Functional 3.Psychological	7	Categorical	Yes/no	Self	NM	At the moment
		Modified child version		3 years and above	4	1. Pain/Discomfort 2. Functional 3.Psychological 4.Social	9	Categorical	Yes/no	Self	NM	At the moment
		Parent version					10	1-5	5 point scale	Proxy	NM	At the moment
6. MIQ (126, 127)	UK		2016	10-16 years	3 themes	1.Appearance of teeth 2.Effect on social interactions 3. Oral health and function	17	0-34	3 point scale	Self	NM	NM
7. OHRQoL Hypodontia (128, 132)	UK		2011	11-18 years	4 themes	1.Treatment 2. Effect on daily activities 3.Appearance 4. Other peoples' reactions	7		5 point scale	Self	7 min	At the moment
8.PedsQL Oral Health Scale (122)	USA	Toddlers, Young child, Child and	2009	2-4 years, 5-7 years,	1	1.Oral Health	5	0-100	5-point scale (0 to 4) and 3-point	Self/ Proxy	NM	Past one month

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of scores	Scoring method	Respondent	Avg. time to complete	Recall period
		Adolescent versions		8-12 years and 13-18 years					scale (0 to 2) for young Child self-report			
9. POQL (116)	USA	Pre-school version, school-age and pre-teen version	2011	Pre-school and school-age	4	1.Emotional 2. Physical 3. Role 4. Social	10	0 -100	Likert-scale	Self/proxy	NM	Past 3 months
10. SOHO-5 (124)	UK		2012	5 years	NS	NS However included questions from functional and psychosocial domains	7	0-14	3-point scale	Self	5–6 minutes	Life time (ever)
11. TOQOL (117)	USA		2015	13-18 years	5	1.Physical functioning 2. Role functioning 3. Social functioning 4. Oral problems 5.Emotional functioning	16	1-100	Event (Likert scale 1-4, how bothered 1-5)	Self	NM	Past 3 months

**manuscript received year or year of publication of the development study was considered as the year of development of the instrument.*

COHIP - Child Oral Health Impact Profile; COHIP-SF 19 - Child Oral Health Impact Profile-Reduced; COHQoL-Child Oral Health Quality of Life Questionnaire; CPQ 11-14- Child Perceptions Questionnaire for 11 to 14 years; CPQ 8-10- Child Perceptions Questionnaire for 8 to 10 years; CPQ11-14 short- Short forms of CPQ11-14 (16 and 8 items); FIS- Family Impact Scale; P-CPQ - Parental-Caregiver Perceptions Questionnaire; C-OIDP- Child Oral Impacts on Daily Performances; ECOHIS- Early Childhood Oral Health Impact Scale; Michigan OHRQoL scale- Michigan oral health related QoL scale; MIQ- Malocclusion Impact Questionnaire; NM-Not mentioned; OHRQoL hypodontia- Oral health related QoL questionnaire for hypodontia; PedsQL Oral Health Scale-Paediatric Quality of Life Inventory Oral Health Scale; POQL- Paediatric Oral Health-related Quality of life Questionnaire; SOHO-5- Scale of Oral Health Outcomes for 5-year-old children; TOQOL- Teen Oral Health-Related Quality of Life instrument

Table 2.8: Psychometric properties of oral health-specific QoL instruments

Instrument	Validity						Reliability		Conclusion from the authors
	Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
1. COHIP									
COHIP (114, 130)	Done	NM	NM	Construct –factor analysis Discriminant validity significant differences among the three clinical groups and community group: Convergent validity by statistically significant ($P < 0.05$) partial Spearman correlations between COHIP scores and Global Health Ratings	Pearson's correlations between domains of the COHIP and the domains of Dento-facial Image, the Social Anxiety Scale, and Multidimensional Self Concept Scale were in expected directions (133).	NM	0.91	ICC = 0.84 no significant score shift over time	'COHIP showed excellent scale reliability and test–retest reliability. Discriminant, convergent and concurrent validity were established' (130, 133).
COHIP-SF 19 (118)	NM	NM	NM	Construct – confirmatory factor analysis Discriminant validity significant differences among the disease severity in paediatric, orthodontic and cleft lip and palate samples Convergent validity by statistically significant ($P < 0.05$) partial Spearman correlations between COHIP scores and Global Health Ratings.	NM	NM	0.82	NM	'Reliability and validity testing demonstrate that the COHIP-SF 19 is a psychometrically sound instrument' (118)
2.COHQoL									
CPQ 8-10 (120)	NM	NM	NM	Positive correlations between the overall scores and the ratings for oral health ($r = 0.17$; not significant) and overall well-being ($r = 0.45$; $P < .001$). CPQ8-10 score and number of decayed surfaces $r = 0.29$, do not demonstrate discriminative validity	NM	NM	0.89	0.75	'Results suggest good construct validity, internal consistency and test retest reliability, but do not demonstrate discriminative validity' (120).

Instrument	Validity						Reliability		Conclusion from the authors
	Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
CPQ 11-14 (119)	NM	NM	NM	Discriminant- significant differences among the overall scores of the clinical groups Significant positive correlation between CPQ 11-14 scores and global ratings of oral health ($p = 0.013$) and overall wellbeing ($p < 0.001$).	NM	NM	0.91	0.90	'These results suggest that the CPQ11-14 is valid and reliable' (119)
CPQ11-14 short (121)	NM	NM	All short forms except the ISF-8 were correlated with the long-form (Spearman's rho 0.87 to 0.98)	Discriminant – All short forms detected differences in impact on the quality of life among the three clinical groups (Oro-facial, orthodontic and paediatric dentistry groups) in the expected direction Correlational construct validity – all short-forms demonstrated positive significant correlations with the ratings of oral health and overall well-being	NM	Relative validity (RV) computed as the ratios of F statistics for the short-forms and the original CPQ11–14.	CPQ11–14-ISF-16 0.83 ISF-8 0.83 RSF-16 0.71 RSF-8 0.73	ICCs ranged from 0.71 to 0.77	'All short forms demonstrated excellent criterion validity and good construct validity. The reliability coefficients exceeded standards for group-level comparisons' (121).
P-CPQ (131)	Done	Done	NM	The P-CPQ discriminated the three clinical groups (Oro-facial, orthodontic and paediatric) in the expected direction. Construct validity – significant associations between the total scores and global ratings of oral health and overall well-being ($P < 0.0001$) in the expected direction.		Feasibility was assessed by floor and ceiling effects	0.94	0.85	'P-CPQ has good construct validity, good internal consistency reliability, and excellent test-retest reliability' (131).
FIS (129)	Done	Done	NM	Item impact study and factor analysis Discriminant – significant differences were observed across the three clinical		FIS sensitive to variations in family	0.83	0.80	FIS shows good reliability and construct validity.

Instrument	Validity						Reliability		Conclusion from the authors
	Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
				groups included in the study for 10 of the scale's 14 items. Construct validity- significant associations between the FIS score and parent–caregiver global ratings of the child's oral health ($P < 0.001$) and overall wellbeing ($P < 0.0001$) in the expected direction.		impact and floor and ceiling effects were minimal			'Discriminant validity of the scale needs to be further tested' (129).
3. C-OIDP (108)	Done	Done	NM	NM	Strongly significant ($p<0.001$) with perceived oral treatment need and perceived oral health problems.	NM	0.82	0.4–0.7	'CHILD-OIDP index is a valid and reliable' (108).
4. ECOHIS (115)	NM	NM	NM	Discriminant - children with either 1–3 or ≥ 4 decayed and/or treated teeth had higher ECOHIS scores than those who were free of dental disease. Convergent-ECOHIS scores were significantly correlated with the global dental and general health measures in the expected direction. The correlation between the child and family impact sections was statistically significant (Spearman's $r = 0.36$, $P \leq 0.001$).	NM	Substantial floor effects and no ceiling effects were observed	child section 0.91 family section 0.95	0.84	'Study provided evidence for the construct validity, internal consistency and test-retest reliability of the ECOHIS' (115).
5. Michigan OHRQoL scale (125)									
Child version	Done	NM	NM	NM	NM	NM	0.54	NM	Not reported the validity and reliability of the questionnaire except for original child version
Modified child version	NM	NM	NM	NM	NM	NM	NM	NM	
Parent version	NM	NM	NM	Factor analysis	NM	NM	NM	NM	

Instrument	Validity						Reliability		Conclusion from the authors
	Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
6.MIQ (126, 127)	Good	Good	Significant correlation with the accepted gold standard (CPQ11–14-ISF16) r = 0·751; P < 0·001)	Correlation with the two global questions ('Overall bother' ρ = 0·733 and 'Life overall' ρ = 0·701) were high.	NM	Floor and ceiling effects were evaluated.	0·906	0·78	'Demonstrated good criterion validity, construct validity and reliability' (127)
7.OHRQoL Hypodontia (128, 132)	Good	Good	High correlations with the seven questions selected from the CPQ (p =0.4–0.7, p < 0.01).	Principal component analysis of the four sections suggested that the proposed underlying hypothetical constructs were largely supported by the data.	NM	NM	0.89	ICC above 0.7 Bland and Almond method-random distribution	'Overall the questionnaire showed good reliability, criterion and construct validity' (132).
8.PedsQL Oral Health Scale (122)	NM	NM	PedsQL oral health scale both parent-proxy report and child self-reports significantly predicted dentist ratings of child oral health.	Construct validity- intercorrelations between the PedsQL 4.0 Generic Core Scales Oral Health Scale and known-group comparisons Convergent validity- PedsQL Oral Health scores significantly correlated with COHQoL scores (parent proxy reports r<0.81, p < 0.01 and child self-report r < 0.65, p < 0.01).	NM	NM	Parent proxy 0.84 Child self-report 0.68	ICC between parent proxy and child self-report 0.56 indicate moderate agreement	'The construct, criterion-related, and convergent validity of the measure was supported by the results of this study' (122)

Instrument	Validity						Reliability		Conclusion from the authors
	Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
9. POQL (116)	Done	NM	NM	Discriminant – there was a significant difference in total scores by caries status and by perceived oral health for both the child self-report (CSR) data and the parent report on child (PRC) data. Convergent – total scores on the POQL correlated significantly with total scores on the PedsQL for both the CSR ($r = -0.52$; $p < 0.001$) and the PRC ($r = -0.25$; $p < 0.001$).	NM	Strong sensitivity to Change	Child self-report (CSR) 0.83 Parent report on child (PRC) 0.86	0.75	'The POQL is a valid and reliable measure of oral health-related quality of life for use in pre-school and school-aged children' (116).
10. SOHO-5 (124)	Done	Done	NM	SOHO-5 scores were significantly associated with subjective oral health outcomes and an aggregate measure of clinical and subjective oral health outcomes.	NM	NM	0.74	Item-total correlation coefficients ranged 0.30 to 0.60	'Initial reliability and validity findings were very satisfactory' (124).
11. TOQOL (117)	Done Satisfactory	Done Satisfactory	NM	Convergent- TOQOL scores were significantly associated with perceived oral health status r=0.29 (P = 0.0001) and PedsQL scores r = –0.26 (P = 0.002) Discriminant – comparison among adolescents with caries and adolescents who were caries free (P = 0.049)	NM	NM	0.88	Mean percentage variation 0.85	'The TOQOL is a valid and reliable OHRQoL measurement in adolescents aged 13-18 years' (117).

** Cronbach's alpha **ICC- intra-class correlation NM-Not mentioned*

COHIP - Child Oral Health Impact Profile; COHIP-SF 19 - Child Oral Health Impact Profile-Reduced; COHQoL - Child Oral Health Quality of Life Questionnaire; CPQ 11-14 - Child Perceptions Questionnaire for 11 to 14 years; CPQ 8-10- Child Perceptions Questionnaire for 8 to 10 years; CPQ11-14 short - Short forms of CPQ11-14 (16 and 8 items); FIS - Family Impact Scale; P-CPQ - Parental-Caregiver Perceptions Questionnaire; C-OIDP - Child Oral Impacts on Daily Performances; ECOHIS - Early Childhood Oral Health Impact Scale; Michigan OHRQoL scale - Michigan oral health related QoL scale; MIQ - Malocclusion Impact Questionnaire; OHRQoL hypodontia - Oral health related QoL questionnaire for hypodontia; PedsQL - Oral Health Scale-Paediatric Quality of Life Inventory Oral Health Scale; POQL - Paediatric Oral Health-related Quality of life Questionnaire; SOHO-5 - Scale of Oral Health Outcomes for 5-year-old children; TOQOL - Teen Oral Health-Related Quality of Life instrument.

Generic QoL instruments used among children and adolescents in oral health research

Five generic QoL instruments used among children and adolescents in oral health research were identified from this review: Child Health Questionnaire (CHQ), Infant and Toddler Child Quality of Life Questionnaire (ITQoL), PedsQL Generic core scale, CHU9D and EQ-5D-Y. Except for PedsQL Generic core scale, all included generic QoL instruments were published after the year 2000. Of the 5 generic QoL instruments, one uses proxy responses (134) and the other four instruments have both self and proxy versions. The number of domains included in the instrument ranged from 4 to 14 and the number of items included in the instruments ranged from 5 to 97. The characteristics of the generic QoL instruments used among children and adolescents in oral health research are provided in the **Table 2.9.**

The use of generic QoL instruments in oral health research was mainly for the purpose to use as a tool to evaluate the construct validity of the oral health-specific QoL instruments (117, 135, 136) and to evaluate the impact of childhood conditions including dental diseases on the QoL of the children (137). Nevertheless, except for the CHQ and the EQ-5D-Y, psychometric properties of the other three generic instruments have been evaluated among paediatric population in oral health research and details of these psychometric evaluations are provided in the **Table 2.10.**

Table 2.9: Characteristics of the generic QoL instruments used among children and adolescents in oral health research

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of score	Scoring method	Respondent	Avg. time to complete	Recall period
1. CHQ	USA	Parent form in 2 lengths CHQ-PF28		5–18 years		14 physical and psychosocial concepts	28 questions	0-100 Population norms available	Likert scale	Proxy	5-10 min	Vary depending on the subscale (ex. past 4 weeks/ in general)
		CHQ-PF50		5–18 years		14 physical and psychosocial concepts	50 questions	0-100 Population norms available	Likert scale	Proxy	10-15 min	
		Child (CHQ-CF87)		10–18 years		14 physical and psychosocial concepts	87 questions	0-100 Population norms available	Likert scale	Self	14 min	
2. CHU9D	UK		2009	7–17 years	9	1.Worried 2.Sad 3.Pain 4.Tired 5.Annoyed 6.School work/ home work 7.Sleep 8.Daily routine able to join in activities	9	Health state utility	5 levels	Self/ Proxy	NM	Today/last night
3. EQ-5D-Y (138)	UK			4–15 years	5	1.Mobility, 2.Self-care 3.Usual activities 4. Pain or discomfort 5.Anxiety/ depression	5	Health state utility	3 levels	Self/ Proxy	NM	At the moment

Instrument	Country of origin	Available formats	Year*	Age group	No. of domains	Domains	No. of items	Range of score	Scoring method	Respondent	Avg. time to complete	Recall period
4. ITQOL	Canada	ITQOL full version	2003	2 months – 5 years		10 multi-item scale (7 infant scales and 3 parent scales)	97	0-100 Population norms are available	Usually 5 levels	Proxy	Vary depending on issues such as the setting, context, age, cognitive functioning, language, layout, etc.	Vary depending on the subscale (ex. past 4 weeks/in general)
		ITQOL-SF47					47					
5.PedsQL 4.0 Generic Core Scales	USA	Toddlers version	1999	2–4 years	4	1. Physical functioning 2. Emotional functioning 3. Social functioning 4.School functioning	23	0-100	5-point Likert scale (0–4)	Proxy	Less than 4 minutes	Past one month
		Young child, Child and Adolescent versions		5–7 years, 8–12 years, 13–18 years		1. Physical functioning 2. Emotional functioning 3. Social functioning 4. School functioning	23	0-100	5-point Likert scale (0–4) and 3-point scale (0–2) for young child self-report	Self/proxy	Less than 4 minutes	Past one month

**manuscript received year or year of publication of the development study was considered as the year of development of the instrument.*
CHQ - Child Health Questionnaire; CHU 9D - Child Health Utility 9D index; EQ-5D -Y - EQ 5D youth version; ITQOL - Infant and Toddler Child Quality of Life Questionnaire; NM - Not mentioned; PedsQL - Paediatric Quality of Life Inventory.

Table 2.10: Psychometric properties of generic QoL instruments among paediatric population in oral health research

Instrument	Study	Validity						Reliability		Conclusion from the authors
		Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
1. CHQ	Validity and reliability not assessed among paediatric populations in oral health research									
2. CHU9D (139, 140)	Evaluated among 6–9 years old children attending for routine dental examinations in community dental clinics (139) and RCT among children 6–9 years old (140)				Construct validity – adequate CHU9D scores showed differences between children with and without dental caries but the difference was not significant	Concurrent validity- excellent. significant correlations with global impact scores	Unresponsive to the changing of dental caries experience (140)	0.66	NM	'CHU9D may be useful in dental research. Further exploration in samples with different caries experience is required' (139)
3. EQ-5D-Y	Validity and reliability not assessed among paediatric populations in oral health research									
4. ITQOL (141)	Evaluated among children with and without early childhood caries-related pain	NM	NM	NM	Discriminant and convergent validity by factor analysis – high success in item validity	NM	Assessment of floor and ceiling effects showed that the ITQOL scales performed well.	> 0.8 for all scales	Not done and justify why not done.	'The ITQOL proved to be a useful instrument for characterizing QOL in this dental caries-afflicted sample' (141).

Instrument	Study	Validity						Reliability		Conclusion from the authors
		Face	Content	Criterion	Construct	Concurrent	Other	Internal consistency*	Retest reliability**	
5. PedsQL 4.0 Generic Core Scale	Chinese version of PedSQL and ECOHIS were compared among children with and without early childhood caries	NM	NM	NM	Discriminant – no significant correlation was found with the correlation with the caries status and PedsQL 4.0 scores ($r = 0.02$; $P > 0.05$) but caries status was highly significant with the ECOHIS scores ($r = 0.66$; $P < 0.01$).	NM		0.74	NM	'The ECOHIS appears more sensitive than PedsQL 4.0 in assessing the impact of dental caries' (142)

** Cronbach's alpha **ICC - intra-class correlation; NM - not mentioned; CHQ - Child Health Questionnaire; CHU9D - Child Health Utility 9D index; ECOHIS - Early Childhood Oral Health Impact Scale; EQ-5D-Y - EQ 5D youth version; ITQOL - Infant and Toddler Child Quality of Life Questionnaire; PedsQL - Paediatric Quality of Life Inventory.*

2.2.5 Discussion

Our systematic review identified 11 paediatric oral health-specific quality of life instruments and five paediatric generic QoL instruments used in oral health research among children and adolescents. Solans et al. (112) reported a marked increase in QoL instruments, especially the disease specific QoL instruments for children and adolescents over the recent past in all areas of health. They identified 94 QoL instruments in all areas of health (30 generic and 64 disease-specific instruments) published during 1980 to 2006. Of these 94 instruments, 51 were published between 2001 and 2006. Except for one, all paediatric QoL instruments identified from our review also were published after the year 2000. The use of QoL instruments to assess the quality of life in children and adolescents has received growing attention within both research and clinical practice over the last decade (112). 'Oral health in America: A report of the Surgeon General' and 'Face of the Child' meeting held in June 2000 highlighted the importance of research to improve QoL of children and families with oral and craniofacial conditions (143-145). Further evidence has shown that there was a marked increase in OHRQoL research in paediatric and orthodontics during the recent past compared to other areas of oral health such as geriatrics and oral medicine/surgery (11). These have facilitated the development and validation of a considerable number of paediatric QoL instruments for children and adolescents in oral health over the recent past as evident in our review.

Many researchers have agreed that the development of child specific QoL instruments is more complex than the development of adult instrument because of the inherited problems associated with the process including lack of concept of QoL in children, problems due to rapid developmental changes among children and the appropriateness of using a proxy (109). Children and adolescents have different perceptions of QoL issues compared to adults, and these perceptions rapidly change with the physical and psychosocial development of a child. Therefore, the instruments specifically developed for them may be more sensitive in capturing the impact of oral disease on their quality of life and effectiveness of an oral health intervention than the information obtained using an adult QoL instrument among children and adolescents.

When deciding on the most appropriate measure of oral health-related QoL in children to use for a study, the choice will be dependent on the purpose of the study (109) as well as the nature of the study population such as age range. The present review identified a wide variety of paediatric QoL instruments used in oral health research,

including generic and disease specific QoL instruments. These instruments cover different age groups such as pre-school and school ages and instruments are available for self-completion by the child or parent proxies. The availability of a wide variety of QoL instruments for children and adolescents will facilitate researchers to choose better instruments for their research. Therefore, reviews of this type are important to assist researchers and programme evaluators in being aware of the range of instruments available and understanding the differences between them to help them make the most appropriate choice.

Psychometric properties of an instrument are an important factor to consider during the selection of the best tool to use in both research and clinical practice (146). A QoL instrument with good psychometric properties will measure what it is really meant to measure (i.e. valid) and be able to reproduce a result consistently (reliable) (146). Except for the Michigan Oral Health-Related QoL scale, all oral health-specific QoL instruments identified in the present review reported the evaluation of validity and reliability during the initial development and validation process. Nevertheless, the methods and the type of validity established ranged widely, thus the direct comparison of the psychometric properties of all identified instruments is not possible. Further psychometric properties of the most commonly used oral health-specific QoL instruments such as the COHQoL, C-OIDP and COHIP are established in different population groups and different cultural settings (63). Therefore, researchers are encouraged to consider the establishment of psychometric properties in the interested target population when choosing the best suited option for their research.

Economic evaluations have become a vital part of decision making in health care and cost utility analysis is the recommended method for many Health Technology Assessment agencies across the world (139, 147). MAUIs are increasingly being used to calculate quality-adjusted life-years in the cost-utility analysis framework (53). Out of 11 oral health-specific QoL instruments identified in this review, none were MAUIs. Therefore, researchers have to depend on direct methods or other generic MAUIs for the estimation of health-related quality of life utility estimates in oral health interventions for use in cost-utility analyses. Chen et al. (53) identified nine generic MAUIs developed for paediatric populations and, as evident in this review, CHU9D and EQ-5D-Y have been used for oral health research among children and adolescents**.** The assessment of psychometric properties of the CHU9D for oral health research among a paediatric population showed adequate construct and concurrent validity (139) and the researchers

concluded that the potential of using CHU9D in oral health research is needed to be further explored (139, 140). The EQ-5D-Y is available in different languages and different modes of administration (138); however the use of the EQ-5D-Y in oral health research was confined to using it as a tool to evaluate the convergent validity of the COHIP-SF19 German version (136). Therefore, whether the CHU9D or the EQ-5D-Y performs best in economic evaluations in paediatric oral health research remains unclear.

The present review did not include paediatric QoL instruments and articles published in languages other than English. This may affect the identification of QoL instruments used in non-English speaking countries to assess the QoL among children and adolescents in oral health. Therefore, we could not provide an overview of the QoL instruments, which can be used in these different sociocultural settings. In addition, we excluded the studies that used QoL instruments developed for adults to assess QoL among children and adolescents. Thus, it was not possible to provide an insight into how common the use of adult measures to assess the QoL among children and adolescents in oral health research.

In conclusion, the present review identified a wide range of paediatric oral health-specific and generic QoL instruments used in oral health research among children and adolescents. The availability of a wide variety of QoL instruments for children and adolescents will facilitate researchers to choose the best-suited QoL instrument for their research.

Research gap identified through the systematic reviews

The two systematic literature reviews identified two key factors related to CUAs among paediatric oral health interventions:

1) Although the use of CUA in economic evaluation of oral health interventions has been increasing, there is only a limited number of CUAs among paediatric oral health interventions. Of the 23 CUAs identified in this review, only four studies were conducted among paediatric populations (48, 51, 96, 97).

2) There is no paediatric oral health-specific preference-based quality-of-life measure (PBM) available to be used in oral health economic evaluations.

The limited number of CUAs is likely due to the fact that there is no oral health-specific paediatric PBM for use in economic evaluations. The availability of paediatric PBMs for oral health would facilitate the economic evaluation of oral health care interventions. Objectives 3 and 4 of this study were focused on developing an oral health-specific PBM to facilitate the economic evaluation of oral health interventions using a CUA framework. Chapter 3 provides a manuscript on developing a classification system for a PBM to achieve Objective 3 of the study. Objective 4 was to generate a preference-based algorithm of utility weights for the health states defined by the new classification system. The resulting three manuscripts are provided as Chapters 4, 5 and 6.

Chapter 3. Development of a classification (descriptive) system for a preference-based quality-of-life measure for dental caries (Dental Caries Utility Index - DCUI) among adolescents

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a submitted co-authored paper. The bibliographic details of the co-authored paper, including all authors, are:

Hettiarachchi RM, Byrnes J, Kularatna S, Chen G, Mulhern B, Scuffham PA. Development of a classification (descriptive) system for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among adolescents- Submitted to Journal of Public Health Dentistry (Manuscript ID-JPHD-OA-05-20-0212)

My contribution to the paper involved:
Designing the study, performing the literature search, data collection and data analysis, and drafting the article.

(Signed) _____ (Date)_____
Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____
Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____
Supervisor: Prof. Paul Scuffham

Development of a classification (descriptive) system for a preference-based quality-of-life measure for dental caries (Dental Caries Utility Index - DCUI) among adolescents

Ruvini M. Hettiarachchi, Sanjeewa Kularatna, Joshua Byrnes, Gang Chen, Brendan Mulhern, Paul A. Scuffham

3.1 Abstract

Objectives

Preference-based quality-of-life measures (PBMs) have been developed in many clinical areas to aid estimation of more accurate utility values for economic evaluations. Existing oral health-related quality-of-life (OHRQoL) instruments are non-PBM and hence, cannot be used to generate utility values. The objective of this study was to develop a classification system for a new PBM (Dental Caries Utility Index- DCUI) for the most prevalent childhood oral health condition: dental caries.

Methods

A systematic literature search of paediatric OHRQoL instruments was conducted to identify possible domains and items to be included in the classification system. Studies eliciting utility values for oral health outcomes and clinical dentistry references were also reviewed and, based on the findings, a draft classification system was developed. To refine the draft classification system, semi-structured interviews were conducted among a convenience sample of 15 12–17-year-old adolescents who had experience with dental caries. The system was further refined and validated by a group of dental experts, using a modified Delphi technique.

Results

The classification system comprised five items (pain/discomfort, difficulty in eating food/drinking, worried, ability to participate in activities, and appearance) and each item had a four-level response scale.

Conclusion

The classification system developed herein is considered an amenable tool for the subsequent development of a new PBM for dental caries. Once the scoring algorithm is completed, the classification system can be used to incorporate economic evaluations of dental caries health interventions in clinical trials or as a routine outcome measure at clinical practice.

Keywords: Preference-based measures, Classification system, Dental caries, Adolescents, Quality of life, Quality-Adjusted Life Years

3.2 Introduction

Oral diseases are one of the most prevalent ailments worldwide, affecting the daily activities and quality of life (QoL) of half of the world's population (2). Moreover, oral disease treatments are costly and have a significant impact on health care expenditure in many countries (2). Thus, strong evidence is required to prioritise limited health resources for oral health interventions. Economic evaluations assist the health care decision-making process by providing information on the interventions and strategies that offer the best value for money (32). Cost-utility analysis (CUA) is a preferred type of economic evaluation recommended by most health technology assessment agencies (HTA) in developed countries (52). In CUA, cost is measured in monetary units and the outcome is measured as a summary outcome: quality-adjusted life years (QALY) (32).

Preference-based quality-of-life measures (PBMs) are essential in calculating QALYs. PBMs are patient-reported outcome measures that consist of a health state classification system and a set of preference weights to generate health utility values (which lie on a 0–1 death-full health QALY scale) corresponding to each of the health states defined by the classification system (60, 61). PBMs can be either generic or disease/condition-specific. Generic PBMs are developed for use among populations with any disease or condition (60, 139); however, these instruments arguably do not include all important domains of every disease and therefore, they may be insensitive to relevant patient changes in certain conditions (61). Condition-specific PBMs (CSPBMs) are designed for a specific disease or condition, and may be more responsive to changes due to health care interventions (61). To date, several oral health-related quality-of-life

(OHRQoL) instruments have been developed and validated among paediatric populations (148). However, none are PBM and therefore, cannot be used in CUAs.

The most prevalent chronic childhood oral health condition is dental caries, which has considerable impacts on QoL, such as eating function, school performance and social interactions (14). A previous systematic review (147) identified four CUAs conducted among paediatric populations and all of them were focusing on dental caries-related interventions. In the absence of CSPBM for dental caries, researchers calculated health state utilities by using a direct valuation approach (48), applying different derivatives (e.g., disability-adjusted life years instead of the typically preferred QALY) (51, 96) or relying on generic PBMs (97). Although the generic paediatric PBMs, such as the EQ-5D-Youth (EQ-5D-Y) (149) and Child Health Utility 9D (CHU9D) (150), have been used in oral health research, their performance has not been extensively evaluated (148). Nevertheless, previous studies have shown that the CHU9D was unable to detect a significant difference between the caries statuses and was unresponsive to changes in the dental caries decayed, missing and filling status among children receiving dental treatment over a period of one year (140). Further, it has been reported that the Child Perceptions Questionnaire (CPQ), a non-PBM oral health quality of life measure, was able to establish significant differences in caries statuses and performed better than the CHU9D among children with dental caries (139). This empirical evidence has indicated that generic PBMs, such as the CHU9D, were unable to significantly detect differences between caries statuses and thus, they may not be appropriately responsive to measure the effectiveness of oral health interventions (139).

Considering that dental caries is the most prevalent chronic childhood oral health condition—and preventable in nature (22)—and given that children and adolescents are the primary target group of the public oral health care provision in many countries (108), there is an urgent demand to develop a CSPBM for economic evaluation. This study aims to meet this demand by developing a classification system for a new paediatric CSPBM for dental caries: the Dental Caries Utility Index (DCUI). This paper reports the first stage of the development process, the construction of the DCUI classification system.

3.3 Methods

A classification system can be developed from an existing non-PBM or as a *de novo* measure (62). Initially, a systematic literature search was conducted to identify available paediatric OHRQoL instruments (details published elsewhere (148)). Of the 11

identified, this study considers three non-PBM OHRQoL instruments specific to dental caries and five instruments developed to assess the impact of OHRQoL due to any oral disease. The former are the Early Childhood Oral Health Impact Scale (ECOHIS) (115), Scale of Oral Health Outcomes for 5-year-old children (SOHO-5) (124) and Michigan OHRQoL scale (125), which are specific to dental caries. The latter include the Child Oral Health Impact Profile (COHIP) (114, 118), Child Oral Health QoL Questionnaire (COHQoL) (119-121), Child Oral Impacts on Daily Performances (C-OIDP) (108), Paediatric OHRQoL Questionnaire (POQL) (116) and Teen OHRQoL instrument (TOQOL) (117), which assess the impact of OHRQoL due to any oral disease. The three excluded instruments were either specific to oral diseases other than dental caries or consisted of only a single item representing overall oral health.

The identified instruments all adopt frequency-type response options. Previous qualitative studies suggested that it is more relevant to include a response format related to the severity than the frequency of the caries-specific OHRQoL measure, as adolescents generally discussed caries symptoms in terms of severity rather than frequency (151). None of the above instruments can be easily transformed between different types of response levels. At this stage, it was decided that a CSPBM would be developed as a *de novo* measure based upon the above literature review and incorporating qualitative interviews and expert validation, a better option for the development of a new classification system (62, 150). Ethical approval for this study was obtained from the Human Research Ethics Committee, Griffith University, Australia (HREC/2018/822) **(Appendix A)**.

Literature search

The eight instruments identified by the previous search were used to determine possible domains, items and response levels for the new classification system. The items included in the relevant versions of original and short forms of these instruments were extracted to an Excel spreadsheet and categorised under the respective domain to form a large pool of domains and items. According to Sischo et al., OHRQoL is a multidimensional concept that includes oral health, functional, emotional and social wellbeing, and satisfaction with care (only for the treatment groups) (11). Therefore, oral health functional, emotional and social wellbeing were considered as main domains to categorise the items in each instrument. Two researchers (RH and SK) with dentistry expertise analysed and removed items irrelevant to dental caries from the list.

The pooled list was then analysed to identify the most frequently occurring items in the domains. PBMs usually include one item from each domain for ease of conducting the valuation exercise. Hence, the most commonly occurring item from each domain was included in the draft classification system. However, some domains may not be captured adequately with only a single item. PBMs such as AQoL-6D include few items per multidimensional domains (152). Therefore, when more than one relevant item occurred in a particular domain (e.g., items were differently worded in separate instruments or there was a potential to include more than one item per domain), the research team collaboratively identified the most suitable item/s and wordings to be included.

To explore any potentially omitted items, we further reviewed previous direct valuation studies for oral health outcomes. Standard clinical dentistry texts (153) were also referenced to identify clinical scenarios related to dental caries. Based on the findings, a draft classification system for the DCUI was developed.

Interviews with adolescents

Next, the draft classification system was refined based on consultation with adolescents, the target population. The aim of the interviews was to identify whether all relevant items were included in the descriptive system, whether all included items were important, and whether wordings were appropriate for the target group. Semi-structured interviews were conducted among a convenience sample of adolescents aged 12–17 years. By the age of 12 years, the mixed dentition period is over and, except for wisdom teeth, most children have all their permanent teeth. In Australia, free dental services are provided only up to 17 years of age. Therefore, 12–17 years was considered a suitable age range for this study.

The snowballing technique was used to recruit the sample and the initial two participants were recruited from a known group of adolescents from two different geographic locations in Queensland (Brisbane and Gold Coast), Australia. Adolescents aged 12–17 years who had active caries or had previous experience of caries and who could speak English were included in the interviews. Adolescents with oral diseases other than dental caries were excluded. Face-to-face interviews were conducted in a quiet and convenient location. Prior to the start of the interview, informed written consent was obtained from the adolescents and their parents, and demographic data, perceived oral health status and frequency of dental visits were recorded for each participant. An interviewer guide **(Text Box 3.1)**, developed based on the items and response levels

included in the draft classification system, provided direction for conducting the interviews.

The interviews were audio-recorded and transcribed non-verbatim. Each included item was considered as a potential theme for the analysis and codes were developed based on the adolescents' reasons for why or why not the included items were important. The participants' opinions were summarised based on these themes and codes. Included items were retained if the majority of respondents agreed that they were sufficiently important to remain in the classification system. However, participants' opinions summarised based on themes and codes were further explored to consider whether the participants has raised any special issues regarding the importance of the included items and the inclusion of specific wordings—even when a special issue was raised by only one participant. These were discussed with the research team to reach consensus on any changes to make the instrument more understandable to the target group.

Interviewer Guide

A. Perception

1. What do you think is meant by 'tooth decay'?
   In your view, what are the main problems associated with a decayed tooth?
   Interviewer: Please record the answers to this question in your notebook and use them to probe the participant's comments on each item of the whole instrument.

B. Draft classification system

I would like to request your opinion about a questionnaire we are planning to give to adolescents in your age group. There are no right or wrong answers. Please give your opinion on each part of the questionnaire.

2. Tooth location
   a) When you think about tooth health, is the location of the tooth important to you?
   b) If yes, what makes the location of the tooth important to you?
   c) Can you understand which teeth are front teeth and which teeth are back teeth?
   d) Would you say that whether or not the tooth is at the front or back is the best way to distinguish or describe location?
3. Pain/discomfort
   Do you think pain/discomfort is an important factor to be considered in tooth decay? Why/why not?
4. Difficult eating/drinking
   Do you think difficult eating/drinking is an important factor to be considered in tooth decay? Why/why not?
5. Worried or anxious about decayed tooth
   Do you think worry or anxiety is an important factor to be considered in tooth decay? Why/why not?
6. Able to join in activities (Playing, Sports, School activities)
   Do you think tooth decay can affect your ability to participate in activities like playing, sports and school activities? Why/why not?
7. Appearance
   a) In your opinion, is appearance an important issue when thinking about your teeth?
   b) Do you that think tooth decay has an impact on appearance?
8. Swelling
   Do you think swelling is an important factor to be considered in tooth decay? Why/Why not?

Interviewer: Please show cue cards 1–7 to the participant one at a time. These show each item and its response levels.

9. Would you please read this and tell me:
   a) Are these wordings easy to understand and clear to you?
   b) If they are not clear, which words are unclear to you? Can you suggest a better word?

Interviewer: Please show the whole instrument to the participant and ask them to read and comment on it.

10. Please would you read this and tell me:
    a) Do you think that adolescents in your age group will be able to understand these questions?
    b) This is all about tooth decay. So, do you think all important factors for tooth decay are included here or is there any other important factor we need to include?

Text box 3.1: Interviewer Guide

Expert validation

The refined draft classification system was then validated based on the consensus of a group of dental experts using a modified Delphi technique (154). The experts' opinions were sought regarding the relevance of the included items and response levels with respect to the QoL aspects of dental caries, appropriateness for the target group and clarity of the wordings. A covering letter, which described the purpose of the study, and a template to obtain expert comments was emailed to all experts in the group. Included items, response levels and wordings of the classification system were refined based on expert opinion after the first round. In the second round, the revised classification system and a summary of the first round comments were sent to all group members to obtain consensus on the finalised classification system.

