University Library

A gateway to Melbourne's research publications

Minerva Access is the Institutional Repository of The University of Melbourne

Author/s:
Christian, Bradley

Title:
Caries Risk Assessment Tools: The Evidence

Date:
2020

Persistent Link:
https://hdl.handle.net/11343/265864

Terms and Conditions:
Terms and Conditions: Copyright in works deposited in Minerva Access is retained by the copyright owner. The work may not be altered without permission from the copyright owner. Readers may only download, print and save electronic copies of whole works for their own personal non-commercial use. Any use that exceeds these limits requires permission from the copyright owner. Attribution is essential when quoting or paraphrasing from these works.

Caries Risk Assessment Tools: The Evidence

Bradley Christian

ORCID identifier 0000-0001-5072-3512

Doctor of Philosophy

September 2020

The University of Melbourne

Faculty of Medicine, Dentistry and Health Sciences

Melbourne School of Population and Global Health

Submitted in total fulfilment for the degree of Doctor of Philosophy

Abstract

Background

Various caries risk assessment tools (CRATs) are currently used in the management of early childhood caries. The evidence to inform the selection of a specific tool is unclear, as CRATs differ in their content and development methodology, mostly due to the absence of a standard reporting and testing guideline. Risk-based and outcomes focused (the shift from volume to value) models of caries management have been gaining popularity in recent times as they attempt to prevent and manage the disease itself rather than its sequela. Risk-based models of oral health care attempt to stratify populations, with those most at-risk prioritised for care, while concurrently reducing unnecessary care for low-risk people. This becomes an important consideration when allocating resources in limited resource settings such as public dental services. If new models of caries management depend on an accurate and precise risk assignment, then evidence is needed to inform each step of the development and selection of CRATs. Without this essential body of work, risk-based models of caries management run the risk of not delivering the intended positive outcomes, for either the patient or the health care organisation. The aim of this PhD was to generate new knowledge on caries risk assessment tools for young children, to demonstrate their utility in models of caries management.

Methods

The philosophical underpinnings of my PhD research were informed by the value-based health care (VBHC) agenda. The theoretical foundations, including the intent, motivation and expectations for the research were guided by two socio-ecological conceptual frameworks – National Institute for Health and Clinical Excellence's (NICE) conceptual framework for public health and the Fisher-Owens conceptual model of influences on child oral health. The PhD research was conducted as three separate research strands: Strand 1. The systematic review – provided evidence for the current state of knowledge including knowledge gaps for CRATs for young

children, relating to content, development process, and testing methods, risk categorisation and use in practice; Strand 2. The calibration study – provided evidence for examiner inter- and intra-examiner reliability for the VicGen data and also generated new knowledge related to the process for calibrating clinical examiners in caries diagnosis when conducting research with young children; and Strand 3: The CRAT testing – provided the evidence, based on an assessment of the reliability and validity required to inform the selection of a CRAT (CAMBRA CRAT used as the case study) for young children in the State of Victoria.

Results

The CRATs reported in the literature for use among children less than 6 years were: Caries Management By Risk Assessment (CAMBRA), Cariogram, National University of Singapore CRAT (NUS-CRAT), MySmileBuddy, Dundee Caries Risk Assessment Model, University of North Carolina Risk Assessment Models, University of Michigan pediatric dental clinic caries risk assessment sheet and American Academy of Pediatric Dentistry (AAPD) CRAT. For examiner reliability in the calibration study, Kappa scores for inter-examiner reliability ranged from 0.56 to 0.70 (moderate to good). In the in vivo community examination, using ICDAS caries codes only, weighted Kappa scores for inter-examiner reliability ranged from 0.50 to 0.64 (moderate to good). In the CRAT testing study, good reliability (inter and intra) was observed when using the CAMBRA CRAT. For the classic CAMBRA three-level risk categorisation (low, moderate and high), at 36 and 48 months of age, respectively, the area under the receiver operating curve was close to 0.50, which is indicative of a low ability for the CRAT to predict future caries development.