Pre-test with adolescents

The finalised instrument was pre-tested with a convenience sample of another three adolescents aged 12–17 years, who had experience with dental caries. The readability of the instrument was assessed using the Flesch Kincaid Reading scale.

3.4 Results

The steps adopted to develop the classification system for the DCUI and the changes to the draft classification system at each stage of the development are illustrated in **Figure 3.1**.

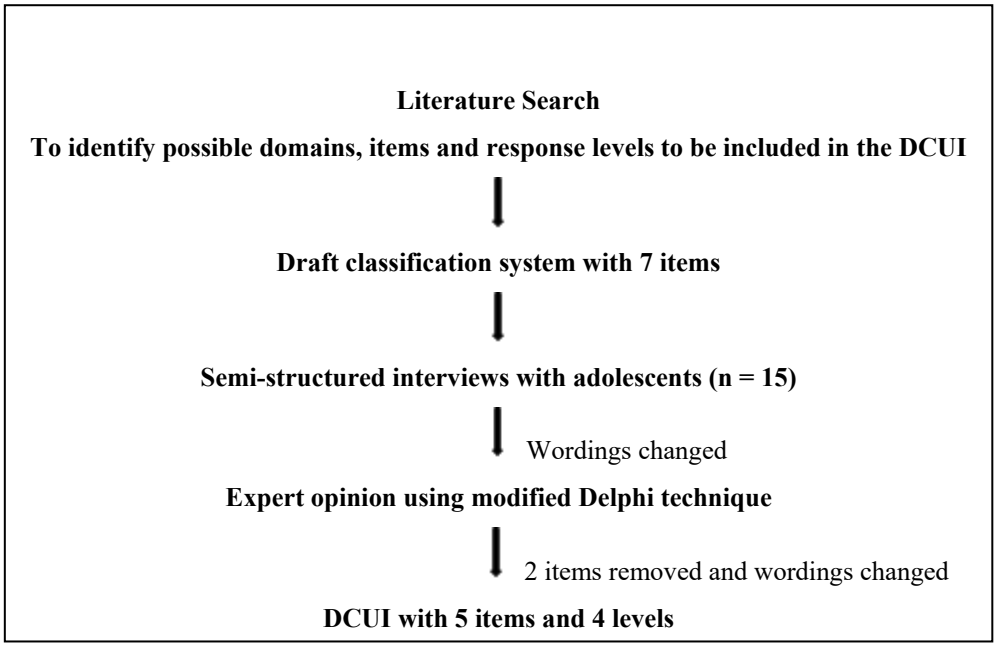


Figure 3.1: Steps to develop a classification system for the Dental Caries Utility Index (DCUI)

Literature search

A large pool of domains and items was constructed from the eight non-PBM instruments **(Table 3.1)**. The pooled list revealed that, except for two single-domain instruments (C-OIDP and SOHO-5), all other instruments mainly included four domains, namely: oral health, functional, social and emotional wellbeing. Further, the items included in the two single-domain instruments were still relevant for the four main domains of interest. Thus, these non-PBM instruments reflected the same construct of the OHRQoL described by Sischo et al. (11) and each of these four domains contained more than one relevant item that could be included in the classification system. Therefore, after discussions among the research team, the best possible item and wordings were identified for selection. For example, the social domain included items related to school activities and participation in other social activities like playing, clubs, et cetera. The research team selected 'able to join in activities' as the optimal item for the social domain. Eventually, four items (one per domain) were identified: oral health – pain/discomfort; functional – difficulty eating foods/drinking; emotional – worried/anxious about tooth and social – able to join in activities. In addition to these, the item 'appearance' with different wordings was included in all eight instruments, either in emotional or social wellbeing domains (e.g., 'Felt that you look different' in the COHIP emotional domain and 'Unhappy with looks 'in the POQL social domain) or as an item in the single-domain

instruments. Thus, the research team identified that ‘appearance’ was an important item to be included in the classification system.

The recall periods for the eight instruments are ‘at the moment’, ‘last 7 days’, ‘past 4 weeks’, ‘past 3 months’ and ‘entire life span’ (148). The options ‘past 4 weeks’ and ‘past 3 months’ are used for the instruments developed for people above five years of age. Considering that recall periods in well-established PBMs range from ‘today’ to ‘past 4 weeks’ (52), and given dental caries are a chronic condition, ‘past 4 weeks’ was selected as the recall period for the draft classification system. Regarding the response levels, as mentioned in the Methods section, severity levels were applied in response scale (rather than frequency levels), as suggested by previous studies (151). The wordings for the response levels of each item were revised with respect to well-established generic PBMs such as EQ-5D-Y (149) and CHU9D (150). A four-level response option was selected, as the evidence suggested that four to seven response options would be optimal to improve the psychometric properties of the scale (155).

Table 3.1: The pool of domains and items extracted from the non-preference-based oral health-related quality-of-life instruments

Instrument	No. domains	No. items	Domains				
			Oral Health	**Functional**	**Emotional**	**Social**	**Other**
COHIP (114)	5	34	**Oral Health Well-being (10 items)** Had pain in your teeth/toothache Had food sticking in or between your teeth Had discoloured teeth or spots on your teeth Had pain or sensitivity in teeth with hot or cold things *Had sores or sore spots in or around your mouth* *Had bad breath* *Had bleeding gums* *Been breathing through your mouth or snoring* *Had crooked teeth or spaces between your teeth* *Had dry mouth or lips*	**Functional Well-being (6 items)** Had difficulty eating foods you would like to eat Had trouble biting off or chewing foods Had trouble sleeping Had difficulty keeping your teeth clean *Had people have difficulty understanding what you were saying* *Had difficultly saying certain words*	**Social/Emotional Well-being (8 items)** Felt worried or anxious Been unhappy or sad Been worried about what other people think about your teeth and mouth Felt that you look different Avoided smiling or laughing with other children Felt shy or withdrawn Been teased, bullied, or called names by other children Been upset or uncomfortable with being asked questions about your teeth	**Self-image (6 items)** Been confident Felt that you were attractive (good looking) I have good teeth I feel good about myself When I am older, I believe (think) that I will have good teeth When I am older, I believe (think) that I will have good health **School Environment (4 items)** Missed school for any reason Had difficulty paying attention in school Not wanted to speak/read aloud in class Not wanted to go to school	

COHIP SF-19 (118)		3	19	**Oral Health Well-being (5 items)** Had pain in your teeth/toothache Had discoloured teeth or spots on your teeth *Had crooked teeth or spaces between your teeth* *Had bad breath* *Had bleeding gums*	**Functional Well-being (4 items)** Had difficulty eating foods you would like to eat Had trouble sleeping Had difficulty keeping your teeth clean *Had difficultly saying certain words*	**Social/Emotional Well-being (6 items)** Been unhappy or sad Felt worried or anxious Avoided smiling or laughing with other children Felt that you look different Been worried about what other people think about your teeth Been teased, bullied, or called names by other children	**Self-Image (2 items)** Been confident Felt that you were attractive **School Environment (2 items)** Missed school for any reason Not wanted to speak/read out loud in class
CPQ (120)	**8-10**	4	25	**Oral Symptoms (5 items)** Pain in your teeth or mouth *Sore spots in your mouth* Pain in your teeth when you drink cold drinks or eat foods Food stuck in your teeth *Bad breath*	**Functional Limitations (5 items)** Needed longer time than others to eat your meal Had a hard time biting or chewing food like apples, corn on the cob or steak Had trouble eating foods you would like to eat Had trouble saying some words Had a problem sleeping at night	**Emotional well-being (5 items)** Been upset Been concerned what other people think about your teeth or mouth Worried that you are not as good-looking as others Felt frustrated Been shy	**Social well-being (10 items)** Missed school Had a hard time doing your homework Had a hard time paying attention in school Not wanted to speak or read out loud in class Tried not to smile or laugh when with other children Not wanted to talk to other children

						Not wanted to be with other children Stayed away from activities like sports and clubs Other children teased you or called you names Other children asked you questions about your teeth or mouth
CPQ 11-14 (119)	4	37	**Oral symptoms (6 items)** Pain in teeth, lips, jaws or mouth *Bleeding gums* *Mouth sores* *Bad breath* Food caught in or between teeth Food stuck to roof of mouth	**Functional Limitations (9 items)** Breathing trough mouth Taken longer than others to eat a meal Trouble sleeping Difficulty to bite or chew food like apples, corn on the cob or steak *Difficulty to open mouth wide* *Difficult to say any words* Difficult to eat foods you would like to eat *Difficult to drink with a straw* Difficult to drink or eat hot or cold foods	**Emotional Well-being (9 items)** Irritable or frustrated Unsure of them self Shy or embarrassed Concerned what other people think about you Worried that you are less attractive than other people Upset Nervous or afraid Worried that you are less healthy than other people Worried that you are different than other people	**Social Well-being (13 items)** Missed school Hard time paying attention in school Difficulty doing homework Avoiding speaking or reading aloud in class Avoiding activities like sports, clubs, drama, music, school trips Avoiding talking to other children Avoiding smiling or laughing when around other children *Difficulty playing a musical instrument, such*

						as a recorder, flute, clarinet, trumpet Avoiding to spend time with other children Argued with other children or family Teased or called names by other children Left out by other children Asked questions by other children
CPQ 11-14 Short Forms (121)	4	8 & 16 versions	**Oral symptoms** Pain in teeth/mouth *Bad breath* *Mouth sores* Food caught between teeth	**Functional Limitations** Difficulty eating/drinking hot/cold foods Difficulty chewing firm foods Difficulty saying words Taken longer to eat a meal Trouble sleeping	**Emotional well-being** Upset Felt irritable/frustrated Felt shy Concerned what people think about your teeth/mouth	**Social well-being** Asked questions Teased/called names Avoided smiling/laughing Argued with children/family Not wanted to speak/read loud in class
C-OIDP (108)	1	8	Eating Speaking Emotion Smiling Study Social contact Cleaning mouth Sleeping			

POQL (116)	4	10	**Physical** Pain Eating food (hard/hot/cold)	**Role** Pay attention in school Miss school	**Emotional** Angry or upset Feel worried Cry	**Social** Not smile/laugh Worry less attractive Unhappy with looks	
TOQOL (117)	5	16	**Oral Problems** *Bad breath* *Food caught between teeth* *Bleeding gums* *Mouth sores*	**Physical Functioning** Pain Eat hot/cold/hard Difficult to eat **Role functioning** Pay attention Sleep Miss school	**Emotional Functioning** Angry/upset/worry Depressed	**Social Functioning** Not smile/laugh Unhappy with looks Worry less attractive	
ECOHIS (115)	6	13	**Child Symptoms** Pain in the teeth, mouth or jaws	**Child Function** Had difficulty drinking hot or cold beverages Had difficulty eating some foods Had difficulty pronouncing any words Missed preschool, day care or school	**Child Psychological** Had trouble sleeping Been irritable or frustrated	**Child Self-image/Social Interaction** Avoided smiling or laughing when around other children Avoided talking with other children	**Parent Distress** Been upset Felt guilty **Family function** Taken time off from work Financial impact on your family
Michigan OHRQoL Original	3	7	**Pain/discomfort** Teeth hurt now	**Functional** Difficult for you to chew	**Psychological** Are you happy with your teeth?		

Child Version (125)			Teeth hurt you in the last days Teeth hurt when you eat something hot or cold	Difficult for you to bite	Do you have a nice smile?	
Michigan OHRQoL Modified Child version (125)	4	9	**Pain/discomfort** Teeth hurt now Teeth hurt when you eat something hot or cold Teeth hurt when you eat something sweet	**Functional** Is it hard for you to chew and bite? Does a hurting tooth wake you up at night?	**Psychological** Do you like your teeth? Are you happy with your teeth and smile?	**Social** Does a hurting tooth stop you from playing? Do kids make fun of your teeth?
SOHO-5 (124)	1	7	Difficulty eating Difficulty drinking Difficulty speaking Difficulty playing Avoiding smiling (due to pain) Avoiding smiling (due to appearance) Difficulty sleeping			

Non relevant items for dental caries are in italics

COHIP - Child Oral Health Impact Profile; COHIP-SF 19 - Child Oral Health Impact Profile-Reduced; C-OIDP - Child Oral Impacts on Daily Performances; ECOHIS - Early Childhood Oral Health Impact Scale; CPQ 11-14 - Child Perceptions Questionnaire for 11 to 14 years; CPQ 8-10 - Child Perceptions Questionnaire for 8 to 10 years; CPQ11-14 short - Short forms of CPQ11-14 (16 and 8 items); P-CPQ - Parental-Caregiver Perceptions Questionnaire for 6 to 14 years old children; POQL - Paediatric Oral Health-related Quality of life Questionnaire; SOHO-5 - Scale of Oral Health Outcomes for 5-year-old children; TOQOL - Teen Oral Health-Related Quality of Life instrument

A further review of previous studies using a direct valuation approach and clinical dentistry books (153) indicated that the location of the tooth, appearance and swelling are important factors for deciding treatment preferences for dental caries **(Table 3.2)**. The item 'appearance' was also identified from the item pool derived from the non-PBM oral health-related QoL measures. This literature search identified three additional items, resulting a draft classification system with seven items **(Figure 3.2)**, which were presented to the adolescents in semi-structured interviews.

Table 3.2: Possible domains and levels to be included in the descriptive system based on the literature on preference elicitation studies generating utility values for oral health outcomes

Reference Study	Preference Elicitation Method and Dental condition	Attributes/Scenario	Remarks
Fyffe & Kay, 1992 (156)	SG - dental caries	Only posterior teeth are considered A decayed and painful posterior tooth A decayed and non-painful posterior tooth A posterior tooth that had been restored and would need further restorative treatment A permanently restored posterior tooth	For SG, sound tooth was considered the best health state and a tooth needing immediate extraction was considered the worst health state.
Fyffe et al., 1999 (71)	TTO and VAS among adolescent sample - dental caries	Scenarios for dental health states Painful decayed front tooth Painless decayed front tooth Missing back tooth Missing front tooth Healthy back tooth Healthy front tooth Painful decayed back tooth Painless decayed back tooth Back tooth with a silver filling Front tooth with white filling Back tooth with a fissure sealant Broken front tooth	Utility values for 12 dental health states using dental free time trade-off and dental visual analogue scale
Espelid et al., 2006 (157)	WTP - restorative material	Appearance (of the restorative material) Not tooth-coloured, highly visible Tooth-coloured, but visible Tooth-coloured and not visible	
Fukai et al., 2012 (158)	VAS for dental health states and TTO for interval of willingness to have dental check-ups	Painless decayed front tooth Painful decayed front tooth Aesthetically filled front tooth Missing front tooth Painless decayed back tooth Painful decayed back tooth Metal filled back tooth	

		missing back tooth	
Augusti et al., 2014 (159)	WTP - Dental treatment	Location (Anterior/posterior)	WTP were elicited for two treatments options to restore a single-tooth gap (anterior and posterior). Anterior area showing a higher WTP index.
Lord et al., 2015 (75)	DCE- Dental Caries	Location Anterior teeth, Pre-molar teeth and Molar teeth Levels for each attribute No problem Decay without pain Decay with pain Tooth needs to be removed	Attribute- based on a study Kay et al. 2014, which found using VAS, that the different values were given to individual teeth and these differences were greatest between anterior (front), pre-molar and molar teeth. Levels- Oral health problems including decay and removal/missing teeth were identified as levels. Following consultation with a clinical expert to refine the level indicating decay to specify whether the decay was associated with or without pain, as these would be likely to affect preferences.

SG - Standard gamble; TTO - Time trade-off; VAS - Visual analogue scale; WTP - Willingness to pay; DCE - Discrete choice experiment

Think of the decayed tooth you are requiring treatment for or had treated during your last visit to the Dental clinic. Please tick one box from each category, which best describes what you have experienced during past 4 weeks due to decayed tooth.

Pain/Discomfort
1. I have no pain or discomfort
2. I have a little pain or discomfort
3. I have quite a lot of pain or discomfort
4. I have lots of pain or discomfort

Difficult eating foods/drinking
1. I have no difficulty in eating food/drinking
2. I have a little difficulty in eating food/drinking
3. I have quite a lot of difficulty in eating food/drinking
4. I have lots of difficulty in eating food/drinking

Worried/anxious about tooth
1. I am not worried/anxious about my tooth
2. I am a little bit worried/anxious about my tooth
3. I am quite worried/anxious about my tooth
4. I am very worried/anxious about my tooth

Able to join in activities (Playing, Sports, School activities)
1. My tooth decay causes no difficulty in join in activities
2. My tooth decay causes a little difficulty in join in activities
3. My tooth decay causes a quite a lot of difficulty in join in activities
4. My tooth decay causes lots of difficulty in join in activities

Location of tooth cavity
1. Front tooth
2. Back tooth

Appearance of my decayed/filled/missing tooth
1. Natural tooth appearance
2. Slightly noticeable tooth cavity/filled tooth
3. Highly noticeable tooth cavity/filled tooth
4. Missing tooth

Swelling
1. I have no swelling associated with decayed tooth/filled tooth
2. I have mild swelling associated with decayed tooth/filled tooth
3. I have a large swelling associated with decayed tooth/filled tooth
4. I have a very big swelling associated with decayed tooth/filled tooth

Figure 3.2 Draft classification system based on literature search

Interviews with adolescents

Fifteen adolescents participated in the semi-structured interviews. The majority (n = 12) were 12–14 years and the group was well balanced in gender. The sample included adolescents whose first language is not English to increase the relevance of the instrument in a culturally and linguistically diverse country such as Australia (160) (Table 3.3). The average duration for the interviews ranged from 15 to 20 minutes.

Table 3.3: Characteristics of the adolescent study sample

Characteristic	Number (n=15)
1. Age	
12–14 years	12
15–17 years	3
2. Gender	
Male	8
Female	7
3. Perceived current oral health status	
Good	12
Neither good nor poor	3
Poor	0
4. Frequency of dental visits	
Once every 6 months	11
When I have oral health problems	4

During the first stage of the interview, the adolescents were asked about tooth decay and they identified a decayed tooth as a rotten tooth, bad tooth or unhealthy tooth. Participants described a decayed tooth as:

> *A tooth that is not in good health, broken down because of build-up of germs in one's mouth. (Male, aged 15 years)*

> *A tooth that is rotten and damaged. It would cause a lot of toothache and pain to a person. (Male, aged 17 years)*

However, during the interviews, it was noted that some adolescents, especially the younger participants, were more familiar with the terms 'tooth cavity' or 'rotten teeth' than the terms 'decayed' or 'carious'. Therefore, the term 'decayed tooth' in the draft classification system was replaced by 'tooth cavity'. According to this group of adolescents, toothache, pain upon biting, pain in the mouth and appearance are the main problems associated with a decayed tooth.

More than 60% of the participants identified all included items in the draft classification system as important factors in dental caries [pain/discomfort (n = 15), difficulty eating foods/drinking (n = 14), worried/anxious about decayed tooth (n = 12), able to join in activities (n = 9), location (n = 11), appearance (n = 14) and swelling (n = 11)]. Participants identified 'pain/discomfort' and 'difficulty eating/drinking' as important factors in tooth decay, as these would indicate the severity of the dental caries and the need to seek dental care:

If my tooth is causing discomfort or pain to me, I know that I have to go to a dentist and describe my pain. (Female, aged 14 years)

Although the majority of participants identified 'worried/anxious about tooth' as an important factor, there was concern for the extremity and potential medical diagnosis of anxiety. For example, a 12-year old girl said that,

I will be worried but I wouldn't be anxious. (Female, aged 12 years)

As this was the opinion of a single participant, the research team further discussed this; to avoid confusion by using both words as a single item, 'anxious' was removed and the item was reworded as 'worried about tooth cavity'.

The adolescents' opinions of the items 'able to join in activities', 'location', 'appearance' and 'swelling' yielded possible interactions with other items in the classification system. Among 15 participants, only nine identified 'able to join in activities' as an important factor to be considered. However, both groups of participants who did and did not consider 'able to join in activities' an important factor related it to the pain:

Yes, the pain will interrupt concentration. (Male, aged 15 years)

If you are in pain, you can't do your best. (Female, aged 14 years)

Not a problem unless your tooth hurts a lot (Female, aged 12 years)

However, a 15-year old participant related this to both pain and appearance as:

Pain, appearance all affect joining in activities. (Male, aged 15 years)

Participants considered the 'location' of teeth an important factor, as it affects appearance or eating ability, depending on whether the tooth decay is in a front or back tooth:

If it is one of my front teeth, it will affect my smile. (Female, aged 12 years)

Eating food could be difficult if the rotten tooth is at the back of your mouth. (Male, aged 17 years)

Considering the item 'Appearance', the majority held the opinion that the tooth decay has an impact on appearance, especially when the decay is in an obvious location:

I think if it is decaying in an obvious spot then it has bad appearance (Female, aged 14 years)

The participants also expressed that ‘swelling’ would be important, as it would cause pain and affect appearance:

If it is a big swelling, it will be ugly and painful. (Female, aged 14 years)

Although these opinions indicate possible interactions with other items in the classification system, they are mixed; therefore, these four items were retained in the classification system until the expert opinion stage while more supportive evidence regarding their inclusion or exclusion was obtained. A summary of adolescents’ opinions were given in **Appendix B**. Following completion of the adolescents’ interviews, the revised instrument consisted of seven items, with only the wording of items changed as described above **(Figure 3.3)**.

Think of the tooth cavity you are requiring treatment for or had treated during your last visit to the Dental clinic. Please tick one box from each category, which best describes what you have experienced during past 4 weeks due to tooth cavity.

Pain/Discomfort
1. I have no pain or discomfort
2. I have a little pain or discomfort
3. I have quite a lot of pain or discomfort
4. I have a lots of pain or discomfort

Difficulty eating foods/drinking
1. I have no difficulty in eating foods/drinking
2. I have a little difficulty in eating foods/drinking
3. I have quite a lot of difficulty in eating foods/drinking
4. I have lots of difficulty in eating foods/drinking

Worried about tooth
1. I am not worried about my tooth
2. I am a little bit worried about my tooth
3. I am quite worried about my tooth
4. I am very worried about my tooth

Able to join in activities (Playing, Sports, School activities)
1. My tooth cavity causes no difficulty in join in activities
2. My tooth cavity causes a little difficulty in join in activities
3. My tooth cavity causes a quite a lot of difficulty in join in activities
4. My tooth cavity causes lots of difficulty in join in activities

Location of tooth cavity
1. Front tooth
2. Back tooth

Appearance of my cavitated/filled/missing tooth
1. Natural tooth appearance
2. Slightly noticeable tooth cavity/filled tooth
3. Highly noticeable tooth cavity/filled tooth
4. Missing tooth

Swelling
1. I have no swelling associated with tooth cavity/filled tooth
2. I have mild swelling associated with tooth cavity/filled tooth
3. I have a large swelling associated with tooth cavity/filled tooth
4. I have a very big swelling associated with tooth cavity/filled tooth

Figure 3.3: Draft classification system revised with adolescents' opinions

Expert validation

The revised draft instrument was then sent to ten dental experts, six of whom agreed to participate. The group of experts consisted of two senior academics and research fellows in dental public health, two clinical dental educators, a general dental practitioner and a dental therapist. These experts advised that the general description provided at the beginning of the instrument should be clearer and emphasised the necessity of reiterating that the questions are related to dental caries and not problems caused by other factors,

such as gingival bleeding or tooth crowding. Therefore, the introductory description was reworded as: *'Please tick one box from each category, which best describes your situation over the past 4 weeks due to a tooth cavity, a filling or missing tooth'.*

The experts also made a number of comments and suggestions on the items of the classification system and the wording of the items. All experts agreed that the items 'pain/discomfort', 'difficulty eating foods/drinking', 'worried', 'able to join in activities' and 'appearance' are important and should be included in the classification system. One expert noted that combining pain and discomfort as one item was potentially problematic, since pain could have a severe impact on QoL while discomfort might not. This is a subjective term and could be interpreted differently (i.e., it might not cause actual pain, but could still feel 'rough' or 'wrong' in some way to participants who might then respond 'yes'). As this issue was raised by only one expert, it was discussed by the research team. Given that the combination of pain and discomfort is consistent with other generic PBMs for adolescents, such as the EQ-5D-Y (149), it was left unmodified as 'pain/discomfort'. The experts stated the item 'worried about my tooth cavity' was generic and that people could be 'worried about [a] tooth' for various reasons, such as being 'worried about losing the tooth' in addition to concerns about pain and appearance. Therefore, this item was edited to 'worried' and an example was provided in the classification system. Although adolescents had mixed opinions regarding the item 'able to join in activities', all experts agreed that it should be included in the classification system. Further, considering that dental caries affect school activities, as evidenced by previous studies (14), that they are an important item in OHRQoL instruments, 'able to join in activities' was retained in the classification system but reworded to 'ability to participate in activities', as suggested by the expert panel. All experts agreed that 'appearance' is an important item, especially for the target group, who would have significant concerns about aesthetics and physical appearance. However, the panel stated that the wordings for the item and the levels identified from the literature search would be confusing and may be unclear to the participants. Therefore, the wording of the first level was changed to 'I am not concerned about my appearance' and other levels were adjusted accordingly to clarify the QoL aspect of appearance.

Four of the six experts were concerned about two included items: 'tooth location' and 'swelling'. These experts stated that 'tooth location' may cause ambiguity if respondents had recent cavities or fillings on both front and back teeth; further, problems with front top teeth may have significantly different impact compared to problems in front

bottom teeth. The experts also added that 'tooth location' could be an important factor to be considered for treatment preferences but that is not an item related to QoL and that the functional form of 'location' was already included in the classification system as 'appearance'. The experts also suggested that the swelling associated with dental caries is uncommon, since few adolescents with caries experience the pulpal involvement that is associated with swelling. They also noted that the significance and implications of 'swelling' were already determined based on the individual answers in regards to the 'appearance', 'activity', 'pain/discomfort' and 'difficulty eating foods/drinking' items. Considering these opinions, and after discussion with the research team, 'tooth location' and 'swelling' were removed from the classification system. A summary of experts' opinions were given in the **Appendix C**.

Based on the expert opinions during the first round, the draft classification system with seven items was refined, which resulted in a classification system of five items. The revised system was again sent to the six experts for their final comments and consensus.

Pre-test with adolescents

The finalised instrument **(Figure 3.4)** was pre-tested by three adolescents. The Flesch Kincaid Reading score was 64.6, which indicates that the new classification system can be easily understood by adolescents aged 12–13 years.

Please tick one box from each category, which best describes your situation over the past 4 weeks due to a tooth cavity, a filling or missing tooth.

Pain/Discomfort
1. I have no pain or discomfort
2. I have a little pain or discomfort
3. I have quite a lot of pain or discomfort
4. I have lots of pain or discomfort

Difficulty eating food /drinking
1. I have no difficulty in eating food/drinking
2. I have a little difficulty in eating food/drinking
3. I have quite a lot of difficulty in eating food/drinking
4. I have lots of difficulty in eating food/drinking

Worried ***(e.g. about losing a tooth, etc.)***
1. I am not worried
2. I am a little bit worried
3. I am quite worried
4. I am very worried

Ability to participate in activities ***(e.g. playing with your friends, sports, school work, etc.)***
1. I have no difficulty participating in activities
2. I have a little difficulty participating in activities
3. I have quite a lot of difficulty participating in activities
4. I have lots of difficulty participating in activities

Appearance
1. I am not concerned about my appearance
2. I am a little concerned about my appearance
3. I am quite concerned about my appearance
4. I am very concerned about my appearance

Figure 3.4: Classification system for new preference-based quality-of-life measure for dental caries among adolescents (Dental Caries Utility Index - DCUI)

3.5 Discussion

To our knowledge, this is the first study to develop a health classification system for dental caries. The new DCUI classification system includes five items: 'pain/discomfort', 'difficulty in eating foods/drinking', 'worried', 'ability to participate in activities' and 'appearance', all of which are commonly used in other non-PBM OHRQoL instruments. A valuation study will be conducted based on the new classification system to generate a tariff (preference-based scoring algorithm) to enable the calculation of health state utility values for future economic evaluations of oral health interventions. Since dental caries is the most prevalent childhood oral disease and highly preventable, this new CSPBM will facilitate better assessment of the impact of oral healthcare services, may improve the decision-making process and have potential evidence-based policy implications.

The classification system for the DCUI was developed based on well-established methodology (62). Although the CSPBM can be developed from an existing non-PBM, it often faces methodological challenges, especially when non-PBMs are complex and do not have a clear multidimensional structure (62). In this study, the new CSPBM was developed by combining a comprehensive literature review, qualitative research with patients and expert opinions. In particular, qualitative interviews with the target patient group (adolescents with dental caries) and consultation with experts in clinical and public health dentistry ensured the necessary face and content validity of the instrument (60). Brazier et al. (62) revealed that 10 of 26 studies developed descriptive systems for CSPBMs using these methodologies either alone or in combination.

This study also has certain limitations. The draft classification system was developed based on the domains and items identified from non-PBM OHRQoL instruments; thus, limitations associated with the identification of relevant items for those instruments would also potentially affect the present study. However, this study gathered information from all relevant existing instruments and was not restricted to a single measure to identify the most suitable items to be included. In addition, the qualitative studies provide reassurance that the most important items have been included. The qualitative interviews were only conducted with adolescents in a certain geographic area, another potential limitation. However, geographic variation is unlikely to significantly affect the identification of the most relevant items. Nevertheless, a cross-cultural validation analysis should be conducted if this new classification will be used in other countries. Although the classification system mainly focused on dental caries, further studies could evaluate the use of this new system more broadly for oral health conditions other than the dental caries as well as for an adult population.

Conclusions

The present paper describes the initial development of a classification system for dental caries to be used among adolescents. The availability of a paediatric CSPBM for dental caries would facilitate better evaluation of oral healthcare interventions compared to generic PBMs and thereby, support improved resource allocation through the use of CUA. Subsequent studies are planned to develop a preference-based scoring algorithm for DCUI and to empirically assess the psychometric properties of the instrument.

Chapter 4. Valuation study for a preference-based quality-of-life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents - Study Protocol

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a submitted co-authored paper. The bibliographic details of the co-authored paper, including all authors, are:

Hettiarachchi RM, Kularatna S, Byrnes J, Mulhern B, Chen G, Scuffham PA. Valuation study for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents - Study Protocol. BMJ Open 2020; 10:e038626. https://10.1136/bmjopen-2020-038626

My contribution to the paper involved the designing the study, planning data collection and data analysis, and drafting the article.

(Signed) _____ (Date)_____

Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Supervisor: Prof. Paul Scuffham

Valuation study for a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents - Study Protocol

Ruvini M. Hettiarachchi, Sanjeewa Kularatna, Joshua Byrnes, Gang Chen, Brendan Mulhern, Paul A. Scuffham

4.1 Abstract

Introduction

A new health state classification system has been developed for dental caries - Dental Caries Utility Index (DCUI) to facilitate the assessment of oral health interventions in the cost-utility analysis (CUA). This paper reports the protocol for a valuation study, which aims to generate a preference-based algorithm for the classification system for the DCUI.

Methods and analysis

Discrete choice experiments (DCEs) will be conducted to value health states generated by the DCUI classification system and preferences for these health states will be modelled to develop a utility algorithm. DCEs produce utility values on a latent scale and these values will be anchored into the full health-dead scale to calculate the quality-adjusted life years (QALYs) in CUA. There is no previous evidence for the most suitable anchoring method for dental caries health state valuation. Hence, we will first conduct pilot studies with two anchoring approaches; DCE including duration attribute (DCE_{TTO}) and DCE anchoring to worst heath state in visual analogue scale. Based on the pilot studies, the most suitable anchoring method among two approaches will be used in the main valuation survey, which will be conducted as an online survey among an age and sex representative sample of 2000 adults from the general population of Australia. Participants will be asked to complete a set of DCE choice tasks along with anchoring tasks, basic social-demographic questions, DCUI, a generic preference-based measure (EQ-5D-5L) and an oral health quality of life instrument (OHIP-14).

Ethics and dissemination

Ethical approval for this study was obtained from the Human Research Ethics Committee, Griffith University (reference number HREC/2019/550). The generated algorithm will facilitate the use of the new dental caries preference-based measure in economic evaluations of oral health interventions. The results will be disseminated through journal articles and professional conferences.

Key words: Dental Caries, Adolescents, Valuation, Discrete choice experiments, Preference-based

Article Summary

Strengths and limitations of this study

- The protocol has been developed for a valuation study, which aims to generate a preference-based utility algorithm for a new health state classification system for dental caries.
- Since there is no previous evidence for the most suitable anchoring method for dental caries health state valuation, pilot studies with two anchoring approaches (DCE$_{TTO}$ and DCE anchoring with VAS) have been planned prior to the main valuation survey.
- The most suitable valuation approach identified based on the pilot studies will be applied as the main valuation survey to generate the utility algorithm.
- The generated algorithm will facilitate the use of the new dental caries preference-based measure in economic evaluations of dental caries interventions.
- Health states defined by the classification system will be valued by the adult general population sample. The methodological constraints associated with conducting health state valuation studies among the paediatric population, limit the ability to value health states with an adolescents sample.

4.2 Introduction

Health economic evaluations play a vital role in assessing health care interventions by providing information on which interventions provides the best value for money (19, 22). Cost-utility analysis (CUA) is a preferred type of health economic evaluation by many Health Technology Assessment authorities across the world (39,

161). CUA compares interventions in terms of their incremental cost per unit of outcome (44) and quality-adjusted life years (QALYs) is the commonest form of outcome measure in CUA (44). The QALY incorporates both quantity and quality of life as a single summary outcome measure. The use of QALY as a summary outcome measure allows CUA to compare health interventions across different diseases (44).

The calculation of QALYs relies on health state utility values (preference weights) assigned to the health states in the condition of interest (162). Preference-based quality of life measures (PBMs) are used to calculate utility values for QALYs in economic evaluations. PBMs are patient-reported outcome measures that consist of a health state classification system and a set of health state utility values corresponding to each of the health states defined by the classification system (60, 61). They are pre-scored, readily available and easy to use rather than the directly eliciting preferences from patients (110). There are two types of PBMs. Generic PBMs can be used for any health condition whereas condition-specific PBMs are tailored for use among patients with a particular disease or condition of interest (62). Generic PBMs such as the EQ-5D (163) are widely used; however, they may not be sensitive to the changes in some disease conditions since they do not include all relevant dimensions for each disease (62). Condition-specific PBMs typically include the dimensions more relevant to a particular disease or condition, thus they may be more responsive to the changes of the disease/ condition over time (61).

Several oral health-related quality of life (OHRQoL) instruments have been developed and validated to date (43, 148). However, none of them are preference-based, thus cannot be used to calculate utility weights for use in health economic evaluations (148). Due to the absence of a condition-specific PBM for oral health, researchers are limited to either elicit oral health utility scores via direct valuation methods (48) and/or use a generic PBM measure (97) in oral health interventions (147). Moreover, children and adolescents are the main focus of publicly funded oral health care services in many countries (108). They have a different perception about the impact of oral disease on their quality of life compared to adults. Therefore, the availability of paediatric condition-specific PBM for oral health will provide better information on how the disease and the oral health interventions affect the target group children and adolescents and will facilitate the use of CUA in assessing oral health care interventions more effectively. Furthermore, the majority of oral health care interventions among children and adolescents focus on dental caries since it is the most common chronic childhood oral disease (22). The

development of a paediatric condition-specific PBM for dental caries is an important area of research in oral health.

Hence, in the first phase of this study, we developed a classification system for dental caries - Dental Caries Utility Index (DCUI) (164) based on a review of the literature, a series of qualitative interviews with adolescents and with expert opinion; these are accepted methodologies to develop a classification system for a CSPBM (60, 62). The DCUI consisted of five items; pain/discomfort, difficulty eating food/drinking, worried, able to join in activities, and appearance, with each item consisted of four levels **(Table 4.1)**. Considering that the mixed dentition period is over by the age of 12 years and that those younger than 12 have less cognitive ability to understand the concepts and terms, we included adolescents above 12 years of age in the development of the classification system. Further, the Flesch Kincaid Reading score of the finalised instrument was 64.6, indicating that adolescents aged 12–13 years can easily understand the classification system. Thus, the target group of this instrument is adolescents above 12 years of age. This paper reports the protocol for a valuation study, which aims to generate a preference-based algorithm for the classification system for the DCUI. To do this, preferences for sets of selected heath states will be elicited that are then modelled to estimate weights for each attribute's level to develop a utility algorithm. The algorithm will facilitate the use of DCUI in health economic evaluation of dental caries interventions among children and adolescents.

4.3 Methods and plan of analysis

Preference elicitation technique

Different preference elicitation techniques have been adopted to elicit preference weights. The cardinal preference techniques such as standard gamble (SG) and time trade-off (TTO) produce utility values anchored by full health and death (55). However, over the recent past, ordinal preference elicitation methods such as discrete choice experiments (DCEs) and ranking orders have become widely used in health state valuations (55). A recent systematic review identified 63 health state valuation studies using DCEs and of them, 36 were published during 2016 to 2018 (65). In a DCE study, participants are requested to state their preference for the series of choices between two or more alternative scenarios describing health problems (75). Best-worst scaling (BWS) is a ranking approach, in which participants are asked to state the best and worst from

typically three or more sets of items or profiles (57). Compared to the traditional valuation methods such as SG and TTO (55), DCEs and BWS are typically conducted without an interviewer and compatible with the online surveys that expedite the data collection process (64). Compared to DCEs, BWS provides additional information related to worst preferences; however, previous studies demonstrated that DCEs performed better than BWS in health state valuations (165). Ordinal preference elicitation methods such as DCE and BWS produce utility values on a latent scale; thus, it is important to anchor the utilities generated onto the full health-dead scale to calculate the QALYs (66). Thus, health state valuation studies using DCE or BWS must include anchoring tasks. In the DCE approach, options such as DCE with duration (DCE_{TTO}) are available and can be used as a standalone valuation approach to test anchoring within the task. Considering all these facts, the DCE approach will be used as the preference elicitation technique for the present study.

Table 4.1: Dental Caries Utility Index (DCUI) Classification System*

Dimension	Description
Pain/Discomfort	1. I have no pain or discomfort 2. I have a little pain or discomfort 3. I have quite a lot of pain or discomfort 4. I have lots of pain or discomfort
Difficulty eating food/drinking	1. I have no difficulty in eating food/drinking 2. I have a little difficulty in eating food/drinking 3. I have quite a lot of difficulty in eating food/drinking 4. I have lots of difficulty in eating food/drinking
Worried ***(e.g., about losing a tooth, etc.)***	1. I am not worried 2. I am a little bit worried 3. I am quite worried 4. I am very worried
Ability to participate in activities ***(e.g. playing with your friends, sports, schoolwork, etc.)***	1. I have no difficulty participating in activities 2. I have a little difficulty participating in activities 3. I have quite a lot of difficulty participating in activities 4. I have lots of difficulty participating in activities
Appearance	1. I am not concerned about my appearance 2. I am a little concerned about my appearance 3. I am quite concerned about my appearance 4. I am very concerned about my appearance

**Hettiarachchi et al.(164)*

Anchoring methods

Different methods such as including duration as an additional attribute (DCE_{TTO}) (64), mapping DCE into TTO, using hybrid models for DCE and TTO, anchoring the worst state using TTO (66), including immediate death in pairwise comparisons(66) and anchoring with visual analogue scale (VAS) have been considered for anchoring DCE values onto the full health to dead scale. The anchoring approaches that need data from separate TTO interviews among participants requires greater time and resources. Dental caries is generally not a life-threatening condition or associated with serious disabilities. Therefore, pairwise comparisons with death would also not be appropriate since dental caries health state would be predominantly the dominant choice over death or living in full dental health for a considerably shorter duration. DCE_{TTO} method is able to convert the latent utility values obtained from DCE to QALY scale while minimising the drawbacks with conventional TTO. The DCE_{TTO} method has been used for valuation studies (68, 166) and the methodology has been tested widely (166, 167). Therefore, the DCE_{TTO} approach will be used as an anchoring approach for the valuation of DCUI. The VAS has been used in recent health state valuation studies to generate utility value sets (168). VAS includes a scale of a single line in which the top of the scale indicates the 'best imaginable health' and the bottom of the scale indicates 'the worst imaginable health'. Individuals are asked to place the health state of interest on this scale (110). VAS is simple and easy to understand compared to the SG and TTO methods (169) and does not attach any trade-off between life years. As mentioned earlier, dental caries is neither life-threatening nor associated with serious disabilities under normal circumstances. Therefore, anchoring with VAS would be an option for a disease condition in which participants will be reluctant to sacrifice life years for the quality of life.

Since there is no previous evidence to identify the most suitable anchoring methods for health state valuations in dental caries, it is worth exploring the two possible options; DCE_{TTO} and anchoring the worst health state with VAS. Therefore, we will first conduct two pilot studies with two DCE designs; DCE_{TTO} (*Valuation approach 1*) and DCE with VAS (*Valuation approach 2*) in order to identify the most suitable method among these two approaches, and this will then be used to generate the utility algorithm in the main valuation survey.

DCE experimental design and construction of choice sets

The DCUI instrument consisted of five items, each with four levels. Therefore, there are 1024 (4^5) possible health states for a full factorial design. As it will not be practical to value all possible combinations, a D-efficient design will be applied to select a subset of these health states while maximising the efficiency of the survey design (170). D-error is a term use to denote the statistical efficiency of a design; a low D-error indicates a more efficient design. Compared to orthogonal designs, D-efficient designs can produce better parameter estimates (171). Two separate D-efficient designs with the model specification as multinomial logit model (MNL) for valuation approach 1 and 2 (to be explained below) will be generated using Ngene software (172). As no previous studies valued dental caries health states derived from a classification system to determine the priors, zero priors will be assumed for all variables to generate the D-efficient designs in pilot surveys. For the main survey, the pilot data will be used to specify the priors for the D-efficient design. The number of choice tasks used in the previous health state valuations using DCE were ranged from 12–3160 and the majority of studies included more than 151 choice tasks (65). The number of choice tasks per respondent ranged from 2-108 (65) to value health states derived from a classification system. For this study, eight choice sets per respondent are chosen and block design will be used with eight choice sets in 25 blocks to value 200 pairwise choice tasks. The block design will ensure an equal number of respondents per block. In addition to these eight choice tasks per respondent, the DCE survey will be started, with a practice DCE choice task and a dominant choice question at the beginning of each block as a warm up task and to allow respondents to be familiar with the DCE tasks.

Pilot Study- DCE_{TTO} (Valuation approach 1)

For the valuation approach 1, DCE choice tasks will include duration as an additional attribute (DCE_{TTO} choice tasks). Previous DCE studies in dental caries confined to the evaluation of treatment preferences or health services (74, 75) rather than the valuation of dental caries health states defined by a classification system. Therefore, no previous literature is available to determine the best levels for the duration attribute in the DCE_{TTO} for dental caries. Dental caries is a chronic disease. Progression of dental caries depends on the balance between pathological factors, such as dietary sugars and bacterial count, and protective factors, such as fluoride and good oral hygiene (173). Thus, an assumption was made that the oral health status is constant over time and participants

will be in the same caries state in the given duration when including the duration attribute to the choice tasks. Survival duration in the DCE$_{TTO}$ could be interpreted as an equivalent to 'marginal willingness to trade life years for an improvement in health status' (64). Therefore, it was decided to include 6 months, 1 year, 4 years, 7 years, and 10 years as duration levels. Six months is included as it is recommended to visit the dentist every 6 months. 1 year, 4 years, 7 years, and 10 years were included as these are the commonly used duration levels in conventional TTO tasks to calculate QALY (174). An example DCE$_{TTO}$ choice task is given in **Table 4.2**.

Table 4.2: An example of a discrete choice experiment task including the duration attribute for *DCE$_{TTO}$*

Please consider that you are living a life with tooth decay in health state A or B for the period of time specified. During this time, your health state would not change and then you will die. Which health state do you think is better (health state A or health state B)?		
	Health State A	Health State B
Pain/Discomfort	I have no pain or discomfort	I have a little pain or discomfort
Difficulty eating foods/drinking	I have a little difficulty in eating food/drinking	I have a little difficulty in eating food/drinking
Worried *(e.g., about losing your tooth, etc.)*	I am a little bit worried	I am quite worried
Ability to participate in activities *(e.g. playing with your friends, sports, schoolwork, etc.)*	I have no difficulty participating in activities	I have a little difficulty participating in activities
Appearance	I am a little concerned about my appearance	I am not concerned about my appearance
Duration of life	Stay this health state for 1 year and then die	Stay this health state for 4 years and then die
Which health state do you think is better	○	○

Pilot Study- DCE_{VAS} (Valuation approach 2)

The valuation approach 2 will include DCE tasks without the duration attribute. An example of DCE choice task without the duration attribute is given in **Table 4.3**. In addition to this, VAS tasks will also be included for anchoring. After DCE paired tasks, a VAS task will be included to value the best health state (11111), the worst heath state of the DCUI (44444), a mild health state, a moderate health state, a severe health state and death in a single VAS scale. The two extreme endpoints of the VAS scale indicate 'best imaginable oral health' (Score 100) and 'worst imaginable oral health' (Score 0). An outline of this task is given in **Figure 4.1**.

Further, for both pilot studies, questions will be added at the end of the DCE choice tasks to assess the difficulty in understanding the questions and difficulty in completing the tasks on a response scale of 1-4 (Not difficult at all to extremely difficult). Time taken to complete the whole survey and each task based on the start time and end time will be assessed to check the feasibility and participant burden.

Table 4.3: An example of a discrete choice experiment task for *DCE_{VAS}*

Please consider that you are living a life with tooth decay in health state A or B for a same period of time. During the time, your health state would not change. Which health state do you think is better (health state A or health state B)?		
	Health State A	Health State B
Pain/Discomfort	I have no pain or discomfort	I have a little pain or discomfort
Difficulty eating foods /drinking	I have a little difficulty in eating food/ drinking	I have a little difficulty in eating food/ drinking
Worried ***(e.g. about losing your tooth, etc.)***	I am a little bit worried	I am quite worried
Ability to participate in activities ***(e.g. playing with your friends, sports, schoolwork, etc.)***	I have no difficulty participating in activities	I have a little difficulty participating in activities
Appearance	I am a little concerned about my appearance	I am not concerned about my appearance
Which health state do you think is better	○	○

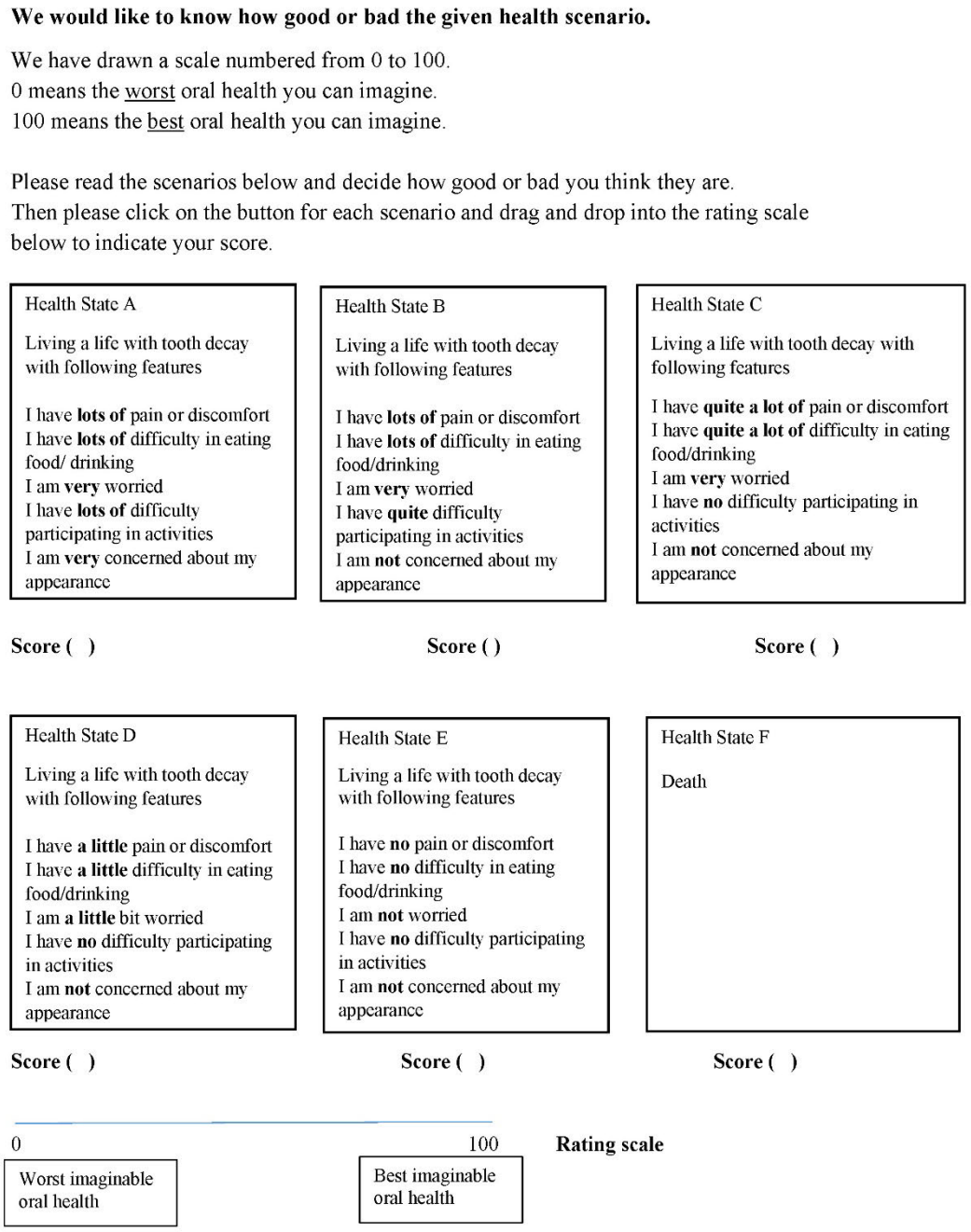


Figure 4.1: Visual Analogue Task (VAS) for DCE_{VAS}

Main valuation survey

Based on the pilot studies, the more suitable anchoring method among these two approaches will be chosen and it will be used to generate a utility algorithm in the main survey conducted in the next phase of the study. In addition, these pilot studies will be

used to assess whether the survey is launching in the way it is designed, to identify any practical issues or to identify changes required for the main valuation survey.

Study sample

There are arguments that continue to be held among researchers regarding 'whose preference should be valued' for health state valuations of paediatric PBMs (175). Researchers have argued that the preferences of children and adolescents should be sought, since they have different preferences than adults, and it is them who are receiving the care (70, 176). However, health state valuation among children is associated with methodological constraints, especially with anchoring tasks (175). These tasks would not only be a cognitive burden for children but there are also ethical issues associated with presenting 'death' to children. Thus, previous studies conducted to develop adolescent-specific algorithm followed different approaches. Child Health Utility-9D adolescent-specific algorithms (67) were developed using two-steps; valuation tasks among adolescents and a separate TTO study with young adults group for anchoring. TTO tasks with adolescent-friendly wordings in face-to-face interviews were used to develop adolescent-specific algorithm for Assessment of Quality of Life (AQoL)-6D (177). However, it is common for child and adolescent health state values to be based on an adult general population sample or use proxy responses from parents/guardian (161). Most of the existing generic paediatric PBM valuation studies were conducted with an adult general population (53, 68, 175) as it is often public money that will be allocated to fund treatment and therefore it is the preferences of the public that matters. The present study is also planned to elicit preferences for health states from an adult general population sample; this is a highly feasible approach for generating health state values using an online sample (175).

Sample size and recruitment

The calculation of sample size for DCE studies is often complex (178) and researchers suggest different formulae based on several factors (179). Johnson and Orme (180) suggest that the sample size (n) required for the main effects model is based on the number of choice tasks (t), the number of alternatives (a), and the number of analysis cells (c), which is equal to the largest number of levels for any of the attributes. Further, Johnson and Orme (180) recommended a sample size of 300 as a rule of thumb for a quantitative study where there is no intention to compare subgroups and a minimum of

about 200 per group for studies that plan to compare groups of respondents to be able to detect significant differences. Further, Lancsar and Louviere (181) stated that 20 respondents per choice set would be sufficient to estimate a reliable model and a sample size of 1000-2000 will be able to produce small confidence intervals.

Pilot Study

Two pilot designs will be tested with an online sample of 400 (200 for each design) participants that is age and sex representative of the adult general population in Australia.

Main valuation survey

Based on the literature reviews and similar Australian online DCE surveys (178), our target is to include a sample size of 2,000 representative respondents, which will be compatible with the above requirements for the main survey.

Administration of the survey

A representative sample of the adult Australian general population in relation to age and gender will be recruited from March 2020. Study participants will be recruited from an existing Australian online panel with the help of online research company SurveyEngine (http://www.surveyengine.com). Around 86% of all households in Australia had access to the internet at home in 2014–15 (182) indicating that online study has a good chance of reaching the target population. Potential participants (adults over 18 years old) who have registered in the online survey panel will receive the invitation for the study and interested panel members will have access to the survey through the provided link. They will be guided through the online survey by screen prompts. The first section of the online survey will be the introduction page where participants will be given all the necessary details about the research project and contact details of the investigator if they need further clarification. Participants will be informed that the participation in this survey will be voluntary and the survey data will be anonymous and confidential. At the end of the introduction page, participants will be requested to provide their consent. Once the participant gives their consent, they will move to the next section and continue the rest of the survey. Consenting participants will be guided through the online survey by screen prompts and this will enable them to complete the tasks at their convenience. The next section of the survey will include screening questions (age and sex) followed by

DCUI and a set of DCE choice tasks and anchoring tasks. Then the participants will be asked to complete a generic preference-based QoL instrument (the EuroQoL5D 5L questionnaire- EQ-5D-5L) (183), an OHRQoL instrument (Oral health impact profile-14-OHIP-14) (184). EQ-5D-5L and OHIP-14 are commonly used among the Australian general population (185) to assess generic and oral health-related QoL, respectively. The OHIP-14 has shown adequate psychometric properties and has been validated extensively among both adults (186) and adolescent (187) populations. Therefore, EQ-5D-5L and OHIP-14 data will be used to assess the general QoL and OHRQoL of the study participants and to evaluate the performance of the DCUI. The final section of the survey will include questions regarding basic social-demographic characteristics, oral health status and frequency of dental visits.

Patient and public involvement

No patient involved.

Analytical plan

All data will be cleaned prior to the analysis and data will be analysed using STATA version 15.1 (188). DCE data will be analysed using a conditional logit model under a random utility framework, which assumes that respondents choose the alternative that maximises their utility (189). The utility function consists of a vector of observable attributes as well as a random error term (55). Both the main effects and the interactions among attributes will be considered.

Pilot Study- DCE_{TTO} (Valuation approach 1)

The observable component (μ_{ij}) of the utility function will be estimated using a conditional logit model as in Equation 1. The life years t will be included as a continuous variable.

$$\mu_{isj} = \alpha t_{isj} + \beta x_{isj}.\, t_{isj} + \epsilon_{isj} \rightarrow \; Equation\, 1$$

μ_{isj} = utility of the option *j* in choice set *s* for survey respondent *i*

α = utility associated with a life year

t = life years

β= corresponding vector of utility weights associated with each level in each dimension, for each life year

x_{isj} = a vector containing five DCUI dimensions; each dimension was estimated using three dummy variables (with 'no problems' serving as the reference level within each attribute) presented in option *j*

ϵ_{isj} = error term

Anchoring results to a health utility scale will be performed based on the methodology described by Bansback et al. (64). The objective is to derive the mean utility value of the state x_{ij} in DCE that correspond to a 10 year TTO value.

$$\hat{V}_j^{DCE} = 1 + \frac{\hat{\beta}}{\hat{\alpha}} x_j \quad \rightarrow Equation\ 2$$

V_j = value of the health state h_j anchored on the health utility scale

$\hat{\alpha}$ = disutility of living with the health state h_j for one year

$\hat{\beta}$ = coefficient representing the value of living in full health for one year

x_j = value for each health state

Based on this formula, the sample mean DCE_{TTO} for the health state h_j can be calculated from the coefficients of the conditional logit model.

Pilot Study- DCE with VAS (Valuation approach 2)

To obtain the coefficients in DCE latent scale, data from the DCE choice tasks in valuation approach 2 will be modelled with an appropriate regression model with the following specification. The best-fitted model will be selected based on the statistical significance of coefficients, the amount of explained variance, and mean absolute error (MAE- absolute difference between the observed value and estimated value in each health state).

$$\mu_{ij} = \beta_1 x_1 + \beta_2 x_2 + \beta_3 x_3 + \beta_4 x_4 + \beta_5 x_5 \quad \rightarrow Equation\ 3$$

μ_{ij} = observable component of the utility function

β = coefficient for each dimension

x = vector contains five DCUI dimensions whilst each dimension is estimated using three dummy variables (with 'no problems' served as the reference level within each attribute).

Anchoring to the full health-dead scale of the VAS anchoring task will be performed based on the methodology proposed by Brazier et al (169) and Rowen et al (66). Brazier et al (169) proposed a formula (Equation 4) to convert the health state values into full health-dead scale based on the value obtained for best health state and death from

VAS health state valuation tasks and this has been used in previous studies (168). Raw VAS scores for worst heath state of the DCUI (44444), a mild health state, a moderate health state, and a severe health state will be converted to full health-dead scale using Equation 4.

$$V_h = \frac{S_h - S_{dead}}{S_{11111} - S_{dead}} \quad \rightarrow \quad Equation\ 4$$

V_h = adjusted VAS rating for health state *h*

S_h = respondent's unadjusted VAS score for state *h*

S_{dead} = respondent's assigned VAS score for the health state 'death'

S_{11111} = respondent's assigned VAS score for a state 11111 (best state)

Then the value of the worst state in the DCE model will be anchored based on anchoring with worst health states and with mapping DCE onto VAS. Rowen et al (66) converted the coefficients on a latent utility scale estimated in DCE data onto the full health-dead scale using the estimated TTO value of the worst state. The same methodology would be followed here and based on the adopted formula (Equation 5) from Rowen et al (66), DCE data will be anchored to the worst state value obtained in VAS.

$$\beta_{r\lambda\Theta} = \beta_{\lambda\Theta} * \frac{wVAS}{wDCE} \quad \rightarrow \quad Equation\ 5$$

$\beta_{r\lambda\Theta}$ = rescaled coefficient for level λ of dimension Θ

$\beta_{r\lambda\Theta}$ = coefficient for level λ of dimension Θ

wVAS= estimated VAS value for the worst state generated using Equations 4

wDCE= DCE value for the worst state estimated using the DCE model

For the anchoring with mapping DCE onto VAS using a linear regression**,** mean VAS values obtained for the worst health state of the DCUI (44444), a mild health state, a moderate health state, and a severe health state will be used as in Equation 6.

$$dVAS_j = 1 - VAS_j$$

$$dDCE_j = 1 - DCE_j$$

$$dVAS_j = f(dDCE_j) + \varepsilon_j \quad \rightarrow \quad Equation\ 6$$

VASj = mean VAS value of health state j

DCEj = modelled latent utility value of health state j

dVASj = disutility of the mean VAS value of health state j

dDCEj = disutility of the modelled latent utility value of health state j

ε= error term

Comparison of valuation approach 1 and 2 to assess the most suitable anchoring method for dental caries health state valuation

As these two DCE designs are different, they are not directly comparable. However, the pilot studies aim to identify the most suitable method for anchoring among these two approaches. Therefore, data from the two valuation approaches will be compared concerning the respondents' self-reported difficulty of the tasks (based on the answers provided for the two questions- how difficult to understand the questions and how difficult to complete the tasks), dropout rate, time taken by each individual on the whole survey and for each DCE task. Further anchored coefficients obtained from the valuation approach 1 and 2, as outlined in the analytical plan, will be examined for the sign and order of the coefficients; i.e. the sign of the duration coefficient should be positive (since utility increase with the time living in full health) and levels in each domain should follow a logical order in which more severe should have larger utility decrement.

Main valuation survey

Once the most suitable valuation approach is decided based on the pilot surveys, that approach will continue as the main valuation survey to generate the utility algorithm. For the main survey, participants' characteristics will be assessed and a chi-square test will be used to assess sample representativeness of the Australian general population. The EQ-5D-5L, OHIP-14, DCUI, and self-reported oral health variables data will be analysed to assess the respondents' QoL and oral health status. DCE tasks data of the main survey will be modelled based on the selected approach from the pilot study, as outlined in the analytical plan. The utility values generated from the main survey can then be used to calculate QALYs in economic evaluation in dental caries interventions.

Ethics and dissemination

Ethical approval for this study was obtained from the Human Research Ethics Committee, Griffith University (HREC/2019/550) **(Appendix D)**. Informed consent will be obtained from the participants prior to starting the survey. Participants are only required to complete the survey; therefore, there is negligible or low risk for the participants. There are no potential adverse events/side effects. The research team will receive only de-identified anonymous data. The data obtained by this research project will be stored securely with a password-protected computer, and secure server in Griffith University until data will be destroyed after the five years minimum period of retention in accordance with University policy. Any personal details that will lead to the identification of individual participants will not be included in any report or publication arising from this research project. Dissemination of the study results will be through the publication of manuscripts in academic journals and conference presentations.

Chapter 5. Comparison of two approaches to value health states derived by preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a co-authored paper prepared for submission. The bibliographic details of the co-authored paper, including all authors, are:

Hettiarachchi RM, Byrnes J, Kularatna S, Chen G, Mulhern B, Scuffham PA. Comparison of two approaches to value health states derived by preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents

My contribution to the paper involved designing the study and the survey, planning and executing the data collection, analysing the data and drafting the article.

(Signed) _____ (Date)_____

Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Supervisor: Prof. Paul Scuffham

5. Comparison of two approaches to value health states derived by preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI) among Australian adolescents

5.1 Abstract

Introduction

The Dental Caries Utility Index (DCUI), a new health state classification system for dental caries has been developed to facilitate economic evaluations of dental caries interventions. The objective of this study was to identify the most suitable method of anchoring among two possible approaches—discrete choice experiment with duration (DCE_{TTO}) and discrete choice experiment with visual analogue scale (DCE_{VAS})—to generate a preference-based scoring algorithm for the DCUI classification system.

Methods

An online survey was conducted with a sample from the Australian general population. Two separate discrete choice experiment (DCE) designs were created for each valuation approach. DCE data were modelled using conditional logit and the modelled coefficients from DCE_{TTO} were anchored using a coefficient for the duration. DCE_{VAS} data were anchored based on two methods: anchoring using the worst health state from VAS and mapping DCE onto VAS. The rescaled coefficients and utility values from both approaches were compared. Further, the two methods were compared in terms the respondents' perceptions of task difficulty and selected characteristics.

Results

Totals of 200 and 191 participants completed the DCE_{TTO} and DCE_{VAS} surveys, respectively. There were no significant difference between the socio-demographic characteristics of the two samples. DCE_{VAS} produced more ordered and significant coefficients than the DCE_{TTO} models. Further, DCE_{TTO} resulted in greater dispersion among utility decrements and the severe health states were valued worse than death, which is unreliable for a condition like dental caries. However, there was no statistically significant difference between the participants' self-reported difficulty in understanding and completing the valuation tasks.

Conclusion

Compared to the DCE_{TTO} approach, DCE_{VAS} is a feasible and reliable approach to anchor the utility values obtained from DCE on a latent scale onto a full health-dead scale in dental caries health state valuations.

Key words: Dental caries, Valuation, Discrete choice experiments, Preference-based, Visual analogue scale

5.2 Introduction

Preference-based quality of life measures (PBMs) are a type of patient-reported outcome measure. They are often used to calculate quality-adjusted life years (QALYs) to assess the cost-effectiveness of health care interventions using a cost-utility analysis framework (53). PBMs are pre-scored instruments and thus, are easy to use and less time consuming than directly eliciting preferences from patients (52). PBMs consist of two main components: a health state classification system and a scoring algorithm or a set of health state utility values corresponding to each of the health states defined by the classification system (61). A set of utility weights or scoring algorithm is generated by estimating utility values for selected health states, often from a sample of the general population using a preference-elicitation method (65). Then, statistical modelling techniques are used to generate a scoring algorithm and to derive the utility values for all possible health states (65).

Time trade-off (TTO), standard gamble (SG), visual analogue scale (VAS), discrete choice experiment (DCEs) and best-worst scaling (BWS) are the preference-elicitation techniques most widely used in health state valuation studies (52). Among these, DCE methods have recently become prevalent (65). In a DCE study, participants are requested to state their preference for a series of choices between two or more alternative scenarios involving health (75). The TTO and SG are cardinal preference techniques that produce utility values anchored by full health and death (64). Conversely, DCE and BWS are ordinal preference elicitation methods that produce utility values on a latent scale. Therefore, the utility values obtained from these methods must be anchored on to a full health-dead scale prior to their use in economic evaluations to calculate QALYs (66). Different methods have been used to anchor DCE latent utility values to a

full health-dead scale. Common methods include the use of data from a separate study (usually, researchers conduct a separate TTO study and DCE decrements in the latent scale are anchored based on mapping the DCE into the TTO or using hybrid models for DCE and TTO or anchoring the worst state using TTO (66)), the use of duration as an attribute within the choice task (DCE_{TTO}) (64) and the inclusion of a third choice alternative (either full health or a dead state) (66).

The Dental Caries Utility Index (DCUI), a new health state classification system for dental caries, has been developed to facilitate economic evaluations of dental caries interventions. The DCUI consists of five domains: pain/discomfort, difficulty eating food/drinking, worried, ability to participate in activities, and appearance. Each domain has four response levels. As in other areas of health, DCEs are becoming popular in oral health research, although most of these studies are related to treatment preferences and do not elicit utility values for a health state classification system. Therefore, the DCE method was selected to generate a utility algorithm for the health states derived from the DCUI classification system. Compared to other health state valuation methods, DCEs are compatible with online surveys, easy to administer and a lower cognitive burden on participants. However, there is no evidence for the most suitable approach to anchor DCE latent scale utility values onto a full health-dead scale in dental caries health state valuations. DCE_{TTO} is the most common anchoring method (65) and has been widely tested (166, 167, 190). Moreover, in DCE_{TTO}, the anchoring task is integrated within the DCE choice tasks (64) and hence, this method does not require a separate study for the anchoring tasks. Although the VAS has been used to generate utility value sets in health state valuation studies (168), it has not been applied to anchor latent utility values derived from a DCE study onto a full health-dead scale. VAS is simple, easy to understand and does not require a trade-off between life years and risk (169). This is important because dental caries is not associated with high mortality or serious disabilities in which individuals require external support to perform their day-to-day activities. Therefore, anchoring with VAS would be a suitable alternative for conditions like dental caries. As no previous studies identified the most suitable anchoring approach for DCE health state valuation in dental caries, it is necessary to explore these two possible options: discrete choice experiment with duration ($DCE_{TTO)}$ and discrete choice experiment with visual analogue scale (DCE_{VAS}). Therefore, the objective of this study was to evaluate the DCE_{TTO} and DCE_{VAS} to identify the most suitable anchoring method to generate a preference-based scoring algorithm for the DCUI classification system.

5.3 Methods

The detailed description of methods and plan of analysis has been submitted as a protocol paper elsewhere (191). A summary of methods is presented here.

DCE experimental design and construction of choice sets

The DCUI instrument consisted of 1024 (4^5) possible health states. Therefore, a D-efficient design (170) was applied to select a subset of 200 pairwise choice tasks while maximising the efficiency of the survey design. Two separate D-efficient designs were generated using Ngene software (172) with the model specified as a Multinomial Model (MNL). As DCUI is a new health state classification system, no prior evidence for preferences was available. Hence, zero priors were assumed for all variables to generate the D-efficient designs. A block design was applied to ensure an equal number of respondents per block. Eight choice sets per respondent in 25 blocks were included to value 200 pairwise health states. The Ngene design codes and DCE design matrix for the DCE_{TTO} approach are provided in Appendices E and F, respectively; while those for the DCE_{VAS} method are provided in Appendices G and H, respectively.

Pilot study: DCE_{TTO} (Valuation approach 1)

The DCE choice tasks for the DCE_{TTO} approach included five attributes from the DCUI and an additional attribute 'duration' for the purpose of anchoring. As there are no previous studies in dental caries to determine the best levels for the duration attribute in the DCE_{TTO} for dental caries, periods of 6 months, 1 year, 4 years, 7 years and 10 years were selected as duration levels. A period of 6 months was considered as the current recommendation is that patients visit the dentist every 6 months, while 1 year, 4 years, 7 years and 10 years are the commonly used duration levels in conventional TTO tasks to calculate QALYs (174). An example DCE_{TTO} choice task is given in **Figure 5.1**.

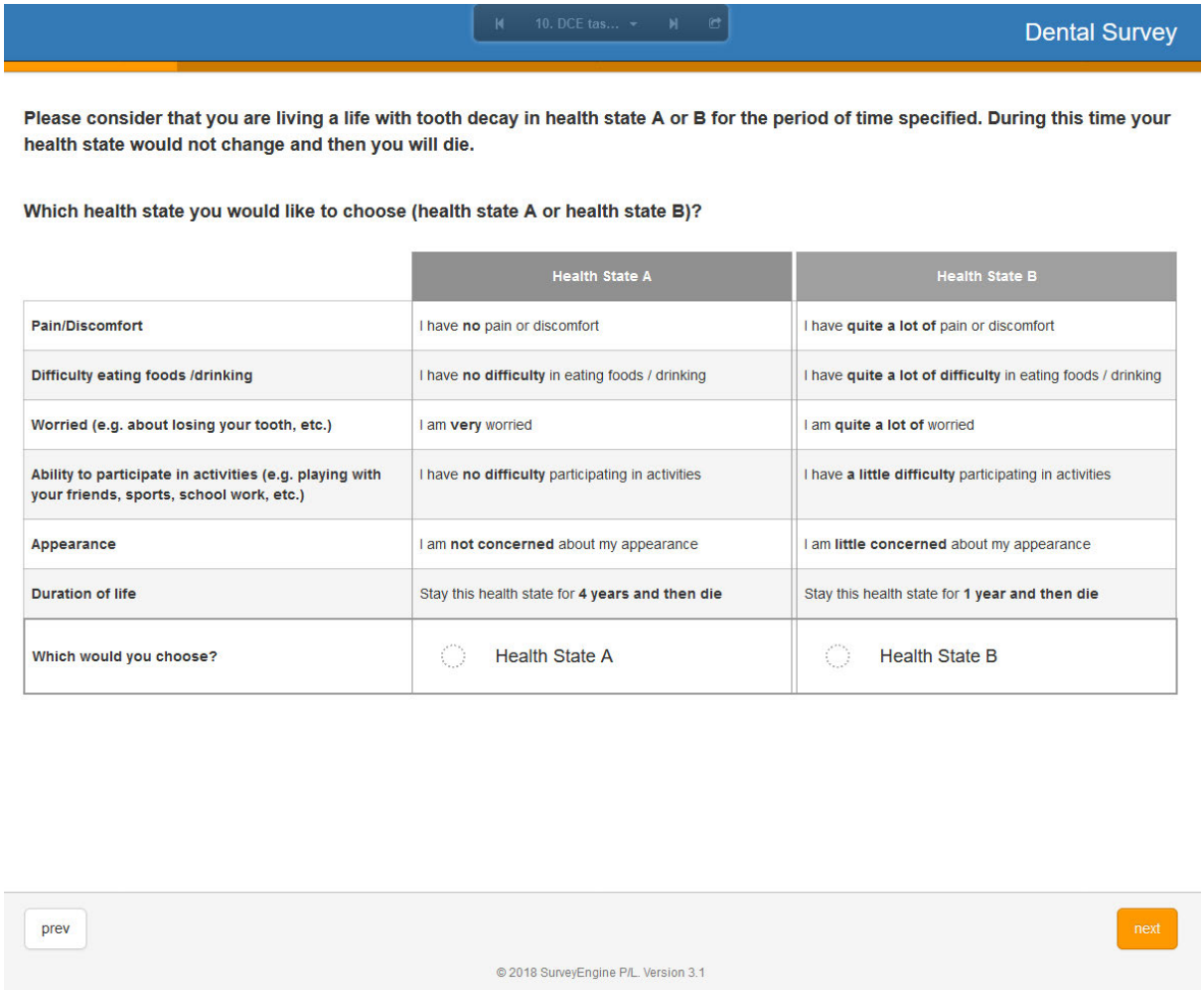


Figure 5.1: An example of a discrete choice experiment task including the duration attribute for the valuation approach 1

Pilot study: DCE$_{VAS}$ (Valuation approach 2)

Valuation approach 2 included DCE tasks without the duration attribute. An example is given in **Figure 5.2**. After the DCE paired tasks, a VAS task was included to value six health states, including the best (11111), the worst (44444), a mild (22211), a moderate (33341) and a severe (44431) state along with death on a single VAS scale. The mild, moderate and severe health states were selected to represent combinations of attribute levels with differing severity. The two extreme endpoints of the VAS scale were marked as 'best imaginable oral health' (Score 100) and 'worst imaginable oral health' (Score 0). An outline of this task is presented in **Figure 5.3**.