Conclusion

To conclude, this PhD study identified a major knowledge gap in the current state of evidence on the subject of caries risk assessment tools (CRATs); which was the lack of methodologically sound studies to provide the evidence to inform decision making on tool selection, for use in practice. The PhD study addressed this knowledge gap through methodologically rigorous research to demonstrate the

process required to generate the evidence to inform the selection of a CRAT. The CAMBRA CRAT which was used as the case study in the PhD research was shown to be ineffective in predicting a caries outcome among young Victorian children.

Declaration

This is to certify that:

I. The thesis comprises only my original work towards the PhD.

II. Due acknowledgement has been made in the text to all other materials used.

III. The thesis is less than 100,000 words in length, exclusive of tables, maps, bibliographies and appendices.

Date: 20th September 2020

Signed:

Preface

In this section the following information is provided: the contributions of all persons involved in any multi-authored publications, the publication status of my chapters in article format and an acknowledgement of all sources of funding.

This PhD included three multi-authored publications. I am the first author on all of these publications and my co-authors have signed the 'declaration thesis with publication' form acknowledging that I contributed more than 50% of the work to these three publications. These co-author forms were submitted with this thesis.

- Paper 1: **Christian B**, Armstrong R, Calache H, Carpenter L, Gibbs L & Gussy M (2018). A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. International Journal of Paediatric Dentistry, 29(2): 106-116. Contributions: Conceived the idea: BC; Designed the methods: BC and RA; Literature search: BC; Paper screening, data extraction and quality assessments: BC, RA and LC; Wrote the paper: BC, RA, HC, LC and LG and MG.
- Paper 2: **Christian B**, Amezdroz A, Calache H, Gussy M, Sore R, Waters E (2017). Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical. Community Dental Health. 34(4):248-253.

  Contributions: Conceived the idea: BC and EW; Designed the methods: BC, HC, RS; Conducted the study: BC and EA; Wrote the paper: BC, EA, HC, MG and EW.
- Paper 3: **Christian B**, Calache H, Adams G, Hall M, Dashper S, Gibbs L and Gussy M (2020). A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children. Journal of Dentistry. 95: 103324.

Contributions: Conceived the idea: BC; Designed the methods: BC, HC and GA; Conducted the study: BC; Data analysis: BC; Wrote the paper: BC, HC, GA, MH, SD, LG and MG.

This thesis includes three chapters that are presented in the article format and all three have been published. Details are presented above.

For this PhD, I was awarded a Professional and Practice-based Australian Postgraduate Award scholarship by the Faculty of Medicine, Dentistry and Health Sciences, University of Melbourne.

Acknowledgements

A PhD is a journey and like an epic saga for many. Along the way this journey was influenced and supported by a number of people who kept me motivated, supported and inspired to see the journey to its end. This section is for them.

First, I would like to acknowledge the participants in my research for their valuable time and for their patience with the research process. My PhD would definitely have not been be possible without their participation and commitment to the project.

Next, I would like to acknowledge Professor Elizabeth Waters who not only encouraged me to commence this PhD journey but also supported my application for a University sponsored Australian permanent residency. You gave me the platform to launch my career and life in Australia, and I will forever be indebted to you for this gesture.

The superstars of my PhD – my supervisory and advisory teams, and my advisory chair. My supervisors: Professor Lisa Gibbs, Professor Hanny Calache and Mr. Geoff Adams – Your cool, calm, reassuring and understanding approach to supervision made it possible for me to get where I am in this PhD. There have been a number of ups, downs and life events (two kids, moved towns, changed jobs, health issues etc) in the past five and a half years; and without your support and encouragement I would not have made it to the finish line! I doubt anyone could have a better supervisor team! My advisory team: Adjunct Professor Martin Hall, Professor Mark Gussy and Professor Stuart Dashper. Your constant support, encouragement, enthusiasm and academic discussion as content and professional experts on my PhD, kept bringing me back to why I undertook the PhD and provided the impetus to keep going. I would also like to acknowledge Professor Dallas

English, who as advisory committee chair kept us all on track and who also provided advice on methodological aspects of the PhD research.

To Professor Wendell Evans, thank you for stimulating my interest and building my foundation in cariology. The learnings through my masters' program with you and our ongoing discussions over the years are now paying dividends. We will continue the effort to ensure that cariology is the recognised specialisation.