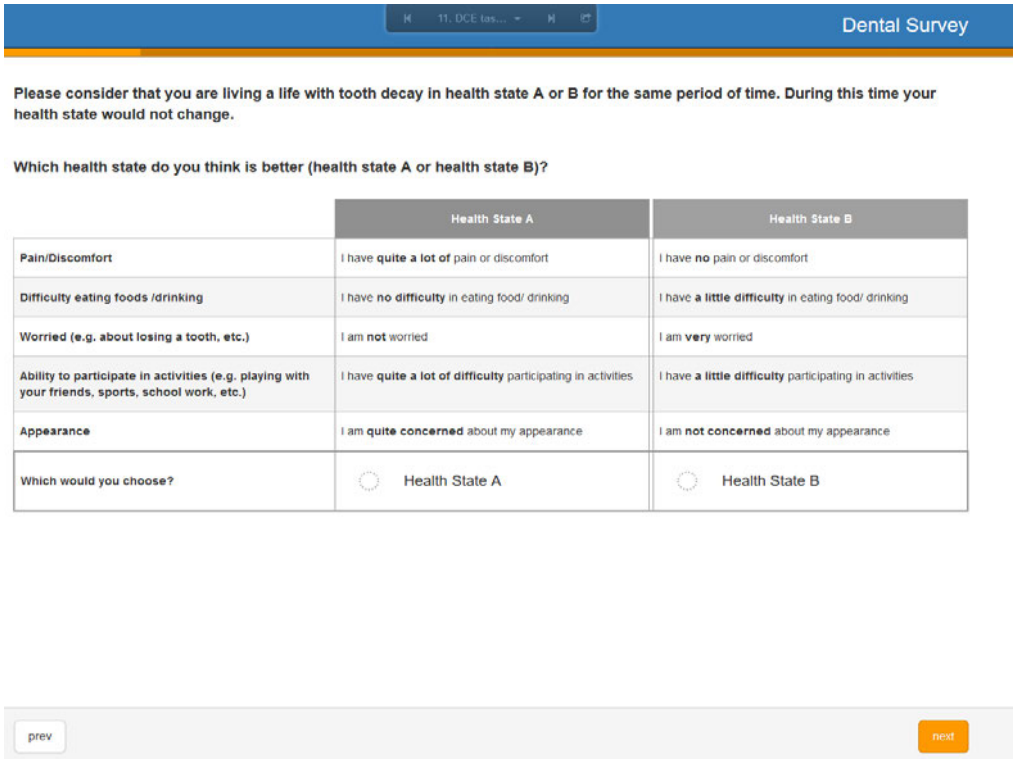


Figure 5.2: An example of a discrete choice experiment task in the valuation approach 1

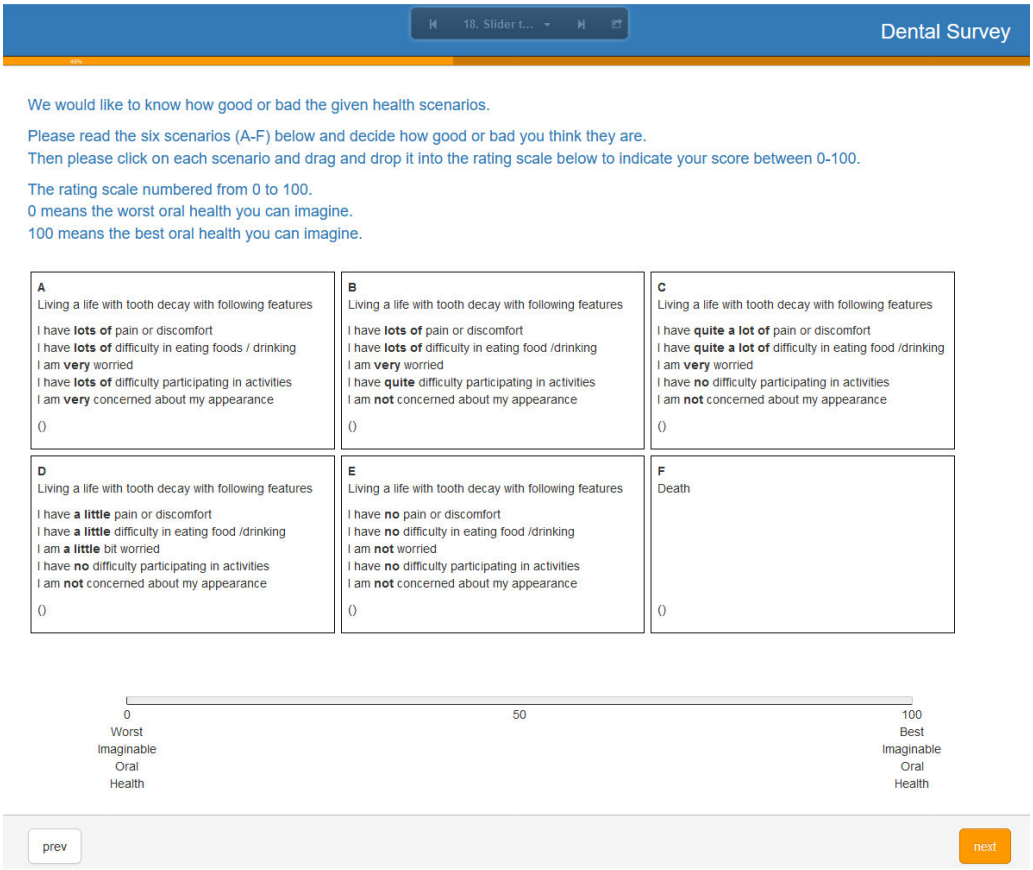


Figure 5.3: VAS valuation task

Study sample and recruitment

The target group of the DCUI instrument is children above 12 years of age. However, designing and conducting a health state valuation study among children is challenging because the valuation tasks could imposed a high cognitive burden on the children (175). Moreover, ethical issues may arise, as some tasks require the sample to consider 'immediate death' as an option. Therefore, most of the existing generic paediatric PBM valuation studies were conducted with an adult general population sample (53, 68, 175), as it is often public money that will be allocated to fund treatment and therefore, the preferences of the public matter. The present valuation study was also conducted among an adult general population sample (175). Johnson and Orme (180) recommended a sample size of 300 as a rule of thumb for a quantitative study where there is no intention to compare subgroups and a minimum of 200 per group for studies that plan to compare groups of respondents to be able to detect significant differences. Therefore, we aimed to include an online sample of 400 representative adults (200 for each design).

An age and sex representative sample from the Australian adult general population was recruited with the support of online research company SurveyEngine (http://www.surveyengine.com). Potential participants (adults over 18 years old) were invited through the online research company and interested participants accessed the survey using the provided link. The survey **(Appendix I)** contents were similar for both valuation approaches except for the DCE choice tasks and the anchoring tasks. The online survey started with an introduction page with a concise description of the research project and the participants were requested to provide their consent. Consenting participants were guided through the online survey by screen prompts and they were able to complete the tasks at their convenience. The subsequent section of the survey included screening questions (age and sex) and the participants were then asked to complete DCUI while thinking about their own oral health status as a warm-up task. Next, the survey included a set of DCE choice tasks. In addition to the eight choice tasks, another two tasks (a practice DCE choice task and a dominant choice question) were included as warm-up tasks at the beginning of each block. In the DCE_{VAS} approach, a VAS anchoring task was included after the DCE choice tasks. Further, the survey included questions at the end of the valuation tasks to assess participants' self-reported difficulty in understanding and completing the tasks on a response scale of 1–4 (Not difficult at all to extremely difficult).

After the valuation tasks, the participants were asked to complete a generic preference-based QoL instrument (the EuroQoL5D 5L questionnaire, EQ-5D-5L) **(Appendix J)** (183), an OHRQoL instrument (Oral health impact profile-14, OHIP-14) (184) and questions regarding basic socio-demographic characteristics, oral health status and frequency of dental visits. OHIP-14 and EQ-5D-5L are the most commonly used instruments to assess oral health and generic QoL among the Australian general population (185). Ethical approval for this study was obtained from the Human Research Ethics Committee, Griffith University (HREC/2019/550) **(Appendix D)**.

Data analysis

Data analysis was preformed using STATA version 15.1 (188) and all data were cleaned prior to the analysis. DCE data were analysed using a conditional logit model under a random utility framework, which assumes that respondents choose the alternative that maximises their utility (189).

Pilot study: DCE_{TTO} (Valuation 1)

The observable component (μ_{ij}) of the utility function was estimated using a conditional logit model (Equation 1) and life years t was included as a continuous variable:

$$\mu_{isj} = \alpha t_{isj} + \beta x_{isj}.\ t_{isj} + \epsilon_{isj} \rightarrow \ Equation\ 1$$

μ_{isj} = utility of the option j in choice set s for survey respondent i

α = utility associated with a life year

t = life years

β= corresponding vector of utility weights associated with each level in each dimension, for each life year

x_{isj} = a vector containing five DCUI dimensions; each dimension was estimated using three dummy variables (with 'no problems' serving as the reference level within each attribute) presented in option j

ϵ_{isj} = error term

The DCE modelled data on a latent scale were anchored onto a full health-dead scale based on the methodology described by Bansback et al (64). The objective was to derive the mean utility value of the state x_{ij} in DCE that corresponds to a 10-year TTO

value. Based on this formula, the sample mean DCE_{TTO} for the health state h_j was calculated from the coefficients of the conditional logit model:

$$\hat{V}_j^{DCE} = 1 + \frac{\hat{\beta}'_2}{\hat{\beta}_2} x_j \quad \rightarrow Equation\ 2$$

V_j = value of the health state h_j anchored on the health utility scale

$\hat{\beta}'_2$= disutility of living with the health state h_j for one year

$\hat{\beta}_2$= coefficient representing the value of living in full health for one year

x_j = value for each health state

Pilot study: DCE_{VAS} (Valuation approach 2)

Data from the DCE choice tasks in valuation approach 2 were modelled with conditional logit model with the following specifications:

$$\mu_{ij} = \beta_1 x_1 + \beta_2 x_2 + \beta_3 x_3 + \beta_4 x_4 + \beta_5 x_5 \quad \rightarrow Equation\ 3$$

μ_{ij} = observable component of the utility function

x = vector containing five DCUI dimensions; each dimension is estimated using three dummy variables (with 'no problems' serving as the reference level within each attribute).

β = coefficient for each dimension

The DCE modelled data on a latent scale were anchored onto a full health-dead scale with VAS data based on two methods: the worst health state using VAS and mapping DCE onto VAS. The raw VAS data were cleaned prior to the data analysis. Participants expected to give logical answers to the death, best or worst health states to support the meaningful results in anchoring procedure. Strict exclusion criteria were applied to remove all relevant logical inconsistencies and obtain reliable adjusted VAS scores. Therefore, participants were excluded from the VAS data analysis if they met at least one of the following exclusion criteria:

- Participants valued best ≤ death or worst or mild or moderate or severe.
- Participants valued death ≥ mild or moderate.
- Participants valued worst ≥ mild or moderate or severe.
- Participants were missing either death, best or worst VAS data (due to a technical issue).

Prior to anchoring, VAS scores for the worst health state of the DCUI (44444), a mild health state, a moderate health state and a severe health state were converted to a full health-dead scale at the individual level using Equation 4 proposed by Brazier et al (169):

$$V_h = \frac{S_h - S_{dead}}{S_{11111} - S_{dead}} \quad \rightarrow \quad Equation\ 4$$

V_h = adjusted VAS rating for health state *h*

S_h = respondent's unadjusted VAS score for state *h*

S_{dead} = respondent's assigned VAS score for the health state 'death'

S_{11111} = respondent's assigned VAS score for a state 11111 (best state)

Anchoring with worst health state using VAS

Rowen et al (66) proposed a methodology to rescale the coefficients on a latent utility scale onto the full health-dead scale using the estimated TTO value of the worst health state. The same formula was adopted (Equation 5) here to anchor DCE data onto the full health-dead scale.

$$\beta_{r\lambda\Theta} = \beta_{\lambda\Theta} * \frac{wVAS}{wDCE} \quad \rightarrow \quad Equation\ 5$$

$\beta_{r\lambda\Theta}$ = rescaled coefficient for level λ of dimension Θ

$\beta_{r\lambda\Theta}$ = coefficient for level λ of dimension Θ

wVAS= estimated VAS value for the worst state generated using Equations 1

wDCE= DCE value for the worst state estimated using the DCE model

Mapping DCE onto VAS

For anchoring by mapping the DCE onto the VAS using a linear regression, utility values for the worst health state of the DCUI (44444), a mild health state, a moderate health state and a severe health state were calculated based on the coefficients obtained in the DCE latent scale and regressed on to the adjusted VAS scores for these four health states using Equation 6:

$$dVAS_j = 1 - VAS_j$$

$$dDCE_j = 1 - DCE_j$$

$$dVAS_j = f(dDCE_j) + \varepsilon_j \rightarrow \ Equation\ 6$$

VASj = mean VAS value of health state *j*

DCEj = modelled latent utility value of health state *j*

dVASj = disutility of the mean VAS value of health state *j*

dDCEj = disutility of the modelled latent utility value of health state *j*

ε= error term

Comparison of valuation approaches 1 and 2 to assess the most suitable anchoring method for dental caries health state valuation

As these two DCE designs are different, they are not directly comparable. However, the pilot studies aim to identify the most suitable anchoring method of these two approaches. Therefore, data from both methods were compared concerning the respondents' self-reported perceptions of the task difficulty (based on their answers to two questions: How difficult was it to understand the questions? and How difficult was it to complete the tasks?) and selected characteristics. Further, anchored coefficients obtained from valuation approaches 1 and 2 were examined for their sign and the order of the coefficients; that is, the sign of the duration coefficient should be positive (since utility increases with the time living in full health) and the levels in each domain should follow a logical order in which more severe states have larger utility decrement.

5.4 Results

A total of 200 participants completed the survey for valuation approach 1 (DCE_{TTO}) and 191 participants completed the survey for valuation approach 2 (DCE_{VAS}). **Table 5.1** shows the comparison of two samples indicates that there was no statistically significant difference between them in relation to age, sex, education level, weekly income and marital status.

Table 5.1: Characteristics of the samples of valuation approaches

Characteristic	DCE_{TTO} n = 200	DCE_{VAS} n = 191	P-value*
Age, mean (SE)	47.12 (1.26)	46.85 (1.32)	0.87
Sex, n (%)			
Male	104 (52.0)	90 (49.6)	0.363
Female	96 (48.0)	101 (50.4)	
Highest level of Education*, n (%)			
Year 11 or below	21 (10.5)	25 (13.1)	0.247
Completed high school	42 (21.0)	31 (16.2)	
Trade certificate	23 (11.5)	35 (18.3)	
Diploma/advanced diploma	33 (16.5)	32 (16.8)	
Bachelor's degree and above	81 (40.5)	68 (35.6)	
Weekly Income* AUD, n (%)			
Less than $1,000	66 (33.0)	65 (33.5)	0.632
$1,000–$2,999	81 (40.5)	74 (39.6)	
$3,000–$4,499	19 (9.5)	20 (10.0)	
$4,500–$5,999	17 (8.5)	22 (10.0)	
$6,000 or more	17 (8.5)	10 (6.9)	
Marital Status*, n (%)			
Never married	57 (28.5)	56 (29.3)	0.948
Married/de facto	110 (55)	107 (56.0)	
Divorced/separated	26 (13.0)	21 (11.0)	
Widowed	6 (3.0)	5 (2.6)	
Prefer not to say	1 (0.5)	2 (1.1)	

** t-test for continuous data and chi-square with continuity correction for categorical data*

Table 5.2 shows the comparison of two valuation approaches based on selected criteria. Among the participants who completed DCE_{TTO} tasks, 21.5% reported that the tasks were difficult to understand and 29% reported having difficulty in completing the tasks. Of the participants completed DCE_{VAS} tasks, 26.7% reported that the tasks were difficult to understand and around 32% reported that it was difficult to complete the tasks. However, there was no significant difference between the participants' self-reported difficulties in understanding and completing the valuation tasks. The time taken to complete all the valuation tasks was significantly higher for the DCE_{VAS} method **(Table 5.2)**. However, there was no significant difference between the time taken to complete the DCE tasks between two valuation approaches **(Figure 5.4)**. The number of participants with data problems in two approaches were not significant.

Table 5.2: Comparison of responses between two valuation approaches

	DCE_{TTO}	DCE_{VAS}		
		DCE tasks	**VAS task**	**All valuation tasks**
Difficulty in understanding, n (%)				
Not Difficult	157 (78.5)	142 (74.4)	138 (72.2)	140 (73.3)
Somewhat difficult	36 (18.0)	24 (12.5)	35 (18.3)	36 (18.9)
Difficult	1 (0.5)	12 (6.3)	11 (5.8)	6 (3.1)
Extremely difficult	6 (3.0)	13 (6.8)	7 (3.7)	9 (4.7)
P value*	0.178			
Difficulty in completing the tasks, n (%)				
Not Difficult	142 (71.0)	126 (66.0)	133 (69.6)	122 (63.9)
Somewhat difficult	46 (23.0)	42 (22.0)	41 (21.5)	51 (26.7)
Difficult	6 (3.0)	13 (6.8)	7 (3.7)	9 (4.7)
Extremely Difficult	6 (3.0)	10 (5.2)	10 (5.2)	9 (4.7)
P value*	0.437			
Length of interview (LOI)- Median	9.8 min	14.3 min		
Time taken for valuation tasks (min)				
Mean (SD)	3.823 (3.508)	4.684 (5.402)	1.981 (1.461)	6.665 (6.022)
Min–Max	0.422–25.663	0.402–46.514	0.157–7.531	0.559–49.494
P-value**	0.061			
P-value*				0.000
Participants with data problems	48 (24%)[@]	40 (20.94%)^		
P value[#]	0.236			

** P-value between DCE_{TTO} and DCE_{VAS}, all valuation tasks*

***P-value between DCE_{TTO} and DCE_{VAS}, DCE tasks only*

[@]For the DCE_{TTO} approach, the number of participants with data problems was calculated as the number of participants who submitted more than two tasks with different duration and never selected the health state with shorter duration (48/200=24%)

^For the DCE_{VAS} approach, the number of participants with data problems was calculated as the number of participants with best, worst and/or death VAS data missing and/or those who valued death ≥ best or worst ≥ best (40/191=20.94%)

[#] Z test for proportions; Z=0.7237 not significant at p<0.05

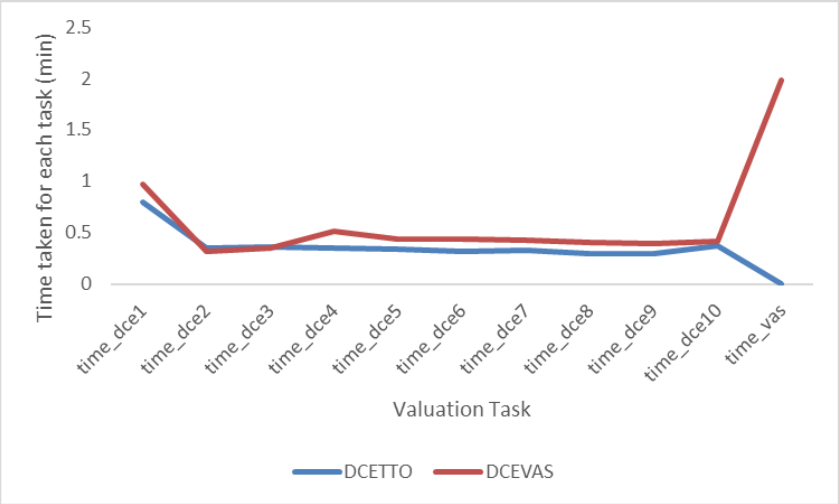


Figure 5.4: Mean time taken to complete each valuation task in the two valuation approaches

Valuation Approach 1: DCE$_{TTO}$

Parameters from the DCE$_{TTO}$ models (on the latent utility scale) were estimated using conditional logit. Table 5.3 reports parameter estimates from the unadjusted model (DCE$_{TTO}$ Model 1) and shows that the duration coefficient was both significant and in the expected direction. Of the 15 coefficients estimated for each level of the five dimensions, five were non-significant and coefficient was not in the expected direction (Level 2 of 'difficulty eat/drink'). The coefficients of all dimensions except 'difficulty eat/drink' and 'ability to participate activities' were ordered as expected (Table 5.3). Adjusted DCE$_{TTO}$ Model 2 was built by combining levels 1 and 2 of 'difficulty eat/drink' and DCE$_{TTO}$ Model 3 was built by combining levels 3 and 4 of 'ability to participate activities' (Table 5.3). However, these two adjusted models improved only the combined dimension. Therefore, another adjusted model was estimated by combining both levels 1 and 2 of 'difficulty eat/drink' and levels 3 and 4 of 'ability to participate activities' (DCE$_{TTO}$ Model 4) (Table 5.3). All coefficients including duration coefficient estimated from this adjusted model were in the expected direction and were logically consistent. Of the 14 coefficients estimated, 10 were significant, including the coefficient for duration. Therefore, anchoring the coefficients to a full health-dead scale using the coefficient of duration continued based on DCE$_{TTO}$ Model 4.

Table 5.3: Parameter estimates from the unadjusted and adjusted DCE_{TTO} models (on the latent utility scale) in valuation approach 1, DCE_{TTO}

Variable	Model 1: Unadjusted model		Model 2: Combined 'difficulty eat/drink' level 1 and 2		Model 3: Combined 'ability to participate' level 3 and 4		Model 4: Combined 'difficulty eat/drink' level 1 and 2 / 'ability to participate' level 3 and 4	
	β	SE	β	SE	β	SE	β	SE
Life years	0.3912*	0.0475	0.4075*	0.04606	0.3882*	0.0470	0.4045*	0.0456
pain/discom2 × life years	–0.0537*	0.0193	–0.0570*	0.01899	–0.0540*	0.0192	–0.0571*	0.0189
pain/discom3 × life years	–0.1670*	0.0246	–0.1656*	0.02454	–0.1683*	0.0249	–0.1669*	0.0248
pain/discom4 × life years	–0.2054*	0.0244	–0.2048*	0.02415	–0.2049*	0.0243	–0.2043*	0.0241
difficult eat/drink2 × life years	0.0443*	0.0209			0.0453*	0.0208		
difficult eat/drink3 × life years	–0.0369	0.0213	–0.0598*	0.01850	–0.0372	0.0213	–0.0604*	0.0186
difficult eat/drink4 × life years	–0.0897*	0.0212	–0.1099*	0.01826	–0.0887*	0.0211	–0.1092*	0.0182
worried2 × life years	–0.0019	0.0192	–0.0039	0.01902	–0.0014	0.0190	–0.0032	0.0188
worried3 × life years	–0.0224	0.0174	–0.0227	0.01703	–0.0220	0.0172	–0.0222	0.0168
worried4 × life years	–0.0529*	0.0216	–0.0527*	0.02171	–0.0506*	0.0213	–0.0501*	0.0214
abilityparticipate2 × life years	–0.0404*	0.0208	–0.0357	0.02030	–0.0411*	0.0207	–0.0365	0.0201
abilityparticipate3 × life years	–0.0626*	0.0196	–0.0598*	0.01907	–0.0501*	0.0182	–0.0466*	0.0175
abilityparticipate4 × life years	–0.0344	0.0217	–0.0298	0.02107				
appearance2 × life years	–0.0288	0.0201	–0.0244	0.01961	–0.0279	0.0198	–0.0233	0.0194
appearance2 × life years	–0.0555*	0.0202	–0.0514*	0.01986	–0.0542*	0.0199	–0.0498*	0.0196
appearance2 × life years	–0.0584*	0.0229	–0.0532*	0.02229	–0.0568*	0.0226	–0.0518*	0.0220
Goodness-of-fit statistics								
Number of observations	3200		3200		3200		3200	
R^2	0.1535		0.1511		0.1524		0.1499	
AIC	1909.7		1912.94		1909.99		1913.54	
BIC	2006.84		2004.01		2001.05		1998.53	

**Significance at $p < 0.05$*

Anchoring based on DCE_{TTO}

Table 5.4 provides the utility decrements estimated from DCETTO Model 4, specified in the **Table 5.3**. The utility decrement for each dimension was estimated using the coefficient for the duration and the estimated utility decrements for each dimension are logically consistent and in the expected direction. The utility values based on the DCETTO approach range from -0.1422 to 1.

Table 5.4: Utility decrement estimates from the DCE_{TTO} models in valuation approach 1, DCE_{TTO}

Dimension	Level	Utility decrement (95% CI)
Pain/discomfort	1	0
	2	–0.1412 (–0.0513 to –0.2312)
	3	–0.4126 (–0.3196 to –0.5058)
	4	–0.5051 (–0.4068 to –0.6033)
Difficulty eating/drinking	1 & 2	0
	3	–0.1492 (–0.0674 to –0.2310)
	4	–0.2700 (–0.1843 to –0.3557)
Worried	1	0
	2	–0.0079 (–0.0826 to 0.0985)
	3	–0.0548 (–0.0258 to 0.1353)
	4	–0.1239 (–0.0288 to –0.2190)
Ability to participate activities	1	0
	2	–0.0902 (–0.0021 to 0.1826)
	3 & 4	–0.1152 (–0.0414 to –0.1890)
Appearance	1	0
	2	–0.0577 (–0.0339 to 0.1493)
	3	–0.1230 (–0.0309 to –0.2152)
	4	–0.1280 (–0.0314 to –0.2246)

From DCE_{TTO} Model 4, conditional logit, dummy coded

Valuation Approach 2 DCE_{VAS}

A total of 191 participants completed the DCEVAS survey. DCE data were analysed using conditional logit and the data were modelled (Unadjusted DCEVAS Model 1) including all 191 participants (**Table 5.5**). In DCEVAS Model 1, all coefficients were in the expected direction except for the coefficient for level 2 of the 'appearance' dimension. Further, the magnitude of the coefficients increased with the severity level of each dimension. Of the 15 coefficients estimated, four were non-significant. **Table 5.5** shows

the coefficient estimates of adjusted DCE$_{VAS}$ Model 2 combining the level 1 and 2 of the dimension 'appearance'. All coefficients were in the expected direction and logically consistent; of the 14 coefficients estimated, 11 were significant. Therefore, anchoring of DCE data on to full health-dead scale was completed based on adjusted DCE$_{VAS}$ Model 2.

Table 5.5: Parameter estimates from the unadjusted and adjusted DCE models (on the latent utility scale) in valuation approach 2, DCE$_{VAS}$

	Model 1: Unadjusted model including all participants (n = 191)		**Model 2: Adjusted model combining 'appearance' levels 1 and 2, including all participants (n = 191)**	
	β	**SE**	**β**	**SE**
pain/discom2	–0.1310	0.1021	–0.1322	0.1020
pain/discom3	–1.2830*	0.1488	–1.2837*	0.1486
pain/discom4	–1.368*	0.1516	–1.3694*	0.1513
difficult eat/drink2	–0.2844*	0.0981	–0.2843*	0.0981
difficult eat/drink3	–0.6419*	0.1241	–0.6404*	0.1246
difficult eat/drink 4	–0.7456*	0.1204	–0.7444*	0.1206
worried2	–0.2708*	0.1135	–0.2700*	0.1134
worried3	–0.4480*	0.1064	–0.4478*	0.1064
worried4	–0.4688*	0.1121	–0.4690*	0.1121
abilityparticipate2	–0.1064	0.1094	–0.1063	0.1093
abilityparticipate3	–0.3131*	0.1221	–0.3136*	0.1218
abilityparticipate4	–0.3356*	0.1167	–0.3369*	0.1162
appearance2	0.0379	0.1089		
appearance3	–0.1418	0.1151	–0.1600	0.0969
appearance4	–0.2776*	0.1278	–0.2966*	0.107
Goodness-of-fit statistics				
Number of observations	1028		1028	
R^2	0.2001		0.2001	
AIC	1724.34		1722.47	
BIC	1814.71		1806.82	

*Significance at $p<0.05$

Analysis of VAS data in valuation approach 2, DCE$_{VAS}$

Of the 191 participants, 93 met at least one of the exclusion criteria. Therefore, a subsample of 98 participants was included in the VAS data analysis. These participants valued six health states using VAS: best health state, worst health state, a mild health state, a moderate health state and a severe health state defined by the DCUI classification system. Raw VAS scores for the worst, a mild, a moderate and a severe health state were

adjusted to a QALY scale in VAS at the participant level using Equation 4 and the mean scores were taken as the adjusted VAS score for each health state. **Table 5.6** shows the unadjusted and adjusted mean VAS values for each health state.

Table 5.6: Unadjusted and adjusted mean VAS values for each health state (n = 98)

Variable	Unadjusted VAS score				Adjusted VAS score			
	Mean	Std. Dev.	Min	Max	Mean	Std. Dev.	Min	Max
Best health state (11111)	91.28	12.73	42	100	1.0000			
Mild health state (22211)	68.88	18.99	10	96	0.7376	0.1909	0	0.9596
Moderate health state (33341)	40.31	19.96	3	97	0.4257	0.1995	0.0309	0.8687
Severe health state (44431)	26.99	17.69	2	96	0.2728	0.1637	–0.0658	0.7111
Worst health state (44444)	14.73	15.24	0	93	0.1248	0.1214	–0.25	0.4949
Death	3.81	12.91	0	94	0.0000			

Sensitivity analysis

For sensitivity analysis, coefficients were estimated using a conditional logit model (DCE_{VAS} Model 3) by excluding the DCE data of the participants who met VAS exclusion criteria (n = 93) **(Appendix K)**. However, the coefficient for level 2 of the 'appearance' dimension estimated by this model was not in the expected direction and of the 15 coefficients estimated, four were non-significant. Further, in DCE_{VAS} Model 3, the coefficients for the dimension 'ability to participate activities' were logically inconsistent. Therefore, excluding the DCE data of the participants who met VAS exclusion criteria did not improve the model estimates.

Anchoring based on DCE_{VAS}

Two methods were applied to rescale the coefficients on to full health-dead scale: rescaling based on the worst health state in VAS data and mapping VAS onto DCE. The procedure for rescaling based on worst health state and mapping are provided in **Appendix L**. Anchoring of DCE data onto full health-dead scale was completed based on the adjusted DCE_{VAS} Model 2 specified in **Table 5.5** and effect coding was done to recover the base level for the purpose of rescaling. To rescale based on worst health state, a utility value for the worst health state (44444) in the DCE latent scale was calculated and then rescaled to the full health-dead scale using the mean adjusted VAS score for

worst health **(Table 5.6)**. To map VAS onto DCE, utility values for four health states (44444, 44431, 33341 and 22211) on the DCE latent scale were calculated and regressed to the adjusted VAS scores shown in **Table 5.6**. **Table 5.7** shows the coefficients estimated from the effect-coded adjusted DCE_{VAS} Model 2 and rescaled coefficients based on worst health state on VAS data and mapping VAS onto DCE. The estimated utility values for each dimension are logically consistent.

Table 5.7: Estimated and rescaled coefficients in valuation approach 2, DCE_{VAS}

Dimension	Level	Estimated coefficients*	Rescaled coefficients worst state[1]	Rescaled coefficients mapping[2]
Pain/discomfort	1	0.6963	0.3076	0.2569
	2	0.5641	0.2716	0.2226
	3	–0.5874	–0.0418	–0.0765
	4	–0.6731	–0.0651	–0.0987
Difficulty eating/drinking	1	0.4173	0.2208	0.1845
	2	0.1330	0.1434	0.1106
	3	–0.2231	0.0466	0.0181
	4	–0.3271	0.0183	–0.0089
Worried	1	0.2967	0.1833	0.1531
	2	0.0267	0.1098	0.0830
	3	–0.1511	0.0615	0.0368
	4	–0.1723	0.0557	0.0313
Ability to participate in activities	1	0.1892	0.1499	0.1252
	2	0.0829	0.1210	0.0976
	3	–0.1244	0.0646	0.0438
	4	–0.1477	0.0582	0.0377
Appearance	1 & 2	0.1522	0.1384	0.1156
	3	–0.0078	0.0949	0.0741
	4	–0.1444	0.0577	0.0386

** DCE_{VAS} Model 2, effect coded, conditional logit (Appendix L)*

Comparison of two valuation approaches: DCE_{TTO} and DCE_{VAS}

As the rescaling on to the full health-dead scale undertaken using different methods in the two valuation approaches, it was not possible to compare them directly based on the rescaled coefficients. Therefore, the two valuation approaches were compared based on the tariff values calculated using the DCE_{TTO} and DCE_{VAS} approaches. The utility decrements from **Table 5.4** provided the rescaled coefficients for calculating utility values based on the DCE_{TTO} anchoring approach. The utility value for a health state

is 1 minus the sum of the relevant utility weights of each dimension. For example, the worst health state based on DCE_{TTO} can be calculated as:

HS (44444) = 1 – (0.5051 + 0.2700 + 0.1239 + 0.1152 + 0.1280) = –0.1422

This contrasts with the utility decrements from **Table 5.7** which provided the rescaled coefficients for calculating utility values based on DCE_{VAS} anchoring approach. The sum of the relevant coefficient for each dimension provided the utility values based on the coefficients rescaled with the VAS worst health state, whereas the sum of the constant and the relevant coefficient for each dimension provided the utility values based on mapping. For example, the utility value for worst health state based on the PITS approach was 0.1248 and it was calculated as:

HS (44444) = –0.0651 + 0.0183 + 0.0557 + 0.0582 + 0.0577 = 0.1248

The utility value for worst health state based on the mapping approach was 0.1646 and it was calculated using a regression coefficient of 0.8354:

(1 – VAS) = 0.8354*(1 – DCE)

VAS = (1 – 0.8354) + 0.8354*DCE

Utility = 0.1646 + 0.8354*DCE

HS (44444) = 0.1646 + (–0.0987) + (–0.0089) +0 .0313 + 0.0377 + 0.0386
= 0.1646

Figure 5.5 shows the tariff values for DCUI health states that were calculated based on rescaled coefficients from the two valuation approaches. The two methods in the DCE_{VAS} (rescaling based on worst health state and mapping) produced largely similar tariff values. The DCE_{TTO} approach valued health states lower than the DCE_{VAS} approach; in particular, severe health states were valued worse than death.

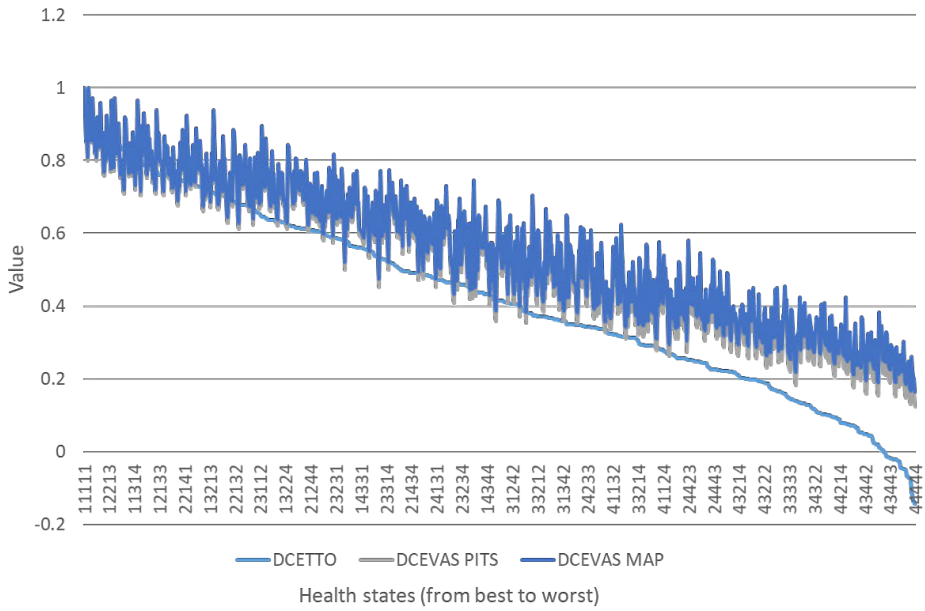


Figure 5.5: A comparison between estimated values of the DCUI health states using DCE_{TTO} as a base

5.5 Discussion

The aim of this study was to select the most suitable anchoring approach between DCE_{TTO} and DCE_{VAS} to develop a utility algorithm for health states defined by the DCUI, a new classification system developed for dental caries. This study provides evidence that the DCE_{VAS} is a feasible and reliable approach to anchor DCE utility values on to a full health-dead scale. VAS has not been tested widely as a method of anchoring DCE utility values in previous research. Hence, the present study has demonstrated the use of VAS as a feasible anchoring approach.

Several methods have been introduced to anchor DCE values onto a full health-dead scale (64, 66). However, techniques that require participants to trade-off health states with life years or immediate death may not be relevant for a condition like dental caries, as it is not associated with serious illness. Webb et al, 2020 first reported the transformation of DCE latent values using the worst health state of VAS (192). The present study further evaluated anchoring DCE values by the mapping VAS onto the DCE or using the worst health state of VAS. Moreover, this study includes a comparison of VAS with the DCE_{TTO} approach. Hence, these findings will facilitate future research using VAS as an anchoring approach, especially for conditions like dental caries.

In the DCE_{VAS} approach, modelled DCE data from both the unadjusted and adjusted models estimated coefficients that were more ordered than those from the

DCE$_{TTO}$ approach. This could be due to the fact that the DCE$_{VAS}$ models' coefficients were main effects DCEs, whereas the DCE$_{TTO}$ included interactions with duration. Further, the utility decrements estimated by DCE$_{TTO}$ were more dispersed and severe health states were valued lower than death. The model implied that the respondents do consider severe DCUI health states worse than death. However, dental caries is neither life-threatening nor associated with severe disability conditions. Therefore, the modelled data estimated from DCE$_{TTO}$ produced unreliable utility values for dental caries health states in which individuals would not be willing to die rather than living with severe dental caries in real life. A previous direct oral health state valuation study reported a similar finding (71). They compared utility values for 12 dental health states obtained from dental free-time trade-off (DFTO) and dental visual analogue scale (DVAS). They reported that the utility values resultant from the DVAS were in an expected and meaningful order (e.g., tooth states with pain had lower utilities than those without pain), whereas the utility values resultant from DFTO were highly skewed, which demonstrated that the participants did not trade off their free time to achieve better tooth health states (71).

This study also has certain limitations. The raw VAS data required adjustment to convert them to a 0–1 scale (169) before they were used to rescale the coefficients. The calculation of VAS adjusted score depended on the respondent's assigned relative VAS score values for the six health states. To obtain a legitimate answer for the adjusted VAS scores, participants should provide logically consistent answers for the health states being valued. However, a substantial number of participants provided illogical answers to at least one of the health states. The underlying reasons for these answers could not be established, as the survey was conducted online. Therefore, participants who valued health states illogically were excluded prior to the calculation of VAS adjusted scores to prevent the production of unreliable values. Similar exclusion criteria were used in previous VAS valuation studies (168).

Although the DCUI is intended for use among children older than 12 years, the health state valuations were collected from an adult sample due to the methodological constraints described in the methods section. Therefore, future research is needed to assess whether these comparison methods perform similarly in adolescent samples. Further, the study was conducted as an online survey and some participants did not complete the whole survey. We could not establish whether this was due to the difficulty of the valuation tasks in either approach. However, the self-reported data of the participants who completed the surveys revealed that there was no significant difference

in the difficulty of either understanding or completing the valuation tasks between the DCE$_{TTO}$ and DCE$_{VAS}$ approaches.

Conclusions

This study has demonstrated that the DCE$_{VAS}$ is a feasible and reliable method to anchor the utility values obtained from DCE on a latent scale onto a full health-dead scale in dental caries health state valuations compared to the DCE$_{TTO}$ approach.

Chapter 6. Valuing a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI): Using DCE and VAS to develop an Australian utility algorithm

STATEMENT OF CONTRIBUTION TO CO-AUTHORED PUBLISHED PAPER

This chapter includes a co-authored paper prepared for submission. The bibliographic details of the paper, including all authors, are:

Hettiarachchi RM, Kularatna S, Byrnes J, Chen G, Mulhern B, Scuffham PA. Valuing a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI): Using DCE and VAS to develop an Australian utility algorithm

My contribution to the paper involved the designing the study and the survey, planning and executing the data collection, analysing the data and drafting the article.

(Signed) _____ (Date)_____

Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Corresponding author of paper: Ruvini Hettiarachchi

(Countersigned) _____ (Date)_____

Supervisor: Prof. Paul Scuffham

Valuing a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI): Using DCE and VAS to develop an Australian utility algorithm

Ruvini M Hettiarachchi, Sanjeewa Kularatna, Joshua Byrnes, Brendan Mulhern, Gang Chen, Paul A. Scuffham

6.1 Abstract

Introduction

The Dental Caries Utility Index (DCUI) is a new health state classification system for dental caries that consists of five domains: pain/discomfort, difficulty eating food/drinking, worried, ability to participate in activities, and appearance. Each domain has four response levels. The aim of this study is to generate an Australian-specific utility algorithm for the DCUI classification system to facilitate health economic evaluation.

Methods

An online survey was conducted among an age and sex representative sample of the adult Australian general population. Participants completed a set of discrete choice experiment (DCE) tasks, visual analogue scale (VAS) tasks, sociodemographic characteristics, the DCUI, a generic preference-based measure (EQ-5D-5L) and an oral health quality of life instrument (OHIP-14). The DCE was used to elicit the preferences on five domains and then the latent utilities were anchored onto death-full health quality-adjusted life years scale combining results from separate VAS tasks. DCE data were modelled using conditional logit and two anchoring procedures were considered, i.e. to either anchor based on the worst heath state, or to adopt a mapping approach. The optimal anchoring procedure was selected based on the mean absolute error (MAE).

Results

A total of 995 adults from the Australian general population completed the survey. The conditional logit estimates on five dimensions and levels were all statistically significant in the expected direction and order (i.e., greater utility decrements were associated with increasing severity levels), except for the second level of the 'worried' and 'appearance' domains. Between two anchoring procedures, the mapping approach was selected based on a smaller MAE. The Australian-specific tariff of DCUI ranges from 0.1681 to 1.0000.

Conclusion

This study developed a utility algorithm for the DCUI. This value set will facilitate utility value calculations from the participants' responses for DCUI application in economic evaluations of dental caries interventions.

Key words: Dental Caries, Adolescents, Valuation, Discrete choice experiments, Preference-based, Visual analogue scale

6.2 Introduction

Cost-utility analysis (CUA) is the preferred type of full economic evaluation for most health technology assessment authorities to assess health care interventions across different disease areas. Quality-adjusted life years (QALYs) are the most common outcome measure in CUA (44) and allow comparison of health interventions in terms of QALY gain or loss for an incremental cost. The QALY is a summary outcome measure that combines utility values assigned to the health states and the duration of time spent in each health state in the condition of interest (162). These health state utility values represent individuals' preferences for different health states and are presented on a full health-dead (1–0) scale (53).

Preference-based quality of life measures (PBMs) can be used to derive utility values for QALYs in economic evaluations. PBMs are pre-scored and utility value sets are readily available. Thus, they are easy to use and less time-consuming than directly eliciting preferences from patients (110). PBMs consist of two components: a health state classification system and a set of utility values (a scoring algorithm). The classification system includes health states described in terms of a set of dimensions with response levels relevant to general health or certain disease conditions. The scoring algorithm, enables the generation of utility values for these health states (60, 61). The algorithms are generated using the values for selected health states in the classification system, which are estimated from a sample of patients or general population preferences using a preference-elicitation method. Once these values are obtained, statistical modelling techniques are used to generate the scoring algorithm and to estimate the utility values for all possible health states derived from the classification system.

The existing paediatric oral health-related quality of life instruments are non-preference-based and hence, cannot be used to calculate utility values in health economic

evaluations (43, 148). Therefore, in the first phase of this study (164) a classification system for dental caries-the Dental Caries Utility Index (DCUI)-was developed. The DCUI consists of five items: pain/discomfort, difficulty eating food/drinking, worried, ability to participate in activities and appearance and each item comprises four levels **(Table 6.1)**. The aim of the present study was to generate a preference-based algorithm for the DCUI classification system. A detailed protocol of this study has been submitted elsewhere (191).

6.3 Methods

Preference elicitation technique

Discrete choice experiments (DCEs) have recently emerged as one of the preferred methods for health state valuations (65). The methodological aspects of DCE valuations have been tested widely and used in health state valuation studies defined by a classification system. DCE surveys are relatively easy to understand and are typically compatible with the online surveys; thus, it offers the added advantage over traditional valuation methods, such as the standard gamble (SG) and the time trade-off (TTO) methods (55). Therefore, the DCE method was used as the preference elicitation technique for the present study. However, DCEs produce utility values on a latent scale. These values should be anchored onto the full health-to-dead scale to calculate the QALYs (193). Several methods (e.g., such as DCE with duration and use of external data from a concurrent TTO study) have been applied to anchor latent DCE values onto a full health-to-dead scale (65). Dental caries is not a severely disabling condition in which patients would be willing to die rather than remain in a severe health state. Therefore, anchoring approaches require a trade-off of life years or else a choice between the dental caries health state and immediate death would be inappropriate for the valuation. Visual analogue scale (VAS) health state valuation tasks are comparatively easy to understand and a lower burden to participants than the SG and TTO methods (169) because they do not attach any trade-off between life years and risk. The VAS approach has been used to generate utility values in several health state valuation studies (168, 194) and oral health studies (71). It has also been used solely as an anchoring approach to rescale DCE utility values onto a full heath-dead scale (192). Two pilot studies were conducted prior to the main survey to evaluate the feasibility and suitability of DCE anchoring with a VAS approach (see Chapter 5 for more details) and compare it to DCE with duration approach

(DCE$_{TTO}$). Pilot data analyses indicated that the DCE anchoring with a VAS approach is more preferable over DCE$_{TTO}$ for a dental caries health state valuation and consequently was chosen for the main survey.

DCE experimental design and choice tasks

The DCUI classification system has 1,024 (4^5) possible health states **(Table 6.1)**. As full factorial design is not feasible, a widely used D-efficient design was adopted to select a manageable 200 pairwise choice tasks of these health states (which were further blocked into 25 versions) using Ngene software (172). This is in line with a recent structured review which identified that most health state valuations studies using DCE included more than 151 choice tasks (65) and the number of choice tasks per respondent ranged from 2 to 108 (65)**.** The values for the coefficients, derived from the pilot study *DCE$_{VAS}$* (see Chapter 5 for further details), were used to define the priors for the D-efficient design in the main survey. An Ngene design was coded with few specifications, such as adding restrictions to eliminate dominant choice tasks from the design. However; the Ngene was unable to yield a D-efficient design with these prior values along with the restrictions to eliminate the dominant tasks. After consulting with the experts in DCE research, it was decided that zero priors would be used for all dimensions and levels in the D-efficient design for the main survey as well; as indicated in the literature, non-zero prior values are not essential for an optimal study design (166). Therefore, the final experiment design in the main survey was the same as the experimental design used in the pilot study *DCE$_{VAS}$* with zero priors. The final experimental design was manually checked and no implausible attribute-level combination was found. The Ngene design codes and DCE design matrix are provided in **Appendices G and H,** respectively. An example of DCE choice task is presented in **Figure 6.1**.

VAS task

A VAS task was included after the DCE choice tasks to facilitate re-scaling the DCE estimates onto the QALY scale. The task included six health states in a single VAS scale: best health state (11111), worst heath state (44444), mild health state, moderate health state, severe health state (defined by the DCUI classification system) and death **(Figure 6.2)**. The mild, moderate and severe health states were selected to represent combinations of attribute levels with differing severity and pilot study data were used to select combinations of significant coefficients to denote these health states. The two

extreme endpoints of the VAS scale were calibrated as 'best imaginable oral health' (Score 100) and 'worst imaginable oral health' (Score 0).

Table 6.1: Dental Caries Utility Index (DCUI) Classification System*

Dimension	Description
Pain/Discomfort	1. I have no pain or discomfort 2. I have a little pain or discomfort 3. I have quite a lot of pain or discomfort 4. I have lots of pain or discomfort
Difficulty eating food/drinking	1. I have no difficulty in eating food/drinking 2. I have a little difficulty in eating food/drinking 3. I have quite a lot of difficulty in eating food/drinking 4. I have lots of difficulty in eating food/drinking
Worried ***(e.g. about losing a tooth, etc.)***	1. I am not worried 2. I am a little bit worried 3. I am quite worried 4. I am very worried
Ability to participate in activities ***(e.g. playing with your friends, sports, school work, etc.)***	1. I have no difficulty participating in activities 2. I have a little difficulty participating in activities 3. I have quite a lot of difficulty participating in activities 4. I have lots of difficulty participating in activities
Appearance	1. I am not concerned about my appearance 2. I am a little concerned about my appearance 3. I am quite concerned about my appearance 4. I am very concerned about my appearance

**Hettiarachchi et al.(164)*

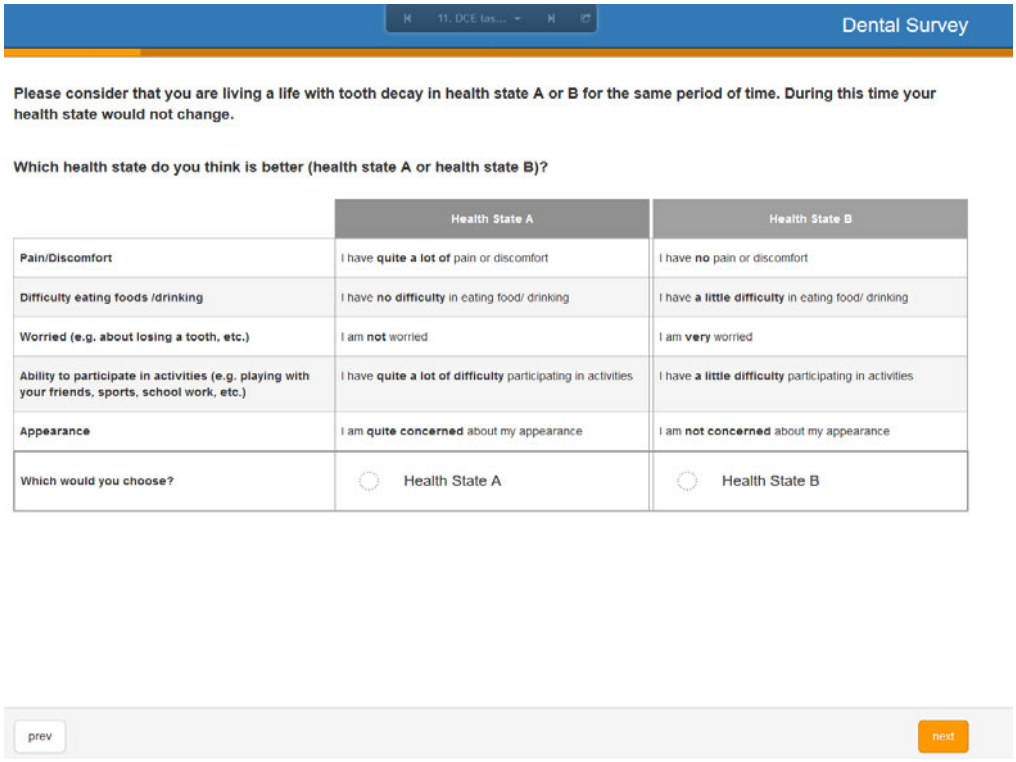


Figure 6.1: An example choice set from the discrete choice experiment valuation task

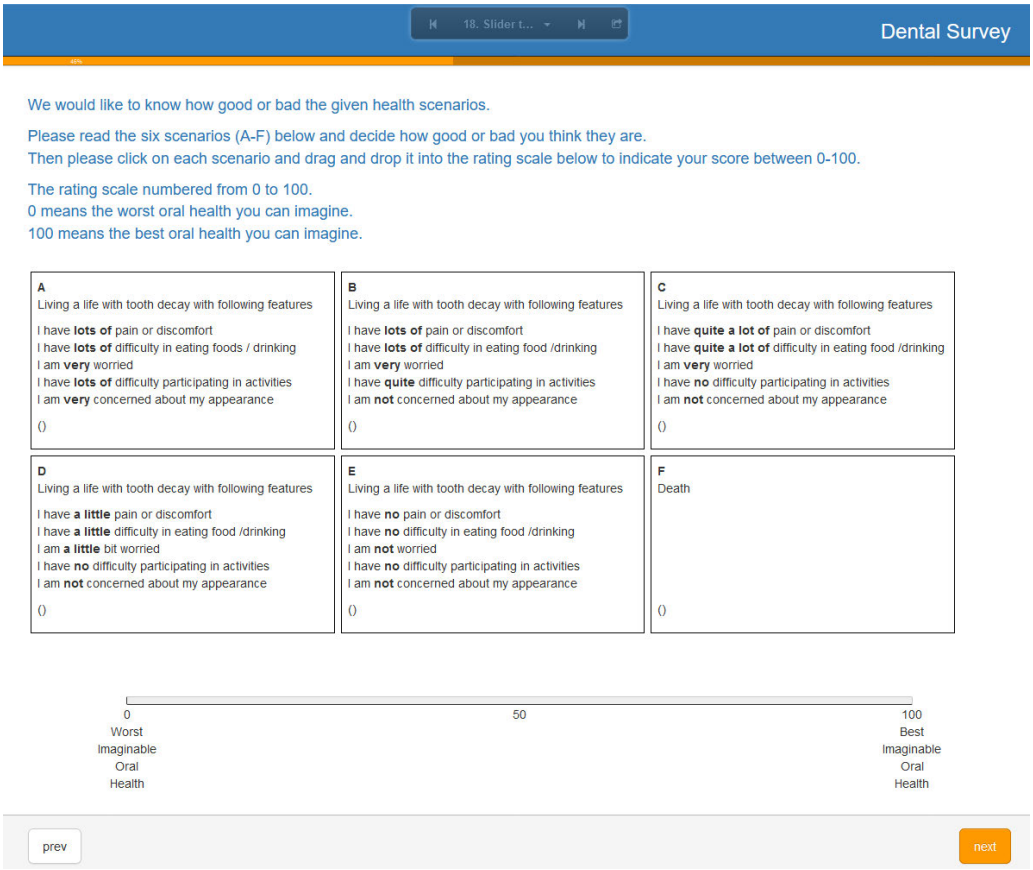


Figure 6.2: The VAS valuation task

Study sample

There exists several options to generate utility algorithms for existing paediatric PBMs, including eliciting preferences from children and adolescents themselves, or from adult general population, or based on proxy responses from parents/guardians (195). Eliciting preferences from a paediatric sample may be a better option to understand their preference given they tend to have different preferences for health states than adults (70, 161); however, it is methodologically challenging, as the elicitation tasks require higher cognitive ability. Another major concern is the ethical issues associated with presenting tasks with the notion/concept of 'dead'. Consequently, it is more common to elicit preferences from adult general population samples to generate utility algorithms for paediatric PBMs (68, 175). This approach is often justifiable, as tax payers (adult general population members) should have greater influence in deciding which health interventions are funded through publically. Furthermore, the anchoring task based on VAS also required participants to value 'death' in relation to the five other health states defined by the DCUI classification system. Considering these facts, the present study elicited preferences for health states from an adult general population sample. Choice tasks were designed in the perspective of participants' 'own health', as dental caries is one of the most prevalent conditions among both children and adults and adults are more likely to experience dental caries in their lives.

Sample recruitment and survey administration

According to Lancsar and Louviere (181), 20 respondents per choice set and a sample size of 1000–2000 is sufficient to estimate a reliable model. Although the use of a larger sample was planned during the study protocol, due to time and financial limitations, the health state valuation study aimed to recruit a sample size of 1000 members from the Australian general population. This sample size was compatible with the sample size requirement and also with other Australian online DCE surveys (178). A representative sample of the adult Australian general population in relation to age and gender was recruited from an Australian online panel via a research company SurveyEngine (http://www.surveyengine.com). Invitations were sent to the potential participants who were registered with the survey company and interested participants accessed the survey using the provided link. The online survey **(Appendix I)** started with the introduction page, where participants were given all the necessary details about the research project and then requested to provide consent for data collection and use. Those

who consented were guided through the online survey by screen prompts and were able to complete the tasks at their convenience. The next section of the survey included screening questions (age and sex) to ensure the inclusion of a representative sample of the Australian general population. The participants were then asked to complete the DCUI classification system with respect to their own oral health as a 'warm-up' task to familiarise them with the words used in the choice tasks. The next section included a set of DCE choice tasks—a practice choice task and a task with a dominant choice—to familiarise the participants with DCE tasks. These were followed by the eight-DCE tasks and the VAS anchoring task. The participants were then asked to complete a generic preference-based QoL instrument (EQ-5D-5L (183)) **(Appendix J)**, an oral health-specific QoL instrument (Oral health impact profile-14, OHIP-14 (184)) and questions regarding their sociodemographic characteristics, oral health status and frequency of dental visits. The OHIP-14 and EQ-5D-5L are the most frequently used instruments in oral health research to evaluate oral health and general health-related QoL in Australian adults (196, 197). Ethical approval for this study was obtained from the Human Research Ethics Committee, Griffith University (HREC/2019/550) **(Appendix D)**.

Data analysis

Sample characteristics

Chi-square tests were used to assess the representativeness of the study sample against Australian general population, which were drawn from Australian Bureau of Statistics data (198-200) as well as the Household, Income and Labour Dynamics in Australia (HILDA) Survey wave 16 (201).

Utility estimation

The data were analysed using the STATA version 15.1 (188) statistical software package. Data from DCE choice tasks were modelled using a conditional logit model under a random utility framework, as specified in equation 1. The random utility framework assumes that respondents choose the option that maximises their utility (189). The utility function consists of a vector of observable attributes as well as a random error term (67):

$$\mu_{ij} = \beta_1 x_1 + \beta_2 x_2 + \beta_3 x_3 + \beta_4 x_4 + \beta_5 x_5 \rightarrow Equation\ 1$$

μ_{ij} = observable component of the utility function

β = coefficient for each dimension

x = vector containing five DCUI dimensions, each of which is estimated using three dummy variables (with 'no problems' serving as the reference level within each attribute)

The best-fit model was selected based on the statistical significance and logical order of the coefficients. The base level of the effect-coded model was recovered using the formula [$L_1 = -1 * (L_2 + L_3 + L_4)$] prior to anchoring (202).

Anchoring

The VAS data first underwent a quality check to make sure that meaningful results are attained, i.e. participants needed to give logical answers regarding the death, best or worst health states. Strict exclusion criteria were applied to remove all relevant logical inconsistencies in order to achieve meaningful results in the anchoring procedure. The participants were excluded if they met any of the following criteria:

- Participants valued best ≤ death or worst or mild or moderate or severe.
- Participants valued death ≥ mild or moderate.
- Participants valued worst ≥ mild or moderate or severe.
- Participants were missing either death, best or worst VAS data (due to a technical issue in the VAS slider task, some missing data exist for participants who initially completed the survey).

The raw VAS scores of the worst, mild, moderate and severe health states were converted to the full health-dead scale based on Equation 2 at the participant level proposed by Brazier et al. (169), as used in in previous studies (168).

$$V_h = \frac{S_h - S_{dead}}{S_{11111} - S_{dead}} \quad \rightarrow \quad Equation\ 2$$

V_h = adjusted VAS rating for health state *h*

S_h = respondent's unadjusted VAS score for state *h*

S_{dead} = respondent's assigned VAS score for the health state 'death'

S_{11111} = respondent's assigned VAS score for a state 11111 (best state)

DCE data were anchored to the full health-dead scale using adjusted VAS scores via two methods: (i) anchoring with the worst (PITS) health state and (ii) anchoring by mapping DCE onto VAS. DCE data were anchored to the worst-state value obtained in

VAS using the adopted formula from Rowen et al. (66) (in which the TTO instead of VAS was used):

$$\beta_{r\lambda\Theta} = \beta_{\lambda\Theta} * \frac{wVAS}{wDCE} \rightarrow Equation\ 3$$

$\beta_{r\lambda\Theta}$ = rescaled coefficient for level λ of dimension Θ

$\beta_{\lambda\Theta}$ = coefficient for level λ of dimension Θ

wVAS= estimated VAS value for the worst state generated using Equations 1

wDCE= DCE value for the worst state estimated using the DCE model

For the anchoring with mapping DCE onto VAS using a linear regression, mean VAS values obtained for the worst heath state (44444), a mild health state, a moderate health state and a severe health state of the DCUI were used as shown in Equation 4:

$dVAS_j = 1 - VAS_j$

$dDCE_j = 1 - DCE_j$

$$dVAS_j = f(dDCEj) + \varepsilon_j \rightarrow Equation\ 4$$

VAS_j = mean VAS value of health state *j*

DCE_j = modelled latent utility value of health state *j*

$dVAS_j$ = mean VAS disutility value of health state *j*

$dDCE_j$ = modelled latent disutility value of health state *j*

ε = error term

The optimal anchoring procedure, among anchoring with the worst (PITS) health state and anchoring by mapping DCE onto VAS, was selected based on the goodness-of-fit mean absolute error (MAE). The MAE provides a measure of fit of predicted values relative to observed or criterion values, with a small MAE being preferred (203). Since there are no directly elicited health state utilities for the vast majority of DCUI health states, VAS health state utilities for the small number of health states included in the VAS task were used as the criterion to evaluate the MAE for the two anchoring approaches. The utility algorithm for the DCUI health states was developed based on the approach with a smaller MAE (204).

6.4 Results

A total of 995 participants completed the survey. Table 6.2 shows the sociodemographic characteristics, health characteristics and self-reported oral health data of the study participants. The sociodemographic characteristics were compared with those of the Australian general population and the sample was more educated and had higher weekly household income than the Australian general population.

Table 6.2: Characteristics of the study sample (n=995)

Characteristic	Sample number	%	Population value[@]	χ2 statistic	P-value*
Mean age (SE)	46.87 (0.583)				
Age (years)					
18–29	210	21.11	21.76	0.83	0.9754
30–39	192	19.30	18.55		
40–49	164	16.48	16.62		
50–59	161	16.18	15.60		
60–69	129	12.96	13.22		
70 or older	139	13.97	14.25		
Sex					
Male	479	48.14	49.2	0.45	0.5039
Female	516	51.86	50.8		
Highest level of education					
Year 11 or below	115	11.56	17.45	600.56	< 0.0001
Completed high school	155	15.58	12.95		
Trade certificate	136	13.67	25.45		
Diploma/advanced diploma	160	16.08	10.8		
Bachelor's degree and above	429	43.12	33.4		
Gross weekly household income AUD					
Less than $1000	329	33.07	29.23	128.22	< 0.0001
$1,000–2,999	360	36.18	47.46		
$3,000–4,499	139	13.97	14.69		
$4,500–5,999	75	7.54	4.91		
$6,000 or more	92	9.25	3.71		
Marital status*, n (%)					
Never married	269	27.04	35.03	57.91	< 0.0001
Married/de facto	590	59.30	48.05		
Divorced/separated	96	9.65	11.74		
Widowed	29	2.91	5.18		
Prefer not to say	11	1.11	-		
DCUI score mean (SD)	8.01 (3.12)		n/a		
EQ5D Utility Score mean (SD)[#]	0.74 (0.30)		n/a		
EQ 5D -VAS Score mean (SD)	73.84 (19.03)		n/a		
OHIP14 Score mean (SD)	26.21 (13.45)		n/a		

Characteristic	Sample number	%	Population value[@]	χ2 statistic	P-value*
Oral Health status					
Excellent	145	14.57			
Very good	277	27.84			
Good	315	31.66			
Fair	173	17.39			
Poor	85	8.54			
Degree of tooth or mouth bother in everyday life					
Not at all	381	38.29			
A little	363	36.48			
Some	131	13.17			
A lot	58	5.83			
Very much	62	6.23			
Dental visits			n/a		
Once every 6 months	442	44.42			
When there is an oral health problem	459	46.13			
Never	94	9.45			
Private insurance for dental treatments			n/a		
Yes	484	48.64			
No	511	51.36			
Experienced toothache in the last 6 months			n/a		
Yes	326	32.76			
No	669	67.24			

[@]Australian age and sex distribution, gross weekly household income and registered marital status were derived from Australian Bureau of Statistics data (198-200). Education levels were derived from the Household, Income and Labour Dynamics in Australia Survey wave 16 (201).

**Chi-square goodness-of-fit test was used to compare observed values with general population values with continuity correction for categorical data*

[#]EQ-5D-5L utility weights were derived from Australian utility weights for EQ-5D-5L (205)

DCE data estimates

DCE choice data were modelled using conditional logit. Model 1 in Table 6.3 reports parameter estimates from the unadjusted model including all participants (n = 995). All coefficients were in the expected direction (all coefficient moves from level 1 of each dimension were valued negatively) and in the expected order for all five dimensions. All coefficients are significant except for level 2 of the dimensions 'worried' and 'appearance'. The conditional logit estimates from **Table 6.3** were used for anchoring in the next stage.

Table 6.3: Parameter estimates from conditional logit model

	Model 1: Unadjusted model including all participants (n = 995)	
	β	**Robust SE**
pain/discom2	–0.2727	0.0451***
pain/discom3	–1.2882	0.0626***
pain/discom4	–1.4026	0.0649***
difficult eat/drink2	–0.1731	0.0433***
difficult eat/drink3	–0.5844	0.0515***
difficult eat/drink 4	–0.6296	0.0505***
worried2	–0.0835	0.0466
worried3	–0.2181	0.0466***
worried4	–0.3767	0.0494***
abilityparticipate2	–0.1649	0.0463***
abilityparticipate3	–0.3466	0.0479***
abilityparticipate4	–0.3637	0.0488***
appearance2	–0.0010	0.0464
appearance3	–0.1364	0.0502**
appearance4	–0.2707	0.0527***
Goodness-of-fit statistics		
Number of observations	7960	
R^2	0.1856	
AIC	9017.09	
BIC	9132.22	

**Significant at $p < 0.01$ ***Significant at $p < 0.001$

Analysis of VAS data

In total, 521 participants were included in the VAS data analysis. The participants included for VAS data analysis were significantly different from the participants who met the VAS exclusion criteria in relation to age, education level and oral health status **(Appendix M).** Raw VAS scores for the worst, mild, moderate and severe heath states were adjusted to the full health-dead scale in VAS at the individual level using Equation 2 and the mean scores were taken as the adjusted VAS scores for each health state. **Table 6.4** shows the unadjusted and adjusted mean VAS values for each health state.

Table 6.4: Unadjusted and adjusted mean VAS values for each health state (n=521)

Variable	Unadjusted VAS score				Adjusted VAS score			
	Mean	Std. Dev.	Min	Max	Mean	Std. Dev.	Min	Max
Best health state (11111)	90.56	14.01	10	100	1.0000			
Mild health state (22211)	66.70	19.82	8	98	0.7253	0.1916	0	0.9899
Moderate health state (33341)	40.47	18.62	3	97	0.4333	0.1970	0.0204	1
Severe health state (44431)	27.29	15.53	0	96	0.2786	0.1755	-0.4062	0.9383
Worst health state (44444)	14.31	11.70	0	93	0.1264	0.1553	-0.8750	0.8842
Death	3.00	8.14	0	94	0.0000			

Sensitivity analysis

For sensitivity analysis, DCE data were modelled by excluding the data of the participants who met VAS exclusion criteria and the coefficients were estimated using a conditional logit model (Model 2, **Appendix M)**. As can be seen, except for the coefficient for level 2 of 'appearance', all coefficients estimated in Model 2 were in the expected direction. Coefficients for each dimension were in the expected order. Of the 15 coefficients estimated from Model 2, two coefficients (level 2 of the dimensions 'worried' and 'appearance') were non-significant. Exclusion of the DCE data of the participants who met VAS exclusion criteria did not improve the model estimates substantially, indicating that it was not necessarily correct to assume that those who failed the VAS task could not complete the DCE tasks. Therefore, anchoring of the DCE coefficients on to a full health-dead scale was performed based on Model 1 (Table 6.3).

Anchoring modelled DCE coefficients to the full health-dead scale using VAS data

The steps of rescaling based on worst health state and mapping are provided in **Appendix N** and were derived from the studies to develop a new scoring algorithm for CHU9D (204, 206). Model 1 specified in the Table 6.3 was effects coded **(Appendix N)** and the base level was recovered. To rescale it based on PITS heath state, effect coded coefficients in the DCE latent scale were rescaled based on the adjusted mean VAS score for the worst health state 0.1264 **(Table 6.4)**. The calculated utility values for four health states (22211, 33341, 44431 and 44444) in the DCE latent scale **(Appendix N)** were 0.8263, 0.2610, 0.0946 and 0.0000, respectively. To map VAS onto DCE, these calculated utility values were regressed onto the adjusted VAS scores shown in **Table 6.4**, as outlined in Equation 4. The estimated coefficient was 0.8319. **Table 6.5** shows the coefficient estimates from effect coded Model 1 and rescaled coefficients based on the worst heath state of VAS data and mapping VAS onto DCE.

Table 6.5: Estimated and rescaled coefficients

	Level	Estimated coefficient*	Rescaled coefficient based on worst state	Rescaled coefficient based on mapping approach
Pain/discomfort	1	0.7409	0.3399	0.2827
	2	0.4682	0.2615	0.2081
	3	–0.5473	-0.0301	-0.0695
	4	–0.6617	-0.0629	-0.1008
Difficulty eating/drinking	1	0.3468	0.2103	0.1750
	2	0.1737	0.1606	0.1276
	3	–0.2376	0.0425	0.0152
	4	–0.2828	0.0295	0.0028
Worried	1	0.1696	0.1521	0.1265
	2	0.0861	0.1281	0.1037
	3	–0.0485	0.0894	0.0669
	4	–0.2071	0.0439	0.0235
Ability to participate activities	1	0.2188	0.1682	0.1400
	2	0.0539	0.1209	0.0949
	3	–0.1278	0.0687	0.0452
	4	–0.1449	0.0638	0.0405
Appearance	1	0.1020	0.1299	0.1080
	2	0.1010	0.1296	0.1078
	3	–0.0344	0.0907	0.0707
	4	–0.1686	0.0521	0.0340

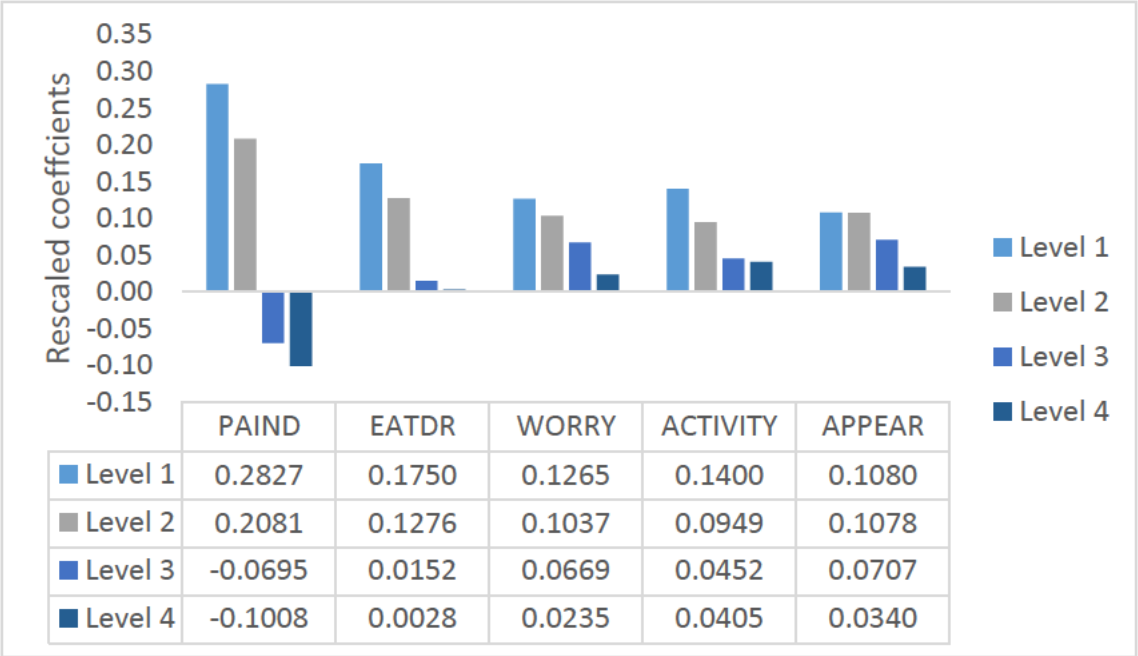
**Model 1in Table 6.3 was effects coded (Appendix N), conditional logit*

The utility decrements from **Table 6.5** provided the weights for calculating utility values for the health states defined by the DCUI. The goodness-of-fit mean absolute error (MAE) value indicated that the DCE estimates based on the mapping approach had the lowest MAE and, thus, performed better than the estimates based on the PITS approach **(Table 6.6)**. Therefore, the utility algorithm was developed based on the rescaled coefficients from the mapping approach.

Table 6.6: Comparison of two rescaling approaches

Health state	DCUI classification	VAS scores	Latent DCE estimates	Rescaled estimates based on PITS approach	Rescaled estimates based on Mapping approach
1	22211	0.7253	0.8263	0.8483	0.8555
2	33411	0.4333	0.2610	0.3544	0.3852
3	44431	0.2786	0.0946	0.2091	0.2468
4	44444	0.1263	0.0000	0.1264	0.1681
MAE	-	-	-	0.0679	0.0629

Figure 6.3 shows the rescaled coefficients (attribute weights) of each dimension based on the mapping approach.



	PAIND	EATDR	WORRY	ACTIVITY	APPEAR
Level 1	0.2827	0.1750	0.1265	0.1400	0.1080
Level 2	0.2081	0.1276	0.1037	0.0949	0.1078
Level 3	-0.0695	0.0152	0.0669	0.0452	0.0707
Level 4	-0.1008	0.0028	0.0235	0.0405	0.0340

(Derived from Model conditional logit, PAIND Pain/discomfort, EATDR Difficulty eating food/ drinking, WORRY Worried, ACTIVITY Ability to participate in activities, APPEAR Appearance).

Figure 6.3: Rescaled coefficients for Dental Caries Utility Index

The 'pain/discomfort' domain was associated with the highest attribute weight decrements for each level, followed by 'difficulty eating food/drinking'. Smaller decrements were observed in the 'appearance' domain (mainly between levels 1 and 2). The utility value for a health state can be calculated as the sum of the constant and the relevant coefficient for each dimension. For example, the utility value for worst health state based on the mapping approach was 0.1681 and it was calculated using a regression coefficient of 0.8319 as:

$$(1 - VAS) = 0.8319*(1 - DCE)$$

$$VAS = (1 - 0.8319) + 0.8319*DCE$$

$$Utility = 0.1681 + 0.8319*DCE$$

$$HS\ (44444) = 0.1681 + (-0.1008) + 0.0028 + 0.0235 + 0.0405 + 0.0340 = 0.1681$$

6.5 Discussion

We estimated a utility algorithm for the newly developed PBM for dental caries (DCUI) using an online survey with DCE and VAS approaches, and an age and sex representative sample of the adult general population in Australia. The value set estimated from this utility algorithm is the first value set for this newly developed PBM for dental caries. We used conditional logit and the resulting model was logically consistent for all dimensions in which utility decreases as severity increases. A logically consistent model is required to apply the utility algorithm in policy decisions (68). Therefore, this value set enables the use of DCUI in economic evaluation of dental caries interventions among Australian adolescents. To the best of our knowledge, the classification system and utility algorithm of DCUI are the first preference-based measures available for use in the economic evaluation of dental caries interventions among this population.