I would also like to acknowledge my colleagues at the Jack Brockhoff Child Health and Wellbeing Program, who made this journey all the more manageable and fun. Particularly, Dana Young, Alana Pirrone, Emily Amezdroz, Hannah Morrice, Rebecca Armstrong, Karen Block, Elise Davis and Lauren Carpenter.

I would also like to acknowledge a number of other colleagues who provided advice, support and encouragement along the way: Associate Professor Rachel Martin, Professor Anthony Blinkhorn, Associate Professor Ajesh George, Dr. Amit Chattopadhyay and Professor Amanda Kenny

My fellow PhD students (past and present): Dr. Lauren Carpenter, Dr. Rachel Boak, Dr. Utsana Tonmukayakul, Dr. Ron Knevel and Dr. Jacki Goode – by completing your PhDs you showed me that there was light at the end of the tunnel! Thank you for all the advice and support provided!!

I would like to acknowledge the support provided by the Centre for Health Equity and the Child and Community Wellbeing Unit in the form of an open access publication grant, professional development award and travel scholarships. You made the PhD experience all the more rewarding.

Last and most important, I would like to thank my family who kept me sane and sacrificed a lot of their family time so that I could see this PhD through to completion. My two beautiful daughters – Catherine and Isabella, while you both did manage to derail my PhD for several months (more like 12+ months), having you kept me grounded and gave me the motivation to get this PhD finished asap. To Rahila, you truly are the wind beneath my wings! Thank you! I will make up for all those lost weekends!!

Table of Contents

Abstract ..... **i**

Declaration ..... **iv**

Preface ..... **v**

Acknowledgements ..... **vii**

List of Figures ..... **xiii**

List of Tables ..... **xiv**

Chapter 1: Introduction **1**

1.1 Problem Statement 1

1.2 Research Context 4

1.3 Research Aim and Objectives 5

1.4 Research Strands 6

1.5 Overview of PhD Chapters 7

Chapter 2: Background **10**

2.1 Dental Caries 12

2.1.1 What is dental caries? 12

2.1.2 Early childhood caries 15

2.1.3 What causes early childhood caries? 17

2.1.4 Burden of early childhood caries 18

2.2 Models of Care 24

2.2.1 Traditional model of oral health care 24

2.2.2 New models of oral health care 25

2.2.3 The North Richmond Model of oral health care 26

2.3 Caries Risk Assessments 29

2.3.1 Risk and risk assessments 29

2.3.2 Caries risk assessment tools 29

2.3.3 Evidence for current ECC risk assessment tools 30

2.4 Summary 30

Chapter 3: Conceptual Frameworks **32**

3.1 Value in Health Care 32

3.1.1 What is value? 33

3.1.2 The Value-Based Health Care (VBHC) Agenda 33

3.1.3 Why is value important in health care? 36

3.1.4 My PhD and value-based health care 38

3.2 National Institute for Health and Clinical Excellence's (NICE) Conceptual Framework for Public Health 39
3.3 Fisher-Owens Conceptual Model of Influences on Child Oral Health 43
Chapter 4: Research Design 47
4.1 Research Strand 1, Chapter 5: The systematic review 47
4.1.1 Method 47
4.2 The Experimental PhD Research Strands 2 and 3 52
4.2.1 The VicGeneration (VicGen) study: An Australian oral health birth cohort 52
4.2.2 Why use the VicGen cohort for the experimental strands of the PhD? 53
4.3 Research Strand 2, Chapter 6: The calibration study 54
4.3.1 Method 54
4.4 Research Strand 3, Chapter 7: The CRAT testing 58
4.4.1 Method 58
4.5 Sequential Stages of the PhD 63
Chapter 5: Research Strand 1: Systematic Review 65
5.1 Preface 65
5.2 The Publication 65
5.2.1 Abstract 65
5.2.2 Introduction 67
5.2.3 Materials and methods 70
5.2.4 Results 73
5.2.5 Discussion 76
5.2.6 Conclusion 82
5.3 Chapter 5 Appendices 90
5.3.1 Appendix 1. COSMIN measurement properties and domains 90
5.3.2 Appendix 2. Quality criteria for measurement properties 91
5.3.3 Appendix 3. Criteria for overall strength of evidence for a particular CRAT 92
5.3.4 Appendix 4. Sensitivity, specificity, positive predictive values and negative predictive values for current CRA tools for children, by study 93
5.3.5 Appendix 5. Content for CRA tools identified in this review 94
Chapter 6: Research Strand 2: The Calibration Study 96
6.1 Preface 96
6.2 The Publication 96
6.2.1 Abstract 96
6.2.2 Introduction 98
6.2.3 Materials and methods 100