The 'pain/discomfort' domain had larger attribute weight decrements than the others, followed by 'difficulty eating food/drinking'. This indicates that the average respondents opposed to have more severe pain or discomfort. The domains 'worried' and 'appearance' showed the smallest decrement from level 1 to 2, which indicated that moving between these levels does not significantly affect the utility value. Previous valuation studies in oral health were mainly confined to preferences for dental treatments (207) or hypothetical vignette-based health states such as 'painful tooth' (47). Although Bellamy et al. (208) developed a multi-attribute utility scale for periodontal disease, we could not compare the utility values obtained from our study with this scale, as its health states were not valued using TTO, SG or any other valuation methods, and all the dimensions are different from those in DCUI. Therefore, we could not compare utility values obtained from our study directly with previous oral health research. However, it has been shown that pain is an important factor influencing dental health state utility values (71, 209).

The DCUI worst health state (have lots of pain or discomfort, lots of difficulty in eating food/drinking, very worried, have lots of difficulty participating in activities and very concerned about appearance) has a utility weight 0.1681. It is lower than the utility weights for the PITS health state in CHU9D. Currently, there are three specific algorithms available for CHU9D. The CHU9D PITS health states based on the UK adult general population (210), the Australian adult general population–specific scoring algorithm (176) and the Australian adolescent-specific scoring algorithm (67) were 0.337, 0.334 and

0.21, respectively. Further, the DCUI worst health state utility weight was lower than the worst health state values in other CSPBMs developed for non-fatal disease conditions. For example, a general population sample valued the worst health state of the PBM developed for atopic dermatitis in children as 0.36 (211). The PITS state of the Asthma Quality of Life-5D (AQL-5D) was 0.39 (212). However, it should be noted that the valuation techniques used in these CSPBMs is quite different from the technique used in DCUI health state valuation.

The VAS approach has been used in previous oral health studies as a direct health state valuation method to value dental health states' utilities (71, 213). However, the health states valued in these studies were not based on a classification system. Further, the VAS approach has not been used to anchor the utility values obtained from DCE studies, except for in a recent valuation study for EQ-5D health states (192). In this study, the authors identified the VAS approach as a feasible method to anchor DCE results to a full health-dead scale and noted that it is more appropriate for a disease condition in which participants may be reluctant to trade-off life years to achieve a better health state. Dental caries is a not a life-threatening condition under normal circumstances. Thus, the present study also supported VAS as a feasible and reliable approach to anchor the utility values obtained from DCE in a latent scale onto a full health-dead scale.

This study has some limitations. The valuation sample was large, and an age and sex representative sample was taken from the adult Australian general population. However, the sample's other sociodemographic characteristics were significantly different from the Australian population average and we could not evaluate how these differences affect health state preferences. Further, although this measure is intended for use among adolescents, health state valuations were conducted among an adult sample due to the methodological constraints described in the methods section. Therefore, future research is needed to assess whether these health state utility values are significantly different from the preferences elicited from an adolescent sample, as well as for populations in other geographical locations. Further, the study was conducted as an online survey. Some participants did not complete the whole survey and others provided logically inconsistent answers for the VAS task. We were unable to establish reasons for these anomalous responses. Further, the VAS task included six healthy states to be valued in a single VAS scale. Due to the technical difficulties, we were not able to randomise the order of these six states. Therefore, it is worth evaluating whether their order in the VAS task affected the VAS score assigned to each health state. However, we performed

sensitivity analysis (Table 6.3, Model 2) by removing the participants who met VAS exclusion criteria and found that their exclusion did not improve the model estimates. This indicates that the inability to understand the VAS task did not affect the performance in DCE tasks.

Conclusion

This study provides the first utility algorithm for the DCUI PBM for dental caries. This value set will facilitate future utility value calculations from participants' responses for DCUI in economic evaluations of dental caries interventions. Further, the present study also supports VAS as a feasible and reliable approach to anchor the utility values obtained from DCE in a latent scale onto a full health-dead scale, especially for a condition like dental caries in which participants may be reluctant to trade life years to achieve better quality of life.

Chapter 7. Discussion

7.1 Introduction

This chapter discusses the main thesis components: Literature review (Chapter 2), development of the classification system for dental caries utility index – DCUI (Chapter 3), development of methods for health state valuation (Chapter 4), comparison between two valuation approaches (Chapter 5) and establishment of the DCUI scoring algorithm (Chapter 6). The strengths and limitations of the study are also discussed and followed by recommendations for future research.

Cost-utility analysis (CUA) is the type of full economic evaluation preferred by most health technology assessment agencies around the world to prioritise health care interventions (37-40). CUA compares the incremental cost of a programme with the resulting health gain, in the form of a summary outcome measure in terms of both quantity and quality of life years (32). CUA is often the recommended form of economic evaluation for disease conditions when quality of life (QoL) is an important outcome measure (81). Many studies have shown that oral diseases have considerable impact on day-to-day activities and QoL, especially among children and adolescents (13-15). Hence, CUA is an important type of economic evaluation to evaluate oral health care interventions, in which generally QoL is improved as a result of an intervention (81).

7.2 Background analysis

7.2.1 CUA in oral health research

The literature review (Chapter 2a) (147) identified increasing use of CUA in oral health research over time. This review identified 23 CUA applications in oral health research from 2000 to 2016. Tonmukayakul et al, 2015 reported that there were only eight CUAs published in dentistry from 1975 to 2013 (22). Of them, seven were published after 2000. Similarly, Chapter 2a noted that around 60% (14) of the CUAs were published after 2010. The majority were able to identify the cost-effective interventions among those being compared and were of good reporting quality, which will assist resource allocation and health care decision-making. This is a promising trend, as CUA is the recommended method of health technology assessment, especially in developed countries.

As the aim of this review was to obtain an overview of the use of CUA in oral health research, it was not confined to studies conducted among paediatric populations.

However, of the 23 studies identified, only four during 2000–2016 involved paediatric samples. Several recent systematic reviews of economic evaluations of oral health interventions among paediatric populations (36, 46, 214) revealed that there have since been no new CUAs in oral health research. While the number of CUAs among paediatric populations and their reporting quality increased in other areas of health (215), this trend was not observed in oral health CUAs among children and adolescents.

The quality adjusted life year (QALY) is the most common outcome measure in CUA, as was confirmed in this review (44). However, a special feature identified here is the use of different derivatives of quality-adjusted outcomes that are specific to dentistry, such as quality-adjusted tooth years (QATYs) and quality-adjusted prosthesis years (QAPYs), or other summary outcome measures used in CUA such as disability-adjusted life years (DALYs). Of the four CUAs identified among paediatric populations, three used QATYs (48) or DALYs (51, 96) as the outcome measure. The fourth study used a generic PBM to calculate QALYs (97). The main advantage of QALY is that it allows comparison of cost-effectiveness across different disease areas and various procedures. This information is valuable for resource allocation and health care planning (32). Therefore, the use of other outcome measures may affect the main strength of the CUA when it is necessary to compare oral health interventions across different health areas to allocate limited resources.

7.2.2 Paediatric quality of life instruments in oral health research

In CUA, outcomes are measured in terms of the quantity and quality of life. Therefore, assessment of available QoL instruments is important to evaluate the application of CUA in oral health research. In particular PBMs report single summary scores for health-related QoL and are used to derive QALYs for economic evaluations (216). Therefore, the literature review of paediatric quality of life (QoL) instruments in oral health research (Chapter 2b) was conducted to provide an overview of paediatric quality of life instruments used in oral health research (148). This review identified 11 paediatric oral health-specific QoL instruments and five generic paediatric QoL instruments used in oral health research. The evidence has shown a recent marked increase in paediatric QoL instruments, particularly disease-specific QoL instruments in all areas of health (112). Similarly, the present review also identified a wide range of paediatric oral health-specific and generic QoL instruments used in oral health research and except for one, all were published after the year 2000. Further, there was remarkable

growth of oral health-related quality of life (OHRQoL) research in the paediatric and orthodontics fields compared to other areas of oral health (11). This was likely a facilitating factor in the development and validation of a considerable number of paediatric QoL instruments in oral health, as evident in Chapter 2b. Moreover, due to the availability of an extensive range of paediatric OHRQoL instruments, researchers are able to use the QoL instrument best suited to their research.

However, one of the main factors identified in this review is that there is no paediatric PBM available for oral health. Out of the 11 oral health-specific QoL instruments identified, none was preference-based. PBMs are used to calculate QALY to assess health care intervention using the cost utility analysis framework (53). As no condition-specific paediatric PBM was available in oral health, studies used direct health state valuation or generic PBMs to estimate QALY in oral health interventions, as evident in Chapter 2a (147). CHU9D and EQ-5D-Y are the two generic PBMs used in paediatric oral health research. However, the performance of these two instruments to capture the changes in oral health-related QoL has still not been adequately evaluated (139, 140).

Public oral health care provision in many countries mainly focuses on the prevention and treatment of oral diseases among children and adolescents (108). Childhood oral diseases are highly preventable in nature and the identification of oral health care interventions that provide the best value for money will reduce the associated economic burden to the health care system (19). However, the background analysis identified only a limited number of CUAs among paediatric oral health interventions. Further, there is no condition-specific preference-based measure (CSPBM) available for oral health in paediatric populations. The limited number of CUAs in this area is likely due to the fact that there is no oral health-specific PBM for use in economic evaluations of oral health care interventions. Recent systematic reviews also identified the need for a paediatric utility measure to quantify outcomes in terms of QALYs and to promote the economic evaluation of oral health care interventions using CUA among children and adolescents (36, 46). Thus, the main aim of this thesis was to meet this need by developing an oral health-specific preference-based quality of life measure to facilitate the identification of high-value oral health interventions in adolescent populations.

7.3 Development of a preference-based quality of life measure

A preference-based quality of life measure generally applicable for all oral diseases would have been ideal to facilitate economic evaluations of oral health

interventions among children. However, common oral diseases among children, such as dental caries, gingivitis, oral trauma and malocclusion, are varied in their prevalence, symptoms and impact on the quality of life. For example, in dental caries pain and discomfort would be more important whereas in gingivitis, bleeding gums and in malocclusion, appearance/ social interactions would be more important. Therefore, considering the time and resource constraints to develop a PBM to cover all oral diseases, efforts were focused on developing a PBM specific for one childhood oral disease.

Dental caries is the most prevalent childhood oral disease in Australia (6) and the world (2). Around 38% of Australian children aged 12–14 years have experienced dental caries in their permanent teeth (6). Due to high prevalence and preventable in nature, the majority of economic evaluations of oral health research among children and adolescents mainly focus on dental caries interventions (36). Therefore, dental caries was considered as the main oral disease condition to develop the oral health-specific PBM. Further, target group for the new instrument was considered as the children above 12 years of age considering that the mixed dentition period is over by the age of 12 years and the less cognitive ability of younger children to understand the concepts and wordings of the PBMs. A preference-based quality of life measure consists of two components: 1) a classification system to define health states and 2) a set of utility weights to generate utility values for the corresponding health state (60, 61).

7.3.1 Development of a classification system for a preference-based quality of life measure for dental caries

A classification system for a PBM can be developed by either converting an existing non-PBM or creating an entirely new measure (62). The literature review of paediatric QoL instruments in oral health research identified 11 non-PBM oral health QoL instruments (148). All of these adopt frequency-type response options rather than severity levels and some contain negatively worded items. A qualitative study conducted to identify adolescents' opinions about dental caries reported that they generally describe caries symptoms in terms of severity rather than frequency (151).Thus, the authors of this study suggested that the response format use severity levels instead of frequency in dental caries-specific QoL measures (151). However, the OHRQoL instruments identified in the present study cannot be easily transformed to use different types of response levels. Moreover, although three of these 11 instruments are PBM QoL measures specific to dental caries (115, 124, 125), all three are targeted for early childhood caries in 3-5-year-

old children. Therefore, a classification system for the new dental caries preference-based measure was developed as a *de novo* measure based upon the above literature review, qualitative interviews with target group adolescents and expert validation (62, 150). These three strategies are commonly used in the development of both new CSPBMs (62) and generic PBMs for children and adolescents (53, 150). The detailed analysis of OHRQoL instruments further revealed that the majority of these instruments were developed using item pools derived from focus group discussions with children, adolescents, parents and health professionals (114, 121). Further, during the development process, the draft classification system was further refined with a sample of adolescents who had dental caries and also with the group of experts in dental public health and clinical setting. These steps ensured the necessary face validity and content validity of the instrument. The new classification system was named Dental Caries Utility Index (DCUI). It includes five items: 'pain/discomfort', 'difficulty in eating foods/drinking', 'worried', 'ability to participate in activities' and 'appearance' and each item has four response levels (see Chapter 3, Figure 3.4).

Oral health, functional, emotional and social (self-image and school environment) are the common dimensions in OHRQoL instruments (11, 217). The new classification system included items to represent these common dimensions (oral health—pain/discomfort; functional—difficulty eating foods/drinking; emotional—worried; social—able to participate in activities and appearance) to cover every aspect of OHRQoL. Further, pain/discomfort, worried, appearance and school activities are also common items appearing in generic paediatric PBM, although the response levels are differently worded to suit health-related QoL more generally (53).

7.3.2 Valuation of a preference-based quality of life measure for dental caries (Dental Caries Utility Index - DCUI)

Study sample and mode of administration

A scoring algorithm allows DCUI to be used in economic evaluations of dental caries interventions and was derived using a health state valuation study. The DCUI classification system has been developed for children above 12 years of age. There is a continuous debate among researchers regarding who should value the health states derived from a classification system, particularly those for children. Understanding the health state valuation tasks and providing a logical value for different health states

imposes a high cognitive demand on the participants, especially the valuation tasks using time-trade-off (TTO) and standard gamble (SG) methods. Further, these tasks require participants to trade life years, risk or immediate death. Therefore, conducting a health state valuation study among paediatric samples would be methodologically challenging (46). Existing paediatric generic PBMs have valued health states from children and adolescents themselves or from adult general population members or based on parents/guardian proxy responses (47). Rowen et al reported wide variation in the approaches used to value existing generic paediatric preference-based measures in relation to the study sample, valuation technique, anchoring method and perspective (47). Chen et al reviewed nine existing paediatric generic preference-based measures and reported that the majority were valued by adult samples (17). Considering the methodological constraints of using a sample of children, the present study valued DCUI health states using a sample of adult general population members. Further, as tax payers and parents, adult general population members spend considerable funds on expensive dental treatments; thus, their preferences are important in the valuation of interventions.

The valuation survey was conducted online. Online data collection is the most common mode of administration of discrete choice experiment surveys (32), as it substantially reduces the time and resources required when compared to face-to-face interviews and postal or telephone methods. However, the online sample in the main survey was significantly different from the Australian general population in terms of their education level, gross weekly household income and marital status. Previous online studies also reported over- or under-representation from the Australian general population (48, 49).

Preference elicitation technique

The health state valuation study was conducted using the DCE method, which has been popular due to its easy administration in online platforms (65). However, DCE is an ordinal preference elicitation technique that produces utility values on a latent scale. Therefore, the utility values obtained must be anchored on a full health-dead scale prior to QALY calculation (66). Several methods have been used in the literature (64, 66); however, dental caries is not a seriously disabling condition. Therefore, most of these methods may not be appropriate for dental caries, as they are based on trading-off life years or immediate death. Further, previous DCE studies in oral health focused on eliciting preferences for dental treatment (74) or the use of dental health services or

programmes (75). Therefore, evidence for the most suitable anchoring approach for a condition like dental caries was scarce. Two possible approaches—discrete choice experiment with duration (DCE_{TTO}) and discrete choice experiment with visual analogue scale (DCE_{VAS})—were compared to identify the most suitable approach for anchoring.

Pilot data revealed that both the unadjusted and adjusted models in the DCE_{VAS} approach produced ordered coefficients when compared to the DCE_{TTO} approach. The more ordered coefficients in DCE_{VAS} could be due to the reason that they were from a main effects DCE and did not include interaction with duration, as in DCE_{TTO}. Further, DCE_{TTO} produced more disperse utility decrements and severe health states were valued worse than death, which is unreliable for a condition like dental caries. A similar observation was reported by Fyffe et al (71), who compared dental free time trade-off (DTTO) and dental visual analogue scale (DVAS) to value 12 dental health states and found that the utility values resultant from the DVAS were more reliable and in order (e.g., tooth states with pain had lower utilities than those without pain). Further Fyffe et al (71) reported that the utility values from DTTO were highly skewed and indicated that the participants were unwilling to trade their free time to achieve better tooth health states.

Based on the pilot data, the main health state valuation study was conducted using the DCE_{VAS} approach. Conditional logit was used to model DCE data and the estimated coefficients for all dimensions were generally monotonic (i.e., in the expected direction and order in which higher severity levels had greater utility decrements). All coefficients were statistically significant, except for those of the second levels of the 'worried' and 'appearance' domains. The utility values obtained from both rescaling with the worst health state and mapping showed largely similar values. The final utility algorithm was completed based on the mapping approach, as the mean absolute error (MAE) value showed that the DCE estimates based on the mapping approach performed better (i.e., MAEs for the mapping approach were lower than those of anchoring based on worst health state). The 'pain/discomfort' dimension was associated with the highest utility decrements.

As there are no previous studies that value health states derived from a classification system in dentistry, it was not possible to directly compare the utility values obtained from this study with previous research. There are three other multi-attribute utility scales in oral health: the multi-attribute utility scale for periodontal disease (208), the multi-attribute utility scale for head and neck cancer (218) and the Minor Oral Surgery Outcome Scale (219). However, the dimensions in these instruments are not related to

dental caries and were not designed to generate QALY weights (220). Therefore, the utility values obtained from the present study could not be compared to these scales. Nonetheless, previous direct health state valuation studies in oral health have shown that pain is an important factor influencing dental health state utility values (71, 209) and a similar observation was found here.

There are often discussions among researchers regarding the suitability of using VAS in health state valuation and cost-utility analysis. VAS is often criticised because it does not involve any trade-off of life years or risk and is associated with certain biases (169). Parkin and Devlin (221) critically analysed the theoretical and empirical evidence and suggested that VAS has many favourable features as a health state valuation technique when compared to the other methods. VAS is comparatively simple, easy to use and reliable (169). It has been applied in previous studies to value health states defined by classification systems (168) and in oral health studies, VAS was used as a direct health state valuation method to value dental health states utilities (71, 213). However, it had not been used to anchor the latent utility values from DCE onto a full health-dead scale until a recent valuation study for EQ-5D health states reported the first use of VAS for this purpose (192). The authors suggested that the VAS approach is a feasible anchoring method and is more suitable for a disease condition in which participants may be reluctant to trade-off life years to achieve better health states (192). However, they used anchoring with VAS for the worst health state approach only (192). The present thesis also supports VAS as a feasible and reliable approach to anchor the utility values obtained from DCE. Further, this study adds that in addition to the rescaling with the VAS worst health state, mapping VAS onto DCE is also another possible method to rescale DCE coefficients onto a full health-dead scale.

7.4 Policy implications

This is the first study to develop a health classification system and a utility algorithm for a preference-based quality of life measure for dental caries. Dental caries is the most prevalent childhood oral disease and is highly preventable in nature. It is likely that the majority of the economic evaluations among paediatric population would focus on dental caries. Therefore, the availability of this new CSPBM for dental caries will facilitate better assessment of the impact of oral health care interventions through CUA and may improve the decision-making process and have evidence-based policy implications.

The Australian Government's total expenditure on dental services was AU$ 2.35 billion in 2016–2017 (18) and dental treatments of eligible children (2–17 years) are supported through public dental health schemes. Moreover, 58% of the total expenditure on dental services (AU$ 5.8 billion) was paid directly by individuals as out-of-pocket costs and 18.7% came from health insurance funds (18). Although government expenditure on dental services is comparatively lower than the private expenditure, the new CSPBM for dental caries would facilitate the identification of the prevention and treatment options for dental caries that offer the best value for money. Hence, DCUI would support the decision-making process to allocate resources effectively within the limited budget. Further, the new CSPBM for dental caries would be used to identify the best preventive and treatment strategies for dental caries through CUA from a societal perspective. Hence, DCUI will facilitate policy decisions that will have positive impact on reducing the direct out-of-pocket expenses of individuals and families.

Sugar consumption is the main risk factor for dental caries (8). A recent report revealed that 8% of 12–13-years old and 12.4% of 13–17-years old Australian children consume sweetened beverages and/or confectionary regularly (30). Therefore, to achieve the dental caries reduction targets set out in the National Oral Health Plan 2015-2024, Australia, there should have been more favourable policy decisions and preventive interventions among children to change their behaviour. In such instances, PBM for dental caries would be supportive for the policy decisions to identify the interventions with the best value for money. Further, sugar consumption is a risk factor for other health conditions such as overweight and diabetes (9). In order to achieve a healthy population, adopting positive behaviour strategies during childhood would be extremely beneficial. For such instances, health policies and population-based health programmes through broad-based government actions with multi-sectoral approaches, such as education and taxing of sugary beverages, are essential to improve overall health (222). The impact of a preventive programme addressing common risk factors among the population would be easily measured for a condition like dental caries, since the progression of dental caries is rapid and visible compared to conditions with slower onset, such as diabetes. The effectiveness of such measures on dental caries could be proven using the new CSPBM for dental caries.

There has been significant attention to improving proper access to oral health care. In 2017–18, more than 72,000 potentially preventable hospitalisations were reported for dental conditions that could otherwise have been prevented with earlier treatment (223).

Among them, more than 25,000 potentially preventable hospitalisations were related to children 0–14 years of age (223). Further, 18% of adults aged 15 years or over avoided or delayed seeking dental care due to its cost (29). Only around 23% of adults aged 18 and over accessed public dental care in 2016–2018.

Policymakers and political leaders expressed interest in improving access to oral health care, such improved access to oral health care is of the utmost importance to achieve the oral health targets set for 2025. For example, Medicare is reviewing items to cover some surgical procedures performed by approved dentists and procedures under the Cleft Lip and Palate Scheme (27). The new CSPBM for dental caries developed in this study has the potential to be developed as an adult measure as well as for assessing other common oral diseases. Thus, future research on adopting the DCUI as an adult measure or for other oral diseases would facilitate the identification of prevention and treatment options offering best value for money and would reorient health services to improve access to dental care services.

7.5 Strengths and Limitations

Strengths

This thesis is the first study to provide comprehensive overviews of the CUAs conducted in oral health interventions (and their reporting quality) and the preference-based and non-preference-based paediatric QoL measures used in oral health research. This is the first study to develop a preference-based QoL measure (a classification system and utility algorithm) to be used in economic evaluations of the most prevalent childhood oral disease among Australian adolescents, dental caries.

This study assessed the two approaches (DCE_{TTO} and DCE_{VAS}) that can be used to anchor the utility values obtained via the DCE method onto full health-dead scale in dental caries health state valuations and showed that VAS is a feasible and reliable anchoring approach for this purpose. This finding may guide research related to anchoring DCE utility values, especially for disease conditions in which participants may be reluctant to trade life years or immediate death to achieve better QoL.

In the DCE_{VAS} approach, this thesis reported two methods to rescale DCE coefficients onto a full health-dead scale: using worst health state in VAS and mapping DCE onto VAS. These methods were adopted from the TTO-based approaches proposed by Rowen et al (66). A recent study reported only rescaling with worst health state in the

DCEVAS approach (192) and therefore, this thesis contributes an additional method for rescaling in DCEVAS.

Limitations

This study also has certain limitations. The draft classification system for DCUI was developed based on the domains and items identified from non-PBM OHRQoL instruments. Therefore, limitations associated with the identification of relevant items for those instruments also affect the DCUI classification system. However, this DCUI classification system was developed using information from all relevant existing instruments rather than a single measure and items were included from the most common dimensions of all OHRQoL instruments. In addition, the qualitative studies and expert opinion supported the proposal that the most important items have been included. Hence, the development process of the DCUI classification system ensured that it covers all aspects of OHRQoL while making the maximum effort to mitigate the impact of limitations associated with the identification of relevant items from non-PBM OHRQoL instruments.

A major limitation is that the DCUI is developed to be used among a relatively small population group, Australian adolescents. Due to the feasibility and resource limitations, the development of the DCUI process did not involve adolescents from different countries. The fact that qualitative interviews were conducted only with adolescents in a certain geographic area in Australia is another potential limitation. However, the geographic variation itself is unlikely to have a significant impact on the identification of the most relevant items for dental caries. The use of qualitative interviews with adolescents and expert opinion at the development stage ensured the face validity and content validity of the instrument. Further, the psychometric properties were not rigorously evaluated with clinical samples as well as with the target group adolescents. This is a major limitation in this study. Although this work was planned, it was not executed due to time and resource restrictions. Instead, a comparison study was performed to identify the most suitable anchoring approach to evaluate dental caries health states—which was identified as a key gap to be filled in literature. A future study is also planned to analyse the performance of the DCUI among adult participants of the valuation study using the available oral health data such as OHIP-14 score and self-

reported oral health status to provide validity on the implicit value of oral health among respondents.

The health state valuation study was conducted as an online survey among a sample of the adult Australian general population. Although the sample was age and sex representative, their other socio-demographic characteristics were significantly different from the Australian general population in relation to education level, income and marital status. Over- or under-representation of some socio-demographic categories is a common limitation in other online surveys (69, 224). However, it was not possible to evaluate the extent to which these differences affect the sample's health state preferences. Further, in the pilot studies and the valuation study, there were participants who did not complete the whole survey and others who provided logically inconsistent answers to the valuation tasks. The underlying reasons for these responses could not be established, as the study was conducted as an online survey. However, by using sensitivity analysis and other methods during the data analysis, the maximum effort was taken to address the impact of these logically inconsistent answers on the final outcome.

7.6 Recommendations for future research

The DCUI was developed for use among Australian adolescents. Rigorous validation of this classification system among child populations, especially in a clinical setting, is recommended to evaluate its psychometric properties.

Although the DCUI is to be used among Australian adolescents, there is high potential that this instrument could be used in CUAs of dental caries economic evaluations in other countries as well. Therefore, cross-cultural validation studies are highly recommended to adapt the new classification system for PBM to be used in other countries. The classification system mainly focused on dental caries due to its prevalence. However, further studies could evaluate its use more broadly for other oral health conditions, considering that the included dimensions are common in most other oral diseases as well.

The DCUI was developed as a paediatric measure since the public health system is mainly focused on providing oral health care services to children and adolescents in many countries. However, the DCUI has the potential to be adapted as an adult measure because dental caries is one of the most common oral diseases in adulthood as well.

Therefore, future studies could be focused on adopting the DCUI to be used in CUAs of adult oral health interventions.

Due to the methodological constraints, the utility algorithm for the DCUI was developed based on preference elicitation among an adult sample of the Australian general population. Although it is relatively common to conduct health state valuation studies for paediatric PBMs using adult general population samples, there is a continuous debate among researchers over 'whose preferences should be valued' for paediatric PBMs. Therefore, future research is needed to assess whether the health state utility values for DCUI derived from an adult sample are significantly different from the preferences of an adolescent sample.

This study used the VAS approach to anchor health state utility values derived from DCE and the VAS anchoring task included six health states to be valued on a single VAS scale. Due to the limitations in technological capacity, it was not possible to randomise the order of these states or to assess the effect of the health state values given by the participants as a result of ordering. Therefore, future research is recommended to evaluate the use of the VAS approach as an anchoring method in other disease areas and to consider whether the order of health states in a VAS task affects the assigned score.

Cross-program comparability is an important factor in funding decisions. It has been proposed that using the same generic PBM for all studies is the main strategy to achieve cross-program comparability (62). However, the generic PBMs may not be sensitive to changes in some conditions or not perform well in relation to validity. For example the CHU9D was unable to detect a significant differences in the changes to dental caries status (140). In such conditions, CSPBMs have an important role in the economic evaluation of health interventions. However, the use of CSPBMs in economic evaluations may compromise cross-program comparability to inform funding allocations (62). Thus, it is important to compare CSPBMs with the available generic PBMs in terms of their validity and responsiveness to assess the trade-off between sensitivity and comparability while using the CSPBM (62). Therefore, future research is recommended to compare the DCUI with the available generic PBMs. Further, CSPBMs may not include the effect of comorbidities and side effects of treatment, which may pose a potentially important threat to both comparability and sensitivity (62). Dental caries shares common risk factors with other diseases and the prevention and management of dental caries might have an impact on quality of life changes in other domains that may not be fully captured in the DCUI domains. For example, changes in diet may also have

an effect on physical and cognitive functions. Therefore, future quantitative and qualitative research studies are recommended to assess the effect of incorporating known side effects and highly prevalent comorbidities as add-on dimensions to the DCUI.

7.7 Conclusions

This thesis is the first study to provide comprehensive overviews of the CUAs conducted in oral health interventions (and their reporting quality) and the preference-based and non-preference-based paediatric QoL measures used in oral health research. The systematic review of CUAs in oral health interventions (Chapter 2.1) reported an increasing trend of using CUAs in economic evaluation of oral health interventions. The majority of the CUAs identified in this systematic review were able to provide conclusions regarding the most cost-effective intervention among the different options compared and were of good reporting quality. Hence, these CUAs could assist in health care decision-making and resource allocation. However, this review identified only a limited number of CUAs conducted among paediatric populations, indicating that CUA is not a frequent method for evaluating oral health interventions among children and adolescents.

The systematic review of paediatric QoL measures used in oral health research (Chapter 2.2) identified a wide range of paediatric QoL instruments used in oral health research among children and adolescents. The availability of these QoL instruments will provide the opportunity to use the best-suited QoL instrument for the research question being addressed and the target age group (i.e., pre-school, school age, etc.) in paediatric oral health research. However, this review reported that there is no oral health-specific paediatric PBM available to be used in economic evaluations of oral health interventions using a CUA framework. The limited number of CUAs identified among paediatric oral health research is likely due to the fact that there is no oral health-specific paediatric PBM. This thesis aimed to fill this gap by developing an oral health-specific paediatric PBM that can be used in the economic evaluation of oral health care interventions.

This is the first study to develop a preference-based quality of life measure for dental caries (a classification system and utility algorithm) to be used among Australian adolescents. The classification system consists of five items and each item has a four-level response scale. The Australian-specific tariff of DCUI ranges from 0.1681 to 1.0000. This will facilitate utility value calculations from participants' responses for

DCUI in economic evaluations of dental caries, the most prevalent childhood disease among Australian adolescents. Hence, it will improve CUA analysis of dental caries interventions and the allocation of resources to effectively to treat and prevent dental caries.

This study assessed the two approaches (DCE_{TTO} and DCE_{VAS}) that can be used to anchor the utility values obtained via the DCE method onto full health-dead scale in dental caries health state valuations and showed that VAS is a feasible and reliable anchoring approach for this purpose. Further, in the DCE_{VAS} approach, this thesis reported two methods to rescale DCE coefficients onto a full health-dead scale: using worst health state in VAS and mapping DCE onto VAS. These findings may guide research related to anchoring DCE utility values, especially for disease conditions in which participants may be reluctant to trade life years to achieve better QoL.

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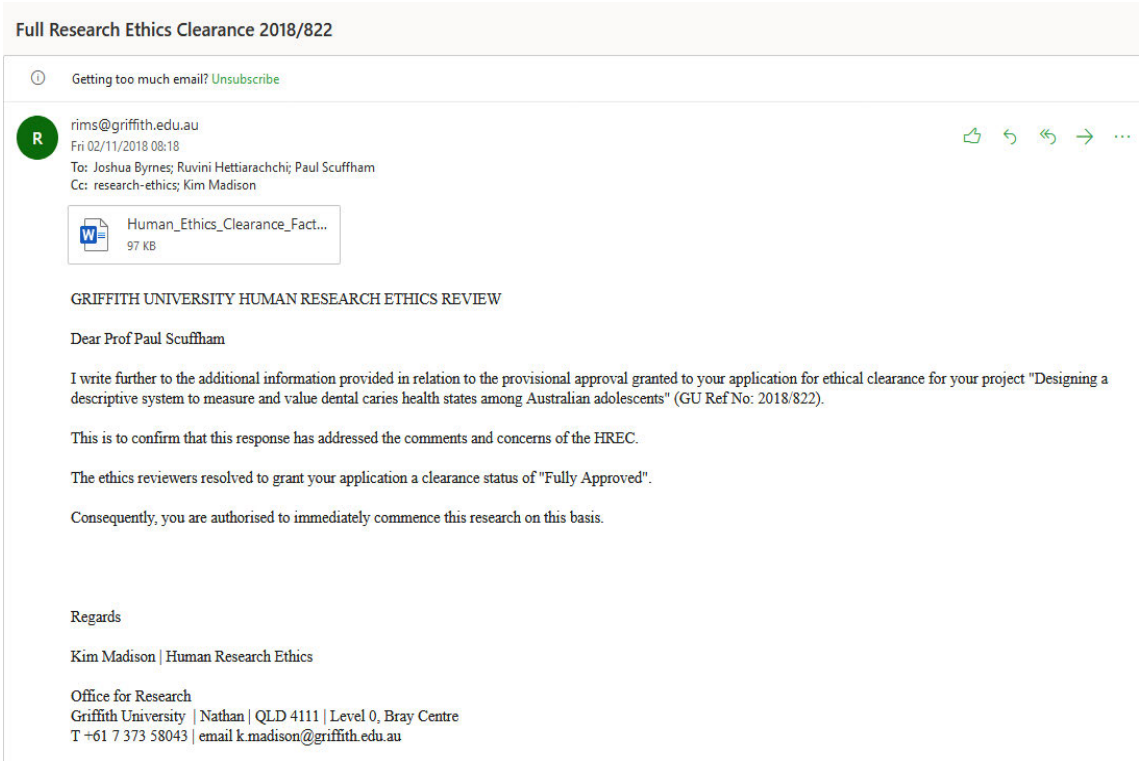
Appendices

Appendix A: Griffith ethical clearance- Development of the classification system for the Dental Caries Utility Index (DCUI)



Full Research Ethics Clearance 2018/822

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R

rims@griffith.edu.au
Fri 02/11/2018 08:18
To: Joshua Byrnes; Ruvini Hettiarachchi; Paul Scuffham
Cc: research-ethics; Kim Madison

Human_Ethics_Clearance_Fact...
97 KB

GRIFFITH UNIVERSITY HUMAN RESEARCH ETHICS REVIEW

Dear Prof Paul Scuffham

I write further to the additional information provided in relation to the provisional approval granted to your application for ethical clearance for your project "Designing a descriptive system to measure and value dental caries health states among Australian adolescents" (GU Ref No: 2018/822).

This is to confirm that this response has addressed the comments and concerns of the HREC.

The ethics reviewers resolved to grant your application a clearance status of "Fully Approved".

Consequently, you are authorised to immediately commence this research on this basis.

Regards

Kim Madison | Human Research Ethics

Office for Research
Griffith University | Nathan | QLD 4111 | Level 0, Bray Centre
T +61 7 373 58043 | email k.madison@griffith.edu.au

Appendix B: Summary of the adolescents' opinions- Development of the classification system for the DCUI

Appendices-Table 1: Summary of adolescents' opinion

	Items	Number of adolescents identified included items and levels as an important factor in tooth decay*	Number of adolescents stated that the wordings are not clear to them
1	Pain/Discomfort	15	2
2	Difficulty eating foods/drinking	14	1
3	Worried/anxious about tooth	12	3
4	Able to join in activities (Playing, Sports, School activities)	9	2
5	Location of tooth cavity	11	2
6	Appearance of my decayed/filled/missing tooth	14	5
7	Swelling	11	3

Summary of important opinions/views of adolescents

1. **What do you think is meant by 'tooth decay'?**

- *When a tooth is decaying this means a tooth is filing up with bacteria and tooth is rotting away. (Participant 1, Male, aged 14 years)*
- *A tooth that is not in good health, broken down because of build-up of germs in one's mouth. (Participant 2, Male, aged 15 years)*
- *I think a decayed tooth is a rotten tooth. (Participant 3, Female, aged 12 years)*
- *I think it is a tooth not healthy and should be taken out. (Participant 4, Female, aged 14 years)*
- *I think a tooth decay is when your tooth has a cavity and it starts hurting. (Participant 5, Female, aged 12 years)*
- *A decayed tooth is a tooth dirty on the sides. (Participant 6, Female, aged 12 years)*
- *A tooth that is rotten and damaged. It would cause a lot of toothache and pain to a person. (Participant 7, Male, aged 17 years)*
- *Something not healthy (Participant 8, Male, aged 13 years)*
- *A tooth starts to decay and fall apart, you see black thing. (Participant 10, Male, aged 12 years)*
- *When your teeth change colour and when it starts to hurt. (Participant 11, Female, aged 14 years)*
- *I don't know decayed tooth. (Participant 12, Male, aged 13 years)*
- *I can't understand what 'decay' means. A bad tooth? (Participant 13, Female, aged 12 years)*
- *When bacteria grows on it. (Participant 14, Female, aged 14 years)*

- *Tooth that is not healthy and should be taken out. (Participant 15, Male, aged 15 years)*

What are the main problems associated with a decayed tooth?

- *Not brushing teeth, not having daily check-ups (Participant 1, Male, aged 14 years)*
- *It could lead to infections and problems for other teeth and possibly on other areas of the body ((Participant 2, Male, aged 15 years)*
- *You have to take teeth out or add filling. (Participant 3, Female, aged 12 years)*
- *Toothache, infection, gum problems (Participant 4, Female, aged 14 years)*
- *Cavities and sugar stuck in teeth (Participant 5, Female, aged 12 years)*
- *I don't know (Participant 6, Female, aged 12 years)*
- *Pain, appearance (Participant 7, Male, aged 17 years)*
- *Have to go to dentist, pain (Participant 8, Male, aged 13 years)*
- *Toothache, weak gums. (Participant 9, Male, aged 14 years)*
- *No idea. (Participant 10, Male, aged 12 years)*
- *Hurt gums, hurt when biting. (Participant 11, Female, aged 14 years)*
- *Pain. (Participant 12, Male, aged 13 years)*
- *Eating too much candy and drinking soft drinks. (Participant 11, Female, aged 14 years)*
- *Hurts when biting (Participant 13, Female, aged 12 years)*
- *Pain. (Participant 14, Female, aged 14 years)*
- *Toothache. (Participant 15, Male, aged 15 years)*

2. **Tooth location**

- *Front teeth-the ones we can see and back teeth- the one we can eat from. (Participant 1, Male, aged 14 years)*
- *Because the dentist need to know where the tooth is if they want to take it out. (Participant 3, Female, aged 12 years)*
- *If it is one of my front teeth, it will affect my smile. (Participant 5, Female, aged 12 years)*
- *Front teeth people usually see (Participant 8, Male, aged 13 years)*
- *No, depending on the person. (Participant 9, Male, aged 14 years)*
- *Because a rotten tooth look bad. (Participant 11, Female, aged 14 years)*
- *Back teeth most easy to get bad because we can't check them often. (Participant 12, Male, aged 13 years)*
- *Front teeth are used for biting back teeth are used for chewing. (Participant 14, Female, aged 14 years)*

3. **Pain/discomfort**

- *If the tooth is causing discomfort or pain to me, I know to go to a dentist and describe my pain (Participant 4, Female, aged 14 years)*

- *Because tooth decay does start to get a bit uncomfortable. (Participant 3, Female, aged 12 years)*
- *Yes, because it could show how bad the tooth is. (Participant 5, Female, aged 12 years)*
- *Because pain can distract you. (Participant 11, Female, aged 14 years)*

4. **Difficult eating/drinking**

- *It is important because it shows how bad the tooth is (Participant 5, Female, aged 12 years)*
- *It hurts when eating (Participant 9, Male, aged 14 years)*
- *Eating food could be difficult if the rotten tooth is at the back of your mouth. (Participant 7, Male, aged 17 years)*

5. **Worried or anxious about decayed tooth**

- *Depends if it is a baby tooth or not. (Participant 1, Male, aged 14 years)*
- *Yes depends on whether it is a baby tooth or not. (Participant 2, Male, aged 15 years)*
- *I will be worried but I wouldn't be anxious. (Participant 3, Female, aged 12 years)*
- *I wouldn't be considered too much. (Participant 4, Female, aged 14 years)*
- *No, because that could just depend on your personality. (Participant 5, Female, aged 12 years)*
- *If it is not a baby tooth I will be worried. (Participant 6, Female, aged 12 years)*
- *I am not too worried (Participant 8, Male, aged 13 years)*

6. **Able to join in activities (Playing, Sports, School activities)**

- *Yes, the pain will interrupt concentration. (Participant 2, Male, aged 15 years)*
- *No, because your running you aren't using your teeth unless if your tooth hurts a lot. (Participant 3, Female, aged 12 years)*
- *No because unless it is causing severe pain, I will not be affected while playing sports. (Participant 4, Female, aged 14 years)*
- *Yes, because it hurts when you move around (Participant 5, Female, aged 12 years)*
- *Not a problem unless your tooth hurts a lot. (Participant 6, Female, aged 12 years)*
- *No, because tooth doesn't affect sporting abilities. (Participant 7, Male, aged 17 years)*
- *Yes Pretty hard to concentrate (Participant 8, Male, aged 13 years)*
- *Pain, appearance all affect joining in activities. (Participant 9, Male, aged 14 years)*
- *Can't eat with decay. (Participant 11, Female, aged 14 years)*
- *No, most activities don't include teeth. (Participant 13, Female, aged 12 years)*
- *If you are in pain, you can't do your best. (Participant 14, Female, aged 14 years)*
- *Because pain can distract you. (Participant 15, Male, aged 15 years)*

7. **Appearance**

- *Having decayed tooth would make me very ugly appearance for everything. (Participant 1, Male, aged 14 years)*
- *Yes because your tooth will look rotten and it will look bad (Participant 3, Female, aged 12 years)*
- *I think if decaying is in obvious spot then it has bad appearance. (Participant 4, Female, aged 14 years)*
- *Yes because your teeth could be too out which means you need braces. (Participant 5, Female, aged 12 years)*
- *It wouldn't affect you. (Participant 10, Male, aged 12 years)*
- *Yes I don't want people to see my decayed tooth. (Participant 11, Female, aged 14 years)*
- *Yes, it could be really bad. (Participant 14, Female, aged 14 years)*
- *It makes you look unhealthy (Participant 15, Male, aged 15 years)*

8. **Swelling**

- *Yes, it can be. (Participant 1, Male, aged 14 years)*
- *Yes, because it can show how bad the tooth is. (Participant 5, Female, aged 12 years)*
- *If it is a big swelling, it will be ugly and painful. (Participant 11, Female, aged 14 years)*

9. **Wordings of the instrument**

- *Yes, no problem. I can understand (Participant 1, Male, aged 14 years)*
- *May be. I am not sure tooth decay. But I know it now because you said it is a rotten tooth (Participant 5, Female, aged 12 years)*
- *Yes ok for me (Participant 8, Male, aged 13 years)*

10. **Suitability to the age group and completeness**

- *I can't remember anything more (Participant 4, Female, aged 14 years)*
- *Only for big kids. Not for small ones (Participant 7, Male, aged 17 years)*
- *I think so (Participant 11, Female, aged 14 years)*

Appendix C: Summary of the expert opinion - Development of the classification system for the DCUI

Appendices-Table 2: Expert panel characteristics

Expert	Highest level of education in Dentistry	Age	current position in Dentistry/ Oral Health field	Number of years' experience in Dentistry	Number of years' experience in current position	Duties/ responsibilities in current position related to paediatric oral diseases
1	Doctorate	56	Associate Professor: Teaching and Research	>30	>3	Conduct paediatric oral health research Plan and implement oral health promotion programmes
2	Doctorate	38	Senior Research Fellow	14	1	Conduct paediatric oral health research Plan and implement oral health promotion programmes
3	Doctorate	43	Lecturer/ Clinical educator	15	2	Work in paediatric dental clinic
4	Degree	55	Lecturer/ Practitioner	30	Lecturer (3) Private practice (30)	Work as a general dental practitioner and treat children and adolescents
5	Masters Degree	44	General Dentist	18	10	Work as a general dental practitioner and treat children and adolescents
6	Degree	64	Clinical Dental Educator	40	4	Work in paediatric dental clinic

Appendices-Table 3: Summary of the expert opinion-1st round

	Items and levels	Number of experts agreed on the included items and levels*	Number of experts raised an issue	Number of experts suggested wordings change	Comments
1	**Pain/Discomfort** 1. I have no pain or discomfort 2. I have a little pain or discomfort 3. I have quite a lot of pain or discomfort 4. I have a lots of pain or discomfort	6	1[#]	1	• 'I am unsure about combining both discomfort and pain into one attribute. Pain could have a severe impact on QoL while discomfort might not. Further, you might need to reiterate for each attribute that the questions are related to dental caries/tooth cavity and not due to other reasons. For eg. Gingival bleeding or tooth crowding could also contribute to discomfort' (Expert 2)
2	**Difficulty eating foods/drinking** 1. I have no difficulty in eating foods/drinking 2. I have a little difficulty in eating foods/drinking 3. I have quite a lot of difficulty in eating foods/drinking 4. I have lots of difficulty in eating foods/drinking	6	0	0	• 'Are you asking about a cavity, and/or filled or missing tooth?' (Expert 1) • 'As commented earlier, difficulty in eating/drinking foods might occur due to several reasons. You might have to clarify 'Difficulty in eating/drinking foods due to tooth cavity' (Expert 2) • Is it important to determine why they are having difficulty in eating/drinking, in this question? Maybe they have difficulty due to pain? Or no pain, but difficulty due to the space resulting from a missing tooth? Or due to another reason, etc). Or, will that information be assumed based upon their answer to the above pain question? (Expert 3)
3	**Worried about my tooth** 1. I am not worried about my tooth 2. I am a little bit worried about my tooth 3. I am quite worried about my tooth	6	0	4	• 'What does worried mean? Or is this essentially for their interpretation?' (Expert 1) • 'This attribute is very generic. If your intention to capture the psychological impact of the tooth cavity, you might

	4. I am very worried about my tooth				have to choose a very specific statement, eg., worry that you might lose your tooth' (Expert 2) • 'Perhaps use 'Concern about tooth cavity', and use 'little' rather than 'bit' in Q2' (Expert 4) • 'Rather than the worried about the cavity their common concerns are discolouration/ aesthetics' (Expert 5)
4	**Able to join in activities (Playing, Sports, School activities)** 1. My tooth cavity causes no difficulty in join in activities 2. My tooth cavity causes a little difficulty in join in activities 3. My tooth cavity causes a quite a lot of difficulty in join in activities 4. My tooth cavity causes lots of difficulty in join in activities	6	0	2	• 'Should it be causes? What about socialising, interacting? Important for adolescents' (Expert 1). • 'Would consider using 'Ability' rather than 'Able', 'My tooth cavity causes' rather than 'my tooth cavity cause', and perhaps use 'participating in activities' rather than 'join in activities' (Expert 4)
5	**Location of tooth cavity** 1. Front tooth 2. Back tooth	2	4	4	• 'Should you think about asking about top and bottom teeth as well? Having a problem with your front top teeth may have very different impacts compared to problems in front bottom teeth' (Expert 1) 'Is it safe to assume adolescents will know what you mean by front and back teeth?' (Expert 1) 'What is they had recent cavities or fillings on both front and back teeth?' (Expert 1) • 'The wording is not clear, as I understand this attribute is related to location of caries, so caries location is more appropriate rather than 'tooth location' (Expert 2) • 'May be confuse when they have multiple cavities. Perhaps more appropriate to refer to 'Front tooth' and 'Back tooth',

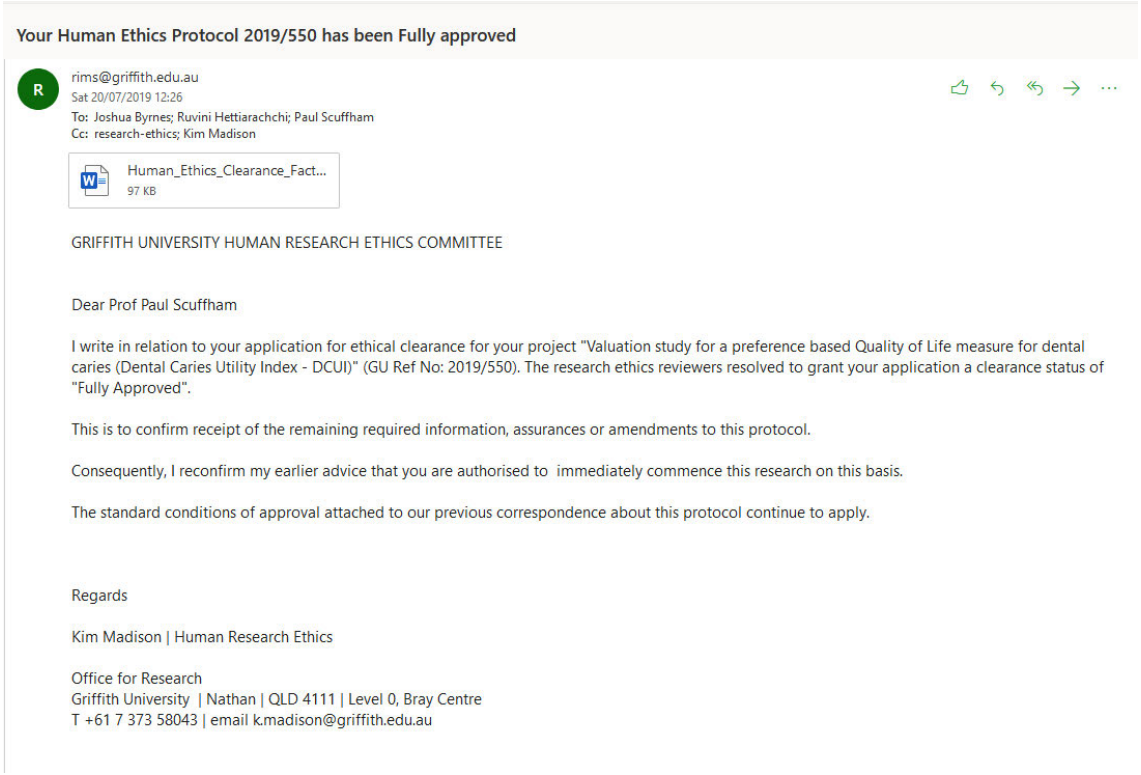
					rather than to 'teeth' (which is plural). As the initial survey question reads: *'Think of the tooth cavity…'* which implies a single tooth, rather than multiple teeth' (Expert 4) • 'Tooth location seems a vague question' (Expert 5)
6	**Appearance of my cavitated/filled/missing tooth** 1. Natural tooth appearance 2. Slightly noticeable tooth cavity/filled tooth 3. Highly noticeable tooth cavity/filled tooth 4. Missing tooth	6	0	3	• Three very different states being asked about (Expert 1). • Noticeability and missing tooth are included in the same question. For instance, a missing posterior tooth might not be noticeable. Also, I do not think noticeability is a major concern when the restoration is in the posterior teeth (Expert 2) • Would consider changing the terms. Cannot see any differences (Expert 6)
7	**Swelling** 1. I have no swelling associated with tooth cavity/filled tooth 2. I have mild swelling associated with tooth cavity/filled tooth 3. I have a large swelling associated with tooth cavity/filled tooth 4. I have a very big swelling associated with tooth cavity/filled tooth	2	4	5	• Again, how is mild and large swelling defined or left to responder to interpret? (Expert 1) • Swelling associated with tooth caries is not a very common finding. You might find very few children with caries who might have pulpal involvement that is associated with swelling (Expert 2) • What a 'mild' swelling is, versus what a 'large' swelling is, Might depend on the individual interpretation of the patient (subjective). Often a patient will report they can feel 'swelling' after/ during a filling, when in fact they are mistaking the sensation of the anaesthetic for an actual swelling (ie there won't actually be swelling, but the patient will think there is 'swelling', as things feel 'odd/different/funny' to them while they are numb) (Expert 4) • Is the significance & implications of 'swelling' already determined by your survey, based on what the individual

	answers in regard to the 'appearance', 'activity', 'pain/discomfort' and 'difficulty eating/drinking' questions? (Expert 3) • Severity of the swelling cannot be assessed by the child (Expert 6)
General comments about the instrument	• 'General comment: At times you specify if you asking them to think about the cavity, or filled tooth, or combination of these plus a missing tooth, could be confusing ... what are most interested in? These three states are quite different' (Expert 1) 'I may struggle if I needed to fill this in ... what are you asking me about, my cavity, filling or missing tooth?' (Expert 1)

*****Assessed the relevance of the included items and response levels with respect to the QoL aspects of dental caries and appropriateness for the target group*

[#]*One expert raised an issue with combining pain and discomfort*

Appendix D: Griffith ethical clearance- Health state valuation study



Your Human Ethics Protocol 2019/550 has been Fully approved

R

rims@griffith.edu.au
Sat 20/07/2019 12:26
To: Joshua Byrnes; Ruvini Hettiarachchi; Paul Scuffham
Cc: research-ethics; Kim Madison

Human_Ethics_Clearance_Fact...
97 KB

GRIFFITH UNIVERSITY HUMAN RESEARCH ETHICS COMMITTEE

Dear Prof Paul Scuffham

I write in relation to your application for ethical clearance for your project "Valuation study for a preference based Quality of Life measure for dental caries (Dental Caries Utility Index - DCUI)" (GU Ref No: 2019/550). The research ethics reviewers resolved to grant your application a clearance status of "Fully Approved".

This is to confirm receipt of the remaining required information, assurances or amendments to this protocol.

Consequently, I reconfirm my earlier advice that you are authorised to immediately commence this research on this basis.

The standard conditions of approval attached to our previous correspondence about this protocol continue to apply.

Regards

Kim Madison | Human Research Ethics

Office for Research
Griffith University | Nathan | QLD 4111 | Level 0, Bray Centre
T +61 7 373 58043 | email k.madison@griffith.edu.au

Appendix E: Ngene codes for the DCE choice tasks design in valuation approach DCE_{TTO}

Dental Caries utility Index- 5 dimensions each 4 levels
DCE_{TTO} with dummy interactions and including duration
Duration attribute: 6 months, 1 year, 4 years, 7 years and 10 years
D-efficient (Zero priors for all betas)

```
Design
? D-efficient (Zero priors for all betas) DCE TTO with dummy interactions
;alts = hs1*, hs2
;rows = 200
;eff = (mnl,d)
;block = 25
;model:
U(hs1) =
b1 [0] * DUR [0.5,1,4,7,10] +
b2.dummy[0|0|0] * Paind[1,2,3,4] +
b3.dummy[0|0|0] * Eatdr[1,2,3,4] +
b4.dummy[0|0|0] * Worry[1,2,3,4] +
b5.dummy[0|0|0] * Ablpa[1,2,3,4] +
b6.dummy[0|0|0] * Apper[1,2,3,4] +

i1[0] * DUR * Paind.dummy[1] +
i2[0] * DUR * Paind.dummy[2] +
i3[0] * DUR * Paind.dummy[3] +
i4[0] * DUR * Eatdr.dummy[1] +
i5[0] * DUR * Eatdr.dummy[2] +
i6[0] * DUR * Eatdr.dummy[3] +
i7[0] * DUR * Worry.dummy[1] +
i8[0] * DUR * Worry.dummy[2] +
i9[0] * DUR * Worry.dummy[3] +
i10[0] * DUR * Ablpa.dummy[1] +
i11[0] * DUR * Ablpa.dummy[2] +
i12[0] * DUR * Ablpa.dummy[3] +
i13[0] * DUR * Apper.dummy[1] +
i14[0] * DUR * Apper.dummy[2] +
i15[0] * DUR * Apper.dummy[3]
/
U(hs2) =
b1 [0] * DUR [0.5,1,4,7,10] +
b2.dummy [0|0|0] * Paind[1,2,3,4] +
b3.dummy [0|0|0] * Eatdr[1,2,3,4] +
b4.dummy [0|0|0] * Worry[1,2,3,4] +
b5.dummy [0|0|0] * Ablpa[1,2,3,4] +
b6.dummy [0|0|0] * Apper[1,2,3,4] +

i1[0] * DUR * Paind.dummy[1] +
i2[0] * DUR * Paind.dummy[2] +
i3[0] * DUR * Paind.dummy[3] +
i4[0] * DUR * Eatdr.dummy[1] +
i5[0] * DUR * Eatdr.dummy[2] +
```

```
i6[0] * DUR * Eatdr.dummy[3] +
i7[0] * DUR * Worry.dummy[1] +
i8[0] * DUR * Worry.dummy[2] +
i9[0] * DUR * Worry.dummy[3] +
i10[0] * DUR * Ablpa.dummy[1] +
i11[0] * DUR * Ablpa.dummy[2] +
i12[0] * DUR * Ablpa.dummy[3] +
i13[0] * DUR * Apper.dummy[1] +
i14[0] * DUR * Apper.dummy[2] +
i15[0] * DUR * Apper.dummy[3]
$
```

Appendix F: Ngene design matrix of the DCE choice tasks design in valuation approach DCE_{TTO}

Appendices-Table 4: Ngene design matrix of the DCE choice tasks design in valuation approach DCE_{TTO}

MNL efficiency measures	
D error	0.012855
A error	0.07817
B estimate	100
S estimate	0

Design													
Choice situation	hs1. dur	hs1. paind	hs1. eatdr	hs1. worry	hs1. ablpa	hs1. apper	hs2. dur	hs2. paind	hs2. eatdr	hs2. worry	hs2. ablpa	hs2. apper	Block
1	0.5	2	3	2	4	1	1	1	4	4	3	3	19
2	4	2	1	1	4	1	4	3	3	4	1	3	18
3	0.5	4	2	2	3	2	1	1	4	1	2	4	14
4	1	3	4	3	3	1	1	1	1	2	2	3	12
5	1	2	2	1	1	2	0.5	4	1	3	3	3	7
6	0.5	1	3	3	2	2	10	1	3	1	2	2	11
7	10	4	2	3	2	2	4	1	3	2	4	3	4
8	7	4	2	3	3	1	7	2	1	1	1	3	22
9	7	1	4	3	3	3	7	4	1	2	2	1	24
10	0.5	4	2	1	1	3	1	1	3	4	3	2	4
11	10	2	2	3	3	2	0.5	2	4	3	1	2	16
12	10	1	1	3	4	4	10	2	3	2	3	3	13
13	7	3	4	2	4	1	7	4	3	1	2	2	18
14	7	2	3	4	1	4	7	1	4	3	2	2	5
15	4	1	2	1	1	1	4	4	1	3	2	2	19
16	4	3	2	3	2	3	7	4	4	1	3	1	2
17	10	3	1	1	3	4	10	2	4	4	4	2	9
18	1	4	3	1	1	3	1	2	1	3	2	1	24
19	1	2	3	1	4	3	0.5	4	2	4	2	4	11
20	7	2	1	3	2	4	10	3	4	2	1	2	10
21	4	2	4	2	4	3	4	3	2	3	1	4	22
22	4	4	4	1	1	2	4	2	2	4	4	1	23
23	1	2	2	4	2	2	1	4	4	2	4	4	8
24	10	3	2	4	3	3	0.5	3	2	4	3	3	19
25	1	1	2	1	1	2	4	3	3	4	3	1	16
26	10	4	2	4	1	3	7	2	4	3	2	4	19
27	0.5	1	3	4	4	3	1	3	4	2	2	2	1
28	0.5	2	2	1	2	3	0.5	1	3	2	3	1	25
29	7	2	4	1	1	2	4	1	2	4	3	3	17
30	1	3	3	2	1	4	4	2	2	4	4	2	20

31	4	4	4	1	1	2	4	2	3	2	4	4	18
32	4	2	3	3	3	3	4	3	4	4	1	4	23
33	7	1	1	2	1	3	4	2	2	3	2	1	25
34	1	4	3	1	2	3	0.5	2	2	3	3	2	7
35	10	1	2	2	2	4	7	2	1	3	4	1	17
36	4	4	3	1	3	1	1	2	4	4	1	4	6
37	10	4	4	3	1	4	10	2	3	1	4	1	5
38	7	4	3	2	4	4	10	1	1	1	1	2	21
39	0.5	1	2	2	3	4	1	3	1	4	4	1	2
40	0.5	2	4	3	2	3	10	2	4	3	2	3	14
41	7	3	4	2	2	3	4	1	2	4	4	4	6
42	0.5	3	3	1	3	3	10	3	3	1	3	3	2
43	10	4	4	4	3	1	0.5	4	4	2	3	1	3
44	1	3	3	4	1	4	4	1	1	2	4	3	24
45	10	4	1	4	2	2	10	1	3	3	1	3	23
46	7	4	3	3	3	1	7	3	2	1	4	3	15
47	0.5	3	1	1	2	1	0.5	4	2	2	4	3	19
48	1	4	4	2	3	2	4	2	3	3	1	1	3
49	7	2	3	1	1	4	7	3	1	4	4	1	18
50	10	3	3	1	1	2	0.5	3	4	1	1	2	4
51	0.5	2	3	4	3	4	0.5	3	2	1	1	3	13
52	4	2	4	3	3	3	10	4	1	2	1	2	9
53	4	4	3	1	4	4	4	3	1	2	3	3	5
54	10	2	2	2	1	3	10	3	4	1	3	2	5
55	0.5	1	3	4	4	2	4	4	2	2	1	3	16
56	0.5	2	4	1	1	1	1	3	1	3	3	3	24
57	10	2	2	4	2	4	1	2	2	4	2	4	11
58	0.5	4	4	1	1	4	10	4	4	1	1	4	4
59	4	4	3	4	4	3	7	2	2	3	1	4	21
60	10	1	2	2	3	1	0.5	1	2	2	3	1	13
61	1	3	2	2	2	2	1	4	4	3	1	4	5
62	1	1	1	3	2	2	0.5	3	2	2	4	3	17
63	10	3	3	4	1	3	10	2	1	1	3	4	14
64	0.5	3	1	4	1	1	10	2	1	3	1	1	21
65	0.5	3	3	2	3	2	0.5	2	4	1	2	1	13
66	1	1	1	4	1	3	0.5	4	3	1	4	4	25
67	4	1	2	1	3	3	7	2	3	2	2	2	1
68	10	1	4	4	3	1	0.5	1	4	4	3	4	25
69	10	3	3	3	4	2	10	1	4	1	1	4	24
70	0.5	3	4	3	4	2	0.5	2	3	2	3	3	3
71	4	2	2	1	4	2	4	1	3	3	3	3	11
72	4	4	3	2	3	3	7	2	1	4	4	4	11
73	1	1	2	2	4	1	1	2	4	3	3	3	13
74	0.5	1	1	1	4	3	10	2	1	1	4	3	8
75	7	4	1	2	3	4	10	2	2	1	2	1	4
76	1	1	4	1	2	1	1	3	2	3	1	3	10
77	0.5	4	2	1	3	2	10	4	2	1	3	2	8
78	1	2	4	2	1	1	4	1	1	1	2	4	3

79	0.5	2	1	3	3	1	1	4	4	4	2	2	14
80	1	2	1	2	3	1	7	4	3	1	4	4	20
81	7	1	1	3	2	4	4	4	2	1	3	1	24
82	1	3	2	3	4	3	0.5	2	4	2	3	4	15
83	7	2	2	3	1	4	7	4	4	1	2	1	18
84	0.5	2	2	1	2	4	10	2	2	1	3	4	22
85	1	2	2	3	3	3	1	4	1	4	2	2	21
86	10	3	2	1	1	1	0.5	3	1	3	1	1	20
87	1	3	1	4	4	4	0.5	4	2	3	1	1	11
88	4	3	4	1	1	2	4	1	1	2	4	4	21
89	4	1	2	2	3	2	1	4	3	3	2	1	11
90	7	1	1	4	1	1	7	4	4	1	4	3	1
91	1	4	2	3	4	4	0.5	2	3	4	1	1	18
92	1	2	3	1	1	4	1	3	4	2	3	1	8
93	4	3	3	3	1	3	7	1	4	2	3	1	25
94	10	2	1	2	3	1	10	1	2	4	4	2	5
95	4	3	3	4	3	2	4	4	1	2	1	4	15
96	10	2	2	2	1	2	10	3	1	3	3	3	16
97	4	1	3	1	2	3	4	4	1	2	1	2	6
98	4	4	4	4	2	3	4	1	3	3	1	1	9
99	7	1	2	1	4	3	7	3	1	4	1	2	7
100	0.5	4	4	3	2	1	0.5	2	2	1	4	4	9
101	10	4	1	4	3	2	0.5	4	1	2	1	2	8
102	0.5	2	1	4	1	2	1	3	4	3	4	1	19
103	4	4	1	4	1	4	4	1	2	3	2	2	21
104	7	3	4	2	4	1	7	1	3	4	2	3	6
105	7	3	4	1	2	4	7	4	1	4	1	3	2
106	10	2	4	4	2	1	7	4	2	3	3	2	2
107	7	1	3	2	1	1	7	2	1	3	3	3	20
108	7	3	2	4	3	4	7	4	4	3	2	1	12
109	0.5	1	1	3	1	3	1	3	2	4	3	4	9
110	0.5	1	4	4	1	3	1	4	3	2	2	2	12
111	7	2	2	3	4	4	7	3	1	1	3	1	8
112	1	1	2	2	2	1	0.5	2	1	1	3	2	3
113	4	4	1	3	4	1	7	2	4	4	2	4	25
114	7	4	1	1	1	1	4	2	2	4	4	3	15
115	7	3	4	4	4	4	7	4	2	3	3	2	21
116	10	4	3	2	1	1	0.5	4	3	1	1	1	22
117	1	1	4	1	3	2	1	2	3	2	2	1	13
118	0.5	1	2	3	4	1	10	1	3	3	4	1	23
119	1	3	2	1	3	4	0.5	1	3	4	4	3	10
120	4	3	3	3	4	2	4	2	2	1	3	1	12
121	10	2	1	2	4	3	10	3	2	3	2	1	1
122	7	1	3	2	2	2	7	3	4	3	1	1	6
123	7	1	1	2	2	2	10	2	4	4	1	4	15
124	7	4	3	2	3	4	7	3	2	4	2	1	14
125	7	3	2	2	3	1	4	1	3	1	1	3	17
126	0.5	1	4	3	1	4	0.5	2	1	1	4	1	10

127	1	3	4	2	4	2	0.5	4	3	1	3	4	20
128	7	1	4	2	4	1	7	4	1	1	1	2	11
129	1	1	4	1	4	1	1	2	3	4	1	2	12
130	1	3	1	2	1	2	4	1	4	3	2	1	25
131	7	1	3	3	1	1	7	2	2	2	3	4	6
132	7	1	2	1	1	4	4	4	3	4	2	2	16
133	10	1	4	4	3	3	0.5	1	4	2	3	3	1
134	1	2	3	4	4	2	10	2	3	4	2	3	3
135	7	1	4	4	2	2	10	3	1	2	1	1	5
136	0.5	1	4	2	2	3	10	1	3	2	2	4	15
137	1	1	3	2	2	2	1	3	1	4	4	1	9
138	4	1	3	3	4	2	1	4	4	4	3	3	16
139	4	2	4	4	3	3	0.5	1	2	2	1	2	8
140	7	3	1	3	2	3	7	4	4	1	1	2	8
141	4	1	1	4	1	1	1	3	3	3	2	2	1
142	7	2	2	4	4	2	7	4	4	3	1	4	14
143	4	2	1	1	3	3	4	3	3	3	2	4	16
144	10	4	2	3	2	3	7	1	3	4	3	2	1
145	1	3	1	4	2	4	10	3	1	2	2	4	20
146	7	3	1	2	3	3	4	4	3	4	2	4	17
147	0.5	3	4	1	2	1	0.5	1	1	3	1	4	7
148	0.5	4	1	4	1	3	1	2	2	2	4	1	4
149	10	2	3	2	3	2	10	4	2	1	2	3	2
150	1	4	1	4	2	2	1	3	3	3	3	4	4
151	0.5	3	2	4	4	4	10	3	2	4	4	4	7
152	1	4	1	2	4	4	0.5	1	2	3	2	3	14
153	0.5	4	2	1	2	2	0.5	3	4	2	1	4	7
154	10	3	3	3	3	4	0.5	3	3	3	4	4	16
155	10	2	1	4	1	2	10	1	2	2	3	4	1
156	1	2	4	2	4	3	1	3	1	1	3	1	18
157	1	3	1	1	2	3	0.5	4	2	4	1	1	7
158	7	1	4	3	2	2	7	3	2	1	1	4	20
159	10	4	2	2	4	1	10	1	3	4	1	4	9
160	4	3	3	4	3	3	1	1	1	2	1	1	18
161	0.5	2	4	2	2	4	1	3	1	1	4	1	13
162	4	2	4	4	4	3	4	3	1	1	2	2	6
163	10	4	4	2	4	2	0.5	4	2	2	4	2	23
164	10	2	4	3	3	2	10	4	3	1	4	3	15
165	10	4	3	1	3	1	10	3	2	2	1	2	2
166	10	4	4	4	4	1	7	1	1	2	2	2	20
167	10	3	4	2	2	4	10	1	2	3	4	1	22
168	10	1	1	3	1	4	1	1	1	3	1	4	5
169	10	1	1	4	4	4	7	2	4	2	3	3	24
170	4	4	1	3	4	4	1	1	3	1	3	2	15
171	4	4	2	3	2	3	4	1	3	2	4	2	12
172	10	1	2	4	2	1	10	3	4	3	4	3	13
173	4	2	1	3	2	4	4	4	4	4	1	2	24
174	4	1	1	4	1	1	7	2	3	1	4	4	2

175	4	2	4	3	2	4	7	3	1	4	4	3	9
176	4	2	1	3	4	4	1	1	3	4	2	1	22
177	10	4	3	4	2	1	10	1	4	1	4	2	19
178	10	4	3	3	4	4	0.5	4	3	3	4	4	17
179	1	3	1	2	2	4	1	2	4	4	3	1	10
180	0.5	2	1	2	4	3	10	2	1	2	4	1	23
181	0.5	2	4	3	4	1	4	3	3	2	2	4	17
182	4	4	2	3	2	4	1	3	4	1	4	2	3
183	1	3	1	2	3	1	1	1	2	4	4	2	3
184	0.5	1	4	1	4	4	1	2	3	3	2	3	17
185	7	4	4	3	1	1	7	3	2	1	3	2	22
186	4	3	4	2	2	3	4	4	2	3	3	2	12
187	7	3	3	4	3	4	7	1	1	3	1	3	21
188	4	3	1	4	2	1	4	1	3	1	4	3	10
189	7	3	3	1	1	1	4	2	2	2	3	2	25
190	4	4	3	1	4	3	4	2	1	4	2	4	10
191	1	1	2	1	1	4	0.5	4	4	3	4	2	14
192	0.5	1	1	3	3	2	0.5	3	3	2	1	3	19
193	7	3	3	1	4	4	10	1	1	4	3	3	10
194	7	3	3	1	3	2	7	4	4	2	2	3	22
195	0.5	2	1	3	3	2	0.5	3	2	4	2	1	4
196	1	3	3	4	1	2	1	4	1	1	4	4	12
197	1	4	2	4	4	2	1	1	4	2	2	3	7
198	0.5	4	2	4	3	1	0.5	3	4	1	4	2	23
199	0.5	4	1	2	2	3	10	1	1	2	2	3	6
200	4	2	3	1	2	1	4	4	4	4	4	4	23

hs1- health state 1; hs 2- health state 1; dur-duration; paind –pain/discomfort; eatdr- difficulty eating food/drinking; worry-worried; ablpa- ability to participate in activities; apper- appearance.