6.2.4 Results 104
6.2.5 Discussion 105
6.2.6 Conclusions 110
Chapter 7: Research Strand 3: CRAT Testing 115
7.1 Preface 115
7.2 The Publication 115
7.2.1 Abstract 115
7.2.2 Introduction 117
7.2.3 Materials and methods 119
7.2.4 Results 124
7.2.5 Discussion 125
7.2.6 Conclusions 132
7.3 Chapter 7 Appendices 137
7.3.1 Appendix 1. COSMIN measurement property domains 137
7.3.2 Appendix 2. Estimates of criterion validity, showing the area under the curve for the CAMBRA CRATs ability to predict caries development at child age 36 (2A) and 48 months (2B) by risk status (low, moderate or high). 137
7.3.3 Appendix 3. Image, the CAMBRA caries risk assessment form in REDCap 138
Chapter 8: Discussion 139
8.1 General Discussion 139
8.2 Contributions to Knowledge 144
8.2.1 Contribution to oral health practice 144
8.2.2 Contribution to research method 145
8.3 Strengths and Limitations 147
Chapter 9: Conclusions, Implications and Recommendations 149
9.1 Conclusions 149
9.2 Implications for Practice and Policy 149
9.3 Recommendations 151
References 153
PhD Appendices 171
Appendix 1. Background published paper 1 171
Appendix 2. Background published paper 2 177
Appendix 3. Research Strand 1, published paper: Systematic Review 185
Appendix 4. Research Strand 2, published paper: Calibration Study 196
Appendix 5. Research Strand 3, published paper: CRAT Testing 202

List of Figures

Figure 1. PhD research strands

Figure 2. The Keyes triad and the classic epidemiologic triad

Figure 3. The ECC burden pyramid

Figure 4A. Prevalence trends for the 30 leading causes of global disease burden with mean percentage change of counts, and all-age and age-standardised rates.

Figure 4B. Incidence trends for the 30 leading causes of global disease burden with mean percentage change of counts, and all-age and age-standardised rates.

Figure 5. Economic burden of oral diseases

Figure 6. The North Richmond model of oral health care

Figure 7. The VBHC Agenda

Figure 8. NICE conceptual framework for public health

Figure 9. The Fisher-Owens conceptual model of influences on child oral health

Figure 10. Sequential representation of the PhD stages

Figure 11. PRISMA flow diagram

List of Tables

Table 1. Inclusion and exclusion criteria

Table 2. Characteristics of included studies

Table 3. Methodological quality of each study by measurement property and caries risk assessment tool

Table 4. Quality of the measurement properties by caries risk assessment tool and study

Table 5. Overall strength of evidence for each caries risk assessment tool, by measurement property.

Table 6. ICDAS Codes

Table 7. Estimates (95% CI) for inter- (at time 1) and intra-examiner reliability

Table 8. Stuart-Maxwell test for marginal homogeneity to assess systematic differences in the in vivo assessment of caries lesions, both between and within examiners

Table 9. Marginal distribution of ICDAS caries codes by examiner from the in vivo community-based dental examination

Table 10. Cross tabulation (marginal frequencies) of caries diagnosis (ICDAS codes) using in vivo visual examinations and intra-oral photographs of the same tooth surfaces for one examiner

Table 11. Distribution of 22 (of the 46) tooth surfaces that were diagnosed as sound (ICDAS code 00) in the in vivo examination but carious when using photographs in Table 5, by tooth location and surface

Table 12. CRAT caries indicators/factors and the corresponding VicGen questions

Table 13. Weighted Kappa scores (95% Confidence Intervals) for reliability and the Stuart-Maxwell test (*P*=) for marginal homogeneity in risk categorisation using CAMBRA

Table 14. Criterion validity of the dichotomised (high-risk vs no-risk) caries risk assessment at 18 months of age, in predicting caries development at 36 and 48 months of age. Responsiveness of the CRAT in identifying children with caries at 48 months who were disease free at 36 months.