Duration levels codes (0.5-6 months, 1-1 year, 2-4 years, 3-7 years, 4-10 years)

Dimension level codes (1-level 1, 2-level 2, 3-level 3, 4-level 4)

Appendix G: Ngene codes for the DCE choice tasks design in valuation approach DCE_{VAS} and main survey

Dental Caries utility Index- 5 dimensions each 4 levels
D-efficient (Zero priors for all betas)
DCE with dummy interactions

```
Design
? D-efficient (Zero priors for all betas) DCE dummy interactions
;alts = hs1*, hs2*
;rows = 200
;eff = (mnl,d)
;block = 25
;model:
U(hs1) =
β1.dummy[0|0|0] * Paind[1,2,3,4] +
β2.dummy[0|0|0] * Eatdr[1,2,3,4] +
β3.dummy[0|0|0] * Worry[1,2,3,4] +
β4.dummy[0|0|0] * Ablpa[1,2,3,4] +
β5.dummy[0|0|0] * Apper[1,2,3,4] +

/
U(hs2) =
β1.dummy [0|0|0] * Paind[1,2,3,4] +
β2.dummy [0|0|0] * Eatdr[1,2,3,4] +
β3.dummy [0|0|0] * Worry[1,2,3,4] +
β4.dummy [0|0|0] * Ablpa[1,2,3,4] +
β5.dummy [0|0|0] * Apper[1,2,3,4]

$
```

Appendix H: Ngene design matrix of the DCE choice tasks design in valuation approach DCE_{VAS} and main survey

Appendices-Table 5: Ngene design matrix of the DCE choice tasks design in valuation approach DCE_{VAS} and main survey

MNL efficiency measures	
D error	0.047634
A error	0.06003
B estimate	100
S estimate	0

Design											
Choice situation	hs1. paind	hs1. eatdr	hs1. worry	hs1. ablpa	hs1. apper	hs2. paind	hs2. eatdr	hs2. worry	hs2. ablpa	hs2. apper	Block
1	2	1	2	2	2	4	4	4	1	3	15
2	4	2	1	1	2	3	1	4	4	3	6
3	3	4	1	1	2	4	1	3	4	3	3
4	1	3	1	4	3	2	1	2	2	4	18
5	1	2	3	1	3	2	3	4	3	4	10
6	2	3	4	3	1	1	4	1	4	3	1
7	2	4	2	4	4	4	3	3	2	3	20
8	3	3	1	3	3	2	2	2	4	1	13
9	1	2	3	1	4	2	4	4	2	3	16
10	3	3	1	2	4	1	4	3	4	2	20
11	3	2	3	1	1	1	1	2	4	4	9
12	1	1	3	2	3	3	4	2	1	2	5
13	4	3	4	2	4	2	4	3	4	2	7
14	3	4	1	3	1	2	2	4	4	2	20
15	2	2	2	3	4	4	1	3	4	1	23
16	4	4	1	3	1	3	1	4	4	2	17
17	4	1	2	1	1	1	4	3	2	4	14
18	1	4	4	2	3	2	2	1	1	1	1
19	3	2	4	4	2	2	3	3	3	1	15
20	1	2	1	2	2	4	3	2	4	3	8
21	1	3	1	3	4	4	1	2	1	1	9
22	1	2	3	1	1	4	1	1	4	4	19
23	1	3	4	3	1	4	4	1	4	3	22
24	2	2	3	1	4	3	4	2	3	1	7
25	4	4	4	1	1	3	3	2	2	3	19
26	4	4	1	2	4	3	1	3	3	2	11
27	3	2	2	3	1	1	1	4	2	4	9
28	4	1	2	2	2	2	4	1	1	4	8

29	4	3	2	2	3	1	1	4	3	4	20
30	3	4	3	1	1	1	3	1	3	2	22
31	1	2	3	3	1	4	3	1	4	4	14
32	4	1	1	4	1	1	2	4	1	3	3
33	2	2	2	4	2	3	3	3	1	1	22
34	3	1	1	4	3	4	2	3	3	2	18
35	4	1	3	3	2	1	3	2	2	1	25
36	4	1	4	1	3	2	2	2	3	2	18
37	4	2	3	2	3	2	1	1	3	4	21
38	1	2	3	4	4	4	1	1	1	2	17
39	2	1	3	1	2	4	2	2	4	3	5
40	4	4	3	3	4	3	1	4	2	2	18
41	3	3	4	3	2	4	1	3	1	4	3
42	3	3	4	1	1	2	2	1	2	3	11
43	4	2	3	3	2	1	4	1	2	3	12
44	4	4	4	2	2	3	3	1	1	3	3
45	4	3	3	4	1	3	2	1	1	4	16
46	4	2	2	1	4	1	4	3	3	2	3
47	3	1	2	3	3	1	3	1	1	4	15
48	4	4	3	1	3	1	2	4	2	4	20
49	4	3	2	3	4	2	1	1	1	2	10
50	1	2	2	3	1	3	1	3	2	2	7
51	2	4	4	3	2	3	2	1	1	4	12
52	3	1	2	4	1	4	3	1	1	2	16
53	1	2	2	1	1	3	1	3	2	2	24
54	2	4	2	3	1	3	3	4	4	2	18
55	3	2	2	2	4	1	3	4	1	1	19
56	1	2	3	2	2	4	4	2	3	3	7
57	2	3	3	3	2	3	2	1	2	1	21
58	2	3	1	1	2	4	2	4	2	4	25
59	1	3	2	2	2	2	4	4	3	3	9
60	1	2	2	1	2	3	1	3	2	1	23
61	3	1	1	2	1	4	2	2	1	2	25
62	3	1	3	2	1	4	2	2	4	3	25
63	3	2	4	3	4	1	3	2	2	1	7
64	4	4	4	4	3	3	3	2	1	4	4
65	1	3	2	1	4	3	2	1	4	3	8
66	2	4	4	3	3	3	2	3	4	4	17
67	1	3	4	4	2	4	4	3	3	3	21
68	2	4	1	1	2	3	3	3	3	1	10
69	1	1	3	1	4	4	4	1	3	3	5
70	1	4	4	4	4	2	3	3	3	2	22
71	2	1	4	1	4	3	4	3	2	3	12
72	4	2	1	4	2	2	3	3	2	3	19
73	4	2	4	3	2	2	1	2	1	1	7

74	2	2	2	4	2	4	3	1	3	4	11
75	1	4	4	2	1	4	1	1	3	4	14
76	2	3	4	4	4	3	4	3	1	2	1
77	4	3	3	4	2	1	1	2	2	4	6
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79	4	3	1	3	1	3	4	4	1	3	2
80	4	1	4	4	1	1	3	2	2	4	6
81	1	3	3	4	4	2	2	1	1	1	4
82	3	3	2	4	2	4	2	1	2	3	10
83	4	1	1	4	1	2	4	2	2	4	22
84	1	1	2	1	3	2	4	3	2	1	14
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87	1	1	2	1	3	2	3	4	2	2	20
88	1	2	1	2	2	2	3	2	1	1	9
89	3	4	1	1	4	1	3	3	3	1	13
90	3	1	3	1	3	4	4	1	4	1	4
91	4	4	2	3	4	2	1	1	2	1	14
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93	3	1	2	2	3	1	2	1	1	2	17
94	1	1	1	3	1	2	2	4	2	4	6
95	4	1	4	2	1	2	3	3	1	4	5
96	3	4	4	1	2	2	2	1	2	4	23
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100	3	4	2	4	4	1	2	4	2	1	5
101	4	3	4	3	3	3	4	2	4	1	15
102	2	1	1	2	3	1	3	2	4	4	25
103	4	2	4	3	4	2	4	2	4	1	25
104	1	4	2	3	4	2	3	4	1	3	1
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106	3	4	2	1	4	2	2	3	3	2	21
107	2	3	4	4	3	3	4	2	2	1	2
108	2	1	1	1	4	3	2	3	2	1	2
109	3	2	1	4	1	4	1	3	1	2	21
110	1	2	4	4	4	3	3	3	2	1	6
111	3	1	1	4	4	2	3	2	2	3	3
112	1	1	3	4	1	4	3	2	3	4	13
113	4	2	1	4	3	3	4	2	2	4	10
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116	3	4	3	4	4	4	3	1	2	2	18
117	2	3	4	4	1	1	1	2	1	2	12
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125	2	1	2	3	3	4	3	1	1	2	24
126	2	3	2	2	3	1	4	4	3	1	20
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132	2	2	2	1	1	3	4	4	4	2	14
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145	3	2	2	4	3	4	4	4	1	4	24
146	3	1	1	3	3	1	2	4	2	1	1
147	1	2	3	2	4	3	1	4	3	2	12
148	1	3	4	2	3	4	4	3	4	4	16
149	4	3	4	4	4	3	2	1	1	2	16
150	4	4	2	2	2	1	1	4	4	3	6
151	2	3	1	4	2	3	4	2	1	3	8
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157	4	2	3	2	3	3	4	1	3	2	22
158	1	4	4	1	3	2	2	1	3	1	25
159	3	2	2	4	1	1	1	3	1	2	16
160	3	2	2	2	2	2	3	4	4	1	20
161	2	4	4	2	1	1	1	3	3	4	24
162	3	3	4	1	4	4	2	3	2	1	3
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184	4	1	2	4	2	2	2	3	3	3	19
185	1	4	3	2	2	2	3	4	1	1	11
186	2	3	1	1	1	3	2	4	3	2	17
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191	1	1	2	1	1	3	4	4	4	3	2
192	4	1	2	3	1	3	3	3	2	3	2
193	3	2	1	3	1	1	4	4	1	2	8
194	2	2	2	1	3	3	3	1	3	4	10
195	3	3	1	2	2	2	2	3	4	4	9
196	1	4	1	3	3	2	1	4	4	4	4
197	2	1	4	3	3	3	3	2	4	2	11
198	3	3	4	1	1	2	1	1	4	2	4
199	1	4	3	3	4	2	2	2	1	2	12
200	2	1	4	2	2	4	3	1	4	3	12

hs1- health state 1; hs 2- health state 1; paind –pain/discomfort; eatdr- difficulty eating food/drinking; worry-worried; ablpa- ability to participate in activities; apper- appearance.

Dimension level codes (1-level 1, 2-level 2, 3-level 3, 4-level 4)

Appendix I: The online survey

Preference-based quality of life measure for dental caries
(Dental Caries Utility Index - DCUI)
(GU ref no: 2019/550)
ONLINE INFORMATION SHEET

Thank you very much for your interest in participating in this study. This study is about tooth decay. Your participation in this research is voluntary, and you will remain anonymous.

Please read the Participant Information sheet in full before consenting to participate in this study.

Who is eligible to participate in the study?

Adults over 18 years of age residing in Australia are eligible to participate.

Why is the research being conducted?

This survey is about tooth decay (tooth cavity/ rotten tooth) and how tooth decay impacts on quality of life and day to day activities. The results of this study will be used to develop a value set that will facilitate economic evaluations of tooth decay prevention programmes. This will assist researchers and policy-makers to identify best programmes to prevent and treat tooth decay.

This study is conducted by the Center for Applied Health Economics, led by Prof. Paul Scuffham, Griffith University, Brisbane and the survey is conducted as a part of the research project leading to a PhD degree for Ruvini Hettiarachchi.

What will you be asked to do?
If you decide to take part in the study, you will be asked to complete a short online survey with a few basic questions about you such as age, and then you will be asked about your preferences with regard to some scenarios of tooth decay. This survey will take approximately 15 minutes to complete.

The expected benefits of the research
It is not expected that there will be direct benefits to you for participating in this survey; however, the results will be used to identify the choices and concerns of the general public around dental health to decide on best programmes to prevent tooth

decay.

Risks to you
There are no anticipated risks to you from participating in this study. The questionnaire is to be completed online at your convenience. It is important for us that you do not disrupt normal activities because of our research.

Your confidentiality
We do not ask for personal identification details in this survey. Your information will be treated confidentially at all times and the survey responses will be de-identified. Participants will remain anonymous in research outputs and your identity will not be revealed in any publication which reports the findings of this research. All research data (survey responses and analysis) will be retained in a password protected electronic file at Griffith University for a period of five years before being destroyed.

Your participation is voluntary
Your participation in this research activity is absolutely voluntary. You are free not to participate. Even after you consent to participate, you can withdraw your consent and information at any time during the survey. Please note that after submitting the survey responses you will not be able to withdraw, as the survey is anonymous, and we will not be able to identify individual responses. You will not be penalized or affected in any way for participating (or not participating) in this research, and your access to healthcare will not be affected in any way.

Questions / further information
If you have any questions about this study or for further clarification, please contact Dr. Ruvini Hettiarachchi, on (07) 37359109 or email ruvini.hettiarachchi@griffithuni.edu.au.

The ethical conduct of this research
This study has been approved by the Griffith University Human Research Ethics Committee (GU Ref No: 2019/550). If you have any concerns or complaints about the ethical conduct of this research project, please contact the Manager, Research Ethics on 3735 4375 or research-ethics@griffith.edu.au.

Feedback to you
The results of this study will not be reported directly to you at the end of the study, however will be published in academic journals and conference presentations. If you wish to access a plain language summary of the research results, please contact Dr. Ruvini Hettiarachchi, on (07) 37359109 or email ruvini.hettiarachchi@griffithuni.edu.au or email at CAHE@griffith.edu.au

CONSENT FORM

By agreeing to participate in the survey, I confirm that:

- I have read the information about this survey;
- I understand that my participation in this research is absolutely voluntary;
- I understand that I am free to withdraw at any time during the survey;
- I understand that the responses I provide to the survey will be combined with those provided by other participants and will be analyzed for presentation in published reports and articles, and that no personally identifiable information about me will appear in any report or article.
- I understand that if I have any additional questions I can contact the research team;

If you have any questions about this survey, or you have problems completing this survey, please contact Dr. Ruvini Hettiarachchi, on (07)37359109 or email ruvini.hettiarachchi@griffithuni.edu.au.

Do you agree to participate in the survey?

Select only one answer

- I agree to participate in the survey
- I do not agree to participate in the survey

Screening Questions

In this section, we would like to know about you.

1. What is your age?

2. What is your gender?

Select only one answer

Male

Female

3. Which state do you live in?

Select only one answer

Dental Caries Utility Index page

In this section, we would like to know about your dental health.

Please tick one box from each category, which best describes your situation over the past 4 weeks due to a tooth cavity, a filling or missing tooth.

Pain/Discomfort

Select only one answer

- I have no pain or discomfort
- I have a little pain or discomfort
- I have quite a lot of pain or discomfort
- I have lots of pain or discomfort

Difficulty eating foods /drinking

Select only one answer

- I have no difficulty in eating food/ drinking
- I have a little difficulty in eating food/ drinking
- I have quite a lot of difficulty in eating food/ drinking
- I have lots of difficulty in eating food/ drinking

Worried (e.g. about losing a tooth, etc.)

Select only one answer

- I am not worried
- I am a little bit worried
- I am quite worried
- I am very worried

Ability to participate in activities (e.g. playing with your friends, sports, school work, etc.)

Select only one answer

- I have no difficulty participating in activities
- I have a little difficulty participating in activities
- I have quite a lot of difficulty participating in activities
- I have lots of difficulty participating in activities

Appearance

Select only one answer

- I am not concerned about my appearance
- I am a little concerned about my appearance
- I am quite concerned about my appearance
- I am very concerned about my appearance

DCE choice tasks

In the next section you will be given two hypothetical scenarios of tooth decay (Health state A and Health state B) in each task. Please read the instructions carefully on how to complete the tasks.

DCE choice tasks

Introduction

In the next section you will be given two hypothetical scenarios of tooth decay (Health state A and Health state B) in each task.

Please read the health states given and choose which health state you think is better compared to the other.

There are 10 tasks to be completed including the practice task.

Please assume that except for what has been described, all else to be the same.

There are no right or wrong answers. We would like to know your choice only.

Example DCE choice task in DCE_{TTO} approach in the comparison study (Chapter 5)

Please consider that you are living a life with tooth decay in health state A or B for the same period of time. During this time your health state would not change and then you will die.
Which health state do you think is better (health state A or health state B)?

	Health State A	Health State B
Pain/Discomfort	I have **a little** pain or discomfort	I have **no** pain or discomfort
Difficulty eating foods /drinking	I have **quite a lot of difficulty** in eating food/ drinking	I have **a little difficulty** in eating food/ drinking
Worried (e.g. about losing a tooth, etc.)	I am **a little bit** worried	I am **not** worried
Ability to participate in activities (e.g. playing with your friends, sports, school work, etc.)	I have **no difficulty** participating in activities	I have **quite a lot of difficulty** participating in activities
Appearance	I am **very concerned** about my appearance	I am **a little concerned** about my appearance
Duration of life	Stay this health state for **6 months and then die**	Stay this health state for **1 year and then die**
Which would you choose?	○ Health State A	○ Health State B

Example DCE choice task in DCE_{VAS} approach in comparison study (Chapter 5) and the main survey (Chapter 6)

Please consider that you are living a life with tooth decay in health state A or B for the same period of time. During this time your health state would not change.
Which health state do you think is better (health state A or health state B)?

	Health State A	Health State B
Pain/Discomfort	I have **a little** pain or discomfort	I have **no** pain or discomfort
Difficulty eating foods /drinking	I have **quite a lot of difficulty** in eating food/ drinking	I have **a little difficulty** in eating food/ drinking
Worried (e.g. about losing a tooth, etc.)	I am **a little bit** worried	I am **not** worried
Ability to participate in activities (e.g. playing with your friends, sports, school work, etc.)	I have **no difficulty** participating in activities	I have **quite a lot of difficulty** participating in activities
Appearance	I am **very concerned** about my appearance	I am **a little concerned** about my appearance
Which would you choose?	○ Health State A	○ Health State B

Please consider 10 tasks with choosing between health state A and B for answering following questions.

What factor did you consider mainly to select between health states A and B?

Select only one answer

Difficulty eating foods /drinking

Worried (e.g. about losing a tooth, etc.)

Ability to participate in activities (e.g. playing with your friends, sports, school work, etc.)

Appearance

What factor did you consider **least** to select between health states A and B?

Select only one answer

Pain/discomfort

Difficulty eating foods /drinking

Worried (e.g. about losing a tooth, etc.)

Ability to participate in activities (e.g. playing with your friends, sports, school work, etc.)

Appearance

How difficult was it for you to understand the above tasks?

Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

How difficult was it for you to complete the above tasks?

Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

Visual analogue scale task in DCE_{VAS} approach in comparison study (Chapter 5) and the main survey (Chapter 6)

In the next task, we would like you to indicate how good or bad the given six health scenarios.

Please read the six health states (A-F) on the next page.

- **A,B,C,D and E - Living a life with a tooth decay with different features**
- **F - Being dead**

We have drawn a scale numbered from 0 to 100.
0 means the worst oral health you can imagine.
100 means the best oral health you can imagine.

We would like you to indicate on the scale how good or bad each of the six health states is, in your opinion.
Please do this by clicking on each box and dragging it into the scale.
We would like you to do this for all the six health states.

Feel free to move each health state on to the scale until you are happy with where you have placed them.
A number indicating, where you have placed each health state, will appear in brackets () at the bottom of each box containing the health state descriptive.

Please click 'Next' to begin

We would like to know how good or bad the given health scenarios.

Please read the six scenarios (A-F) below and decide how good or bad you think they are.

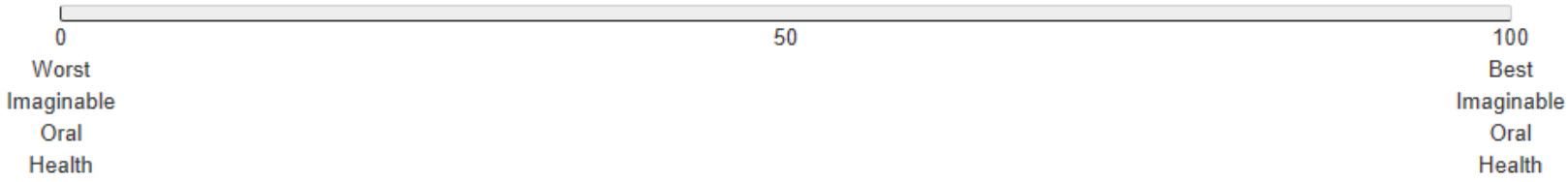
Then please click on each scenario and drag and drop it into the rating scale below to indicate your score between 0-100.

The rating scale numbered from 0 to 100.

0 means the worst oral health you can imagine.

100 means the best oral health you can imagine.

A Living a life with tooth decay with following features I have **lots of** pain or discomfort I have **lots of** difficulty in eating foods / drinking I am **very** worried I have **lots of** difficulty participating in activities I am **very** concerned about my appearance ()	**B** Living a life with tooth decay with following features I have **lots of** pain or discomfort I have **lots of** difficulty in eating food /drinking I am **very** worried I have **quite** difficulty participating in activities I am **not** concerned about my appearance ()	**C** Living a life with tooth decay with following features I have **quite a lot of** pain or discomfort I have **quite a lot of** difficulty in eating food /drinking I am **very** worried I have **no** difficulty participating in activities I am **not** concerned about my appearance ()
D Living a life with tooth decay with following features I have **a little** pain or discomfort I have **a little** difficulty in eating food /drinking I am **a little** bit worried I have **no** difficulty participating in activities I am **not** concerned about my appearance ()	**E** Living a life with tooth decay with following features I have **no** pain or discomfort I have **no** difficulty in eating food /drinking I am **not** worried I have **no** difficulty participating in activities I am **not** concerned about my appearance ()	**F** Death ()



Please consider the rating scale task for answering following questions.

How difficult was it for you to understand the above task?
Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

How difficult was it for you to complete the above task?
Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

Please consider 10 tasks with choosing between health state A and B AND the rating scale task for answering following questions.

How difficult was it for you to understand the above tasks (choosing between health state A and B AND the rating scale task) as a whole?

Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

How difficult was it for you to complete the above tasks (choosing between health state A and B AND the rating scale task) as a whole?

Select only one answer

Not Difficult

Somewhat Difficult

Difficult

Extremely Difficult

Please enter your ideas/ thoughts/ comments about the tasks (choosing between health state A and B AND the rating scale task) you completed above

In this section, we would like to know about your general health.

EQ-5D Online

Please click the ONE box that best describes your health TODAY.

MOBILITY

Select only one answer

I have no problems with walking around

I have slight problems with walking around

I have moderate problems with walking around

I have severe problems with walking around

I am unable to walk around

Please click the ONE box that best describes your health TODAY.

PERSONAL CARE

Select only one answer

I have no problems with washing or dressing myself

I have slight problems with washing or dressing myself

I have moderate problems with washing or dressing myself

I have severe problems with washing or dressing myself

I am unable to wash or dress myself

Please click the ONE box that best describes your health TODAY.

USUAL ACTIVITIES *(e.g. work, study, housework, family or leisure activities)*

Select only one answer

I have no problems doing my usual activities

I have slight problems doing my usual activities

I have moderate problems doing my usual activities

I have severe problems doing my usual activities

I am unable to do my usual activities

Please click the ONE box that best describes your health TODAY.

PAIN / DISCOMFORT

Select only one answer

I have no pain or discomfort

I have slight pain or discomfort

I have moderate pain or discomfort

I have severe pain or discomfort

I have extreme pain or discomfort

Please click the ONE box that best describes your health TODAY.

ANXIETY / DEPRESSION

Select only one answer

I am not anxious or depressed

I am slightly anxious or depressed

I am moderately anxious or depressed

I am severely anxious or depressed

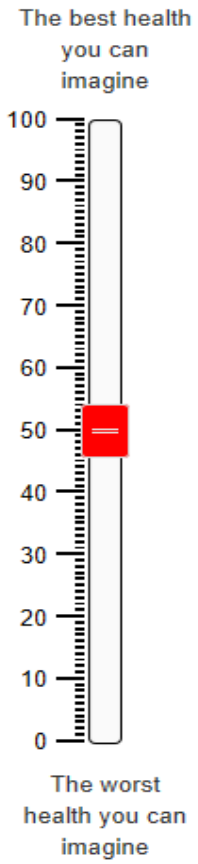
I am extremely anxious or depressed

- We would like to know how good or bad your health is TODAY.
- This scale is numbered from 0 to 100.
- 100 means the best health you can imagine.
  0 means the worst health you can imagine.
- Please click on the scale to indicate how your health is TODAY.

YOUR

HEALTH

TODAY



OHIP-14 Questionnaire

This questionnaire asks how issues with your teeth, mouth or dentures may have caused problems in your daily life. We would like you to complete the questionnaire even if you have good dental health. We would like to know how often you have had each of the 14 listed problems during the LAST MONTH.

HOW OFTEN have you had the problem during the last month?

1. **Have you had trouble pronouncing any words** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

2. **Have you felt that your sense of taste has worsened** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

3. **Have you had painful aching in your mouth?**

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

4. **Have you found it uncomfortable to eat any foods** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

5. **Have you been self-conscious** because of your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

6. **Have you felt tense** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

7. **Has your diet been unsatisfactory** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

8. **Have you had to interrupt meals** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

9. **Have you found it difficult to relax** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

10. **Have you been a bit embarrassed** because of problems with your teeth, mouth or dentures?

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

11. **Have you been a bit irritable with other people** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

12. **Have you had difficulty doing your usual jobs** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

13. **Have you felt that life in general was less satisfying** because of problems with your teeth, mouth or dentures?

Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

14. **Have you been totally unable to function** because of problems with your teeth, mouth or dentures?

Select only one answer
Select only one answer

- Never
- Hardly ever
- Occasionally
- Fairly often
- Very often

Socio-demographics

In this section, we would like to know more about you.

1. What is your highest level of education?

Select only one answer

Bachelor Degree and above

Diploma/ Advanced Diploma

Trade Certificate

Completed high school

Year 11 or below

2. Which best describes your marital status?

Select only one answer

Never married

Married/ De facto

Divorced/ Separated

Widowed

Prefer not to say

3. Roughly how much is your **gross** household income **per week**?

Select only one answer

Less than $1000

$1000- $2999

$3000- $4499

$4500-$5999

$6000 or more

4. Were you born in Australia?

Select only one answer

Yes

No

Oral Health

In this section, we would like to know more about your dental health.

1. How would you consider the condition of your teeth and mouth today?

Select only one answer

Excellent

Very good

Good

Fair

Poor

2. How much do your teeth or mouth bother you in your everyday life?

Select only one answer

Not at all

A little

Some

A lot

Very much

3. How often do you visit the dentist?

Select only one answer

Once every 6 months

When I have an oral health problem

Never

4. Do you have private insurance for dental treatment?

Select only one answer

Yes

No

5. Have you had toothache in the last 6 months?

Select only one answer

Yes

No

6. Do you have untreated decayed teeth (tooth cavities/ rotten teeth)?

Select only one answer

Yes

No

Not Sure

7. Do you have any filled teeth?

Select only one answer

Yes

No

Not Sure

8. Do you have any missing teeth (removed/ extracted) because of tooth decay?

Select only one answer

Yes

No

Not Sure

Appendix J: Approval to use EQ-5D-5L questionnaire

EQ-5D

APPENDIX 1 - Designated use

Licensee is entitled to use and reproduce the (full version of the) EQ-5D-5L Laptop/Desktop version for the sole purposes of carrying out / administrating Licensee's Study/Studies listed below. Licensee shall make available and administer the (full version of the) EQ-5D-5L Laptop/Desktop version to respondents participating in the below Study(ies).

Study	*Starting date*	*Finishing date*
VALUATION STUDY FOR A PREFERENCE-BASED QUALITY OF LIFE MEASURE FOR DENTAL CARIES (DENTAL CARIES UTILITY INDEX- DCUI) AMONG AUSTRALIAN ADOLESCENTS	*January 2020*	*December 2020*

The EQ-5D-5L Laptop/Desktop version will be made applicable to Licensee in the following Language:

English (Australia)

Licensee is entitled to use the above listed Language solely in the Territory referred to between the brackets, unless written permission is obtained from Licensor for the use of a Language in another country.

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Appendix K: Sensitivity analysis- DCE_{VAS} approach of the comparison study

Appendices-Table 6: Parameter estimates from the unadjusted DCE model excluding participants met VAS exclusion criteria in valuation approach 2 DCEVAS

	Model 3: excluding participants who met VAS exclusion criteria (n = 151)	
Dimension and level	**β**	**SE**
pain/discom2	–0.3209	0.1688
pain/discom3	–2.1849*	0.2833
pain/discom4	–2.2590*	0.2895
difficult eat/drink2	–0.5390*	0.1839
difficult eat/drink3	–1.1388*	0.2097
difficult eat/drink 4	–1.3344*	0.2304
worried2	–0.4332*	0.1635
worried3	–0.6956*	0.1615
worried4	–0.7352*	0.1928
abilityparticipate2	–0.0750	0.1999
abilityparticipate3	–0.6002*	0.2183
abilityparticipate4	–0.5357*	0.2030
appearance2	0.0026	0.1880
appearance3	–0.3321	0.1849
appearance4	–0.5553*	0.1961
Goodness-of-fit statistics		
Number of observations	784	
R^2	0.3810	
AIC	702.74	
BIC	783.10	

*Significance at $p < 0.05$

Appendix L: Steps of anchoring based on the DCE_{VAS} approach of the comparison study

Appendices-Table 7: Anchoring modelled DCE coefficients to the full health-dead scale using VAS in comparison study

	Coefficient	Step 1	Step 2 DCE	Rescaled to the PITS*	Rescaled based on mapping**
Pain/Discomfort					
1	0.6963	0.9893	0.3076	0.3076	0.2569
2	0.5641	0.8571	0.2665	0.2716	0.2226
3	-0.5874	-0.2945	-0.0916	-0.0418	-0.0765
4	-0.6731	-0.3802	-0.1182	-0.0651	-0.0987
Difficulty eating food/drinking					
1	0.4173	0.7102	0.2208	0.2208	0.1845
2	0.1330	0.4259	0.1324	0.1434	0.1106
3	-0.2231	0.0698	0.0217	0.0466	0.0181
4	-0.3271	-0.0342	-0.0106	0.0183	-0.0089
Worried					
1	0.2967	0.5896	0.1833	0.1833	0.1531
2	0.0267	0.3196	0.0994	0.1098	0.0830
3	-0.1511	0.1418	0.0441	0.0615	0.0368
4	-0.1723	0.1206	0.0375	0.0557	0.0313
Ability to participate in activities					
1	0.1892	0.4821	0.1499	0.1499	0.1252
2	0.0829	0.3758	0.1169	0.1210	0.0976
3	-0.1244	0.1685	0.0524	0.0646	0.0438
4	-0.1477	0.1452	0.0451	0.0582	0.0377
Appearance					
1&2	0.1522	0.4451	0.1384	0.1384	0.1156
3	-0.0078	0.2851	0.0886	0.0949	0.0741
4	-0.1444	0.1485	0.0462	0.0577	0.0386
HS(11111)	1.7517	3.2163	1.0000	1.0000	1.0000
HS(44444)	-1.4646	0.0000	0.0000	0.1248	0.1646
HS(44444)/5	-0.2929				

** Rescaled to the PITS 0.1248 **Rescaled based on Mapping (regression coefficient 0.8354)*

The rescaling steps are based on the valuing of the Child Health Utility 9D health states to develop a new adolescent-specific scoring algorithm (Personnel communication with authors (1,2)

Model 1 specified in Table 6.3 was effect coded and the base level was recovered. To rescale it based on PITS heath state, effect coded coefficients in the DCE latent scale were rescaled based on the adjusted mean VAS score for the worst health state 0.1248 **(Table 6.4)**. The calculated utility values for four health states (44444, 44431, 33341 and 22211) in the DCE latent scale were 0.0000, 0.7866, 0.2559 and 0.0995, respectively. To map VAS onto DCE, these calculated utility values were regressed onto the adjusted VAS scores shown in **Table 6.4**, as outlined in Equation 6. The regression coefficient was 0.8353827.

```
. gen dVAS=1-vas

. gen dDCE=1-dce

. reg dVAS dDCE, nocons
```

Source	SS	df	MS	Number of obs	=	4
				F(1, 3)	=	437.65
Model	1.68193987	1	1.68193987	Prob > F	=	0.0002
Residual	.011529338	3	.003843113	R-squared	=	0.9932
				Adj R-squared	=	0.9909
Total	1.69346921	4	.423367302	Root MSE	=	.06199

dVAS	Coef.	Std. Err.	t	P>|t|	[95% Conf.	Interval]
dDCE	.8353827	.0399321	20.92	0.000	.7083011	.9624644

Appendix M: Sensitivity analysis- DCE_{VAS} of the health state valuation study

Appendices-Table 8: Characteristics of the participants excluded and not excluded based on VAS exclusion criteria

Characteristic	Participants included n = 521	Participants excluded based on VAS exclusion criteria n = 474	P-value*
Age, mean (SE)	48.06 (0.76)	45.57 (0.89)	0.033
Sex, n (%)			
Male	219 (42.03)	260 (54.85)	0.000
Female	302 (57.97)	214 (45.15)	
Highest level of Education*, n (%)			
Year 11 or below	71 (13.63)	44 (9.28)	0.037
Completed high school	91 (17.47)	64 (13.5)	
Trade certificate	68 (13.05)	68 (14.35)	
Diploma/advanced diploma	73 (14.01)	87 (18.35)	
Bachelor's degree and above	218 (41.84)	211 (44.51)	
Weekly Income* AUD, n (%)			
Less than $1,000	188 (36.08)	141 (29.75)	0.000
$1,000–$2,999	211 (40.5)	149 (31.43)	
$3,000–$4,499	67 (12.86)	72 (15.19)	
$4,500–$5,999	22 (4.22)	53 (11.18)	
$6,000 or more	33 (6.33)	59 (12.45)	
Oral health status*, n (%)			
Excellent	38 (7.29)	107 (22.57)	0.000
Very good	154 (29.56)	123 (25.95)	
Good	181 (34.74)	134 (28.27)	
Fair	109 (20.92)	64 (13.5)	
Poor	39 (7.49)	46 (9.7)	

** t-test for continuous data and chi-square with continuity correction for categorical data*

Appendices-Table 9: Parameter estimates from conditional logit model by excluding the DCE data of the participants who met VAS exclusion criteria

	Model 2: Unadjusted model excluding participants who met VAS exclusion criteria (n = 521)	
	β	Robust SE
pain/discom2	-0.4925***	0.0725
pain/discom3	-1.9156***	0.1085
pain/discom4	-2.1353***	0.1153
difficult eat/drink2	-0.2976***	0.0696
difficult eat/drink3	-0.9864***	0.0827
difficult eat/drink 4	-1.0113***	0.0856
worried2	-0.0759	0.0698
worried3	-0.2584***	0.0740
worried4	-0.5191***	0.0777
abilityparticipate2	-0.1880**	0.0715
abilityparticipate3	-0.5862***	0.0798
abilityparticipate4	-0.5936***	0.0772
appearance2	-0.0534	0.0732
appearance3	-0.3106***	0.0784
appearance4	-0.5664***	0.0819
Goodness-of-fit statistics		
Number of observations	4168	
R^2	0.3289	
AIC	3907.802	
BIC	4013.227	

*Significant at $p < 0.05$ **Significant at $p < 0.01$ ***Significant at $p < 0.001$

Appendix N: Steps of anchoring based on the DCE_{VAS} approach of the health state valuation study

Appendices-Table 10: Anchoring modelled DCE coefficients to the full health-dead scale using VAS data

		Coefficient (CL)	Step-1	Step-2 - DCE	Rescaled to the PITS	Rescale based on Mapping
Pain/Discomfort						
	1	0.7409	1.0339	0.3399	0.3399	0.2827
	2	0.4681669	0.7612	0.2502	0.2615	0.2081
	3	-0.5473574	-0.2543	-0.0836	-0.0301	-0.0695
	4	-0.6617065	-0.3687	-0.1212	-0.0629	-0.1008
Difficulty eating food /drinking						
	1	0.3468	0.6398	0.2103	0.2103	0.1750
	2	0.1736621	0.4667	0.1534	0.1606	0.1276
	3	-0.2376144	0.0554	0.0182	0.0425	0.0152
	4	-0.2828211	0.0102	0.0034	0.0295	0.0028
Worried						
	1	0.1696	0.4626	0.1521	0.1521	0.1265
	2	0.0860552	0.3791	0.1246	0.1281	0.1037
	3	-0.04853	0.2445	0.0804	0.0894	0.0669
	4	-0.2071149	0.0859	0.0282	0.0439	0.0235
Ability to participate in activities						
	1	0.2188	0.5119	0.1682	0.1682	0.1400
	2	0.0538882	0.3469	0.1140	0.1209	0.0949
	3	-0.1278044	0.1652	0.0543	0.0687	0.0452
	4	-0.1449	0.1481	0.0487	0.0638	0.0405
Appearance						
	1	0.1020	0.3951	0.1299	0.1299	0.1080
	2	0.1010371	0.3941	0.1295	0.1296	0.1078
	3	-0.0343988	0.2586	0.0850	0.0907	0.0707
	4	-0.1686523	0.1244	0.0409	0.0521	0.0340
HS(11111)		1.5781	3.0423	1.0000	1.0000	1.0000
HS(44444)		-1.4652	0.0000	0.0000	0.1264	0.1681
HS(44444)/5		-0.2930				

** Rescaled to the PITS 0.1264 **Rescaled based on Mapping (regression coefficient 0.8319)*

The rescaling steps are based on the valuing the Child Health Utility 9D health states to develop a new adolescent specific scoring algorithm- Personnel communication with authors (1,2).

Model 1 specified in the Table 6.3 was effects coded and the base level was recovered. To rescale it based on PITS heath state, effect coded coefficients in the DCE latent scale were rescaled based on the adjusted mean VAS score for the worst health state 0.1264 **(Table 6.4)**. The calculated utility values for four health states (22211, 33341, 44431 and 44444) in the DCE latent scale were 0.8263, 0.2610, 0.0946 and 0.0000, respectively. To map VAS onto DCE, these calculated utility values were regressed onto the adjusted VAS scores shown in **Table 6.4**, as outlined in Equation 4. The estimated coefficient was 0.8319.

```
. gen dVAS=1-vas

. gen dDCE=1-dce

. reg dVAS dDCE, nocons
```

Source	SS	df	MS	Number of obs	=	4
				F(1, 3)	=	225.88
Model	1.65825911	1	1.65825911	Prob > F	=	0.0006
Residual	.02202411	3	.00734137	R-squared	=	0.9869
				Adj R-squared	=	0.9825
Total	1.68028322	4	.420070804	Root MSE	=	.08568

dVAS	Coef.	Std. Err.	t	P>|t|	[95% Conf.	Interval]
dDCE	.8319031	.0553522	15.03	0.001	.6557476	1.008059

References for the appendices

1) Ratcliffe J, Huynh E, Chen G, et al. Valuing the Child Health Utility 9D: Using profile case best worst scaling methods to develop a new adolescent specific scoring algorithm. *Soc Sci Med* 2016;157:48-59. doi: 10.1016/j.socscimed.2016.03.042

2) Ratcliffe J, Chen G, Stevens K, et al. Valuing Child Health Utility 9D Health States with Young Adults: Insights from a Time Trade Off Study. *Applied Health Economics and Health Policy* 2015;13(5):485-92. doi: 10.1007/s40258-015-0184-3