Chapter 1: Introduction

1.1 Problem Statement

Dental caries continues to be a major public health problem and data from the Global Burden of Disease studies showed that dental caries continues to be at the top, for both disease prevalence and incidence (1). Dental caries in children younger than 6 years of age is called early childhood caries (ECC). A recent study estimated that globally, the mean caries prevalence among 1-, 2-, 3-, 4- and 5-year-olds were 17%, 36%, 43%, 55% and 63%, respectively (2) Caries prevalence estimates in the deciduous dentition for Australian five-year-old children were 34.3% (3) and among five-year-old Victorian children were estimated at 60% (unpublished data from the Victoria Generation birth cohort study). The primary reason for this difference is due to the definition for dental caries used across the two reports. The national study (3) defined caries as the cavitated stage of the disease process, whereas in the Victorian study the earliest sign of the caries disease process (white spot lesions) was used; and this broader definition would naturally result in a higher disease prevalence. ECC is of concern for a number of reasons and includes: the age of the child puts them at risk for more serious systemic complications as a result of untreated ECC; the deciduous dentition's role in guiding the eruption of the permanent dentition; the thinner dental hard tissue in the deciduous dentition makes the tooth more susceptible to demineralisation with relatively fast progression of the disease through the tooth structure; and concern about the hospitalisation of very young children and the use of general anaesthetic to extract teeth for a disease that should have been prevented (4-7). Also, the economic cost from oral diseases (predominantly dental caries in children) is very high and creates a significant cost burden on families (8). This economic cost falls broadly under three categories: the direct cost incurred from treatment expenses, an indirect cost due to productivity/time lost from work/school; and an intangible cost resulting from the impact of oral diseases on quality of life (8).

Globally, there is a trend for oral health policies, and programs (including national surveillance programs) to ignore this cohort of children (9); most likely due to the logistical issues associated with accessing children under 6 years of age. Also, young children, under six years of age, are more temperamental, unpredictable and reactive to their environment thus making examinations more difficult to conduct (10). As a result, ECC and its associated burden continues to be very high among this age group and constitutes a major public health problem. For this reason, I decided to focus my PhD on young children.

Traditional models of dental caries management have developed around the surgical management of existing pathology in a tertiary prevention or downstream approach. This approach does little to manage the disease itself and, therefore, has had an insignificant effect on disease prevalence and incidence, the rate of hospitalisation, and the inequitable distribution of dental caries and waiting list times for treatment in public dental services (1, 6, 11). At present, this model still defines the majority of dentistry performed globally, including within Australia. As a result, dental caries continues to be highly prevalent in communities across the world, particularly among children (1). For both providers and consumers, there will be considerable challenges to move from this surgically-based model of care, funded to manage disease symptoms by 'fixing teeth' (an output driven model), to a health promotion, person-centred and risk-based model of care designed to improve oral health outcomes (i.e. manage the disease itself) (12). In this thesis a 'model of care' refers to "a multifaceted concept, which broadly defines the way health services are delivered. It outlines best-practice care and services for a person, population group, or patient cohort as they progress through the stages of a condition, injury, or event" (13).

Contemporary evidence on caries diagnosis, lesion incidence and progression rates, non-invasive primary and secondary preventive strategies, the social determinants of health, and laboratory studies into the biology and chemistry of lesion progression and regression, indicates that timely intervention by

nonsurgical treatment modes could avoid the need for most surgical intervention (including restorations/fillings) (14). Hence, new innovative models of oral healthcare are required to challenge the status quo and improve oral health outcomes.

Risk-based and outcomes-focused (the shift from volume to value) models of caries management have been gaining popularity in recent times as they attempt to prevent and manage the disease itself rather than its sequela (15, 16). Risk-based models of oral health care attempt to stratify populations, with those most at-risk prioritised for care, while concurrently reducing unnecessary care for low-risk people. This becomes an important consideration when allocating resources in limited resource settings such as public dental services. A risk assessment is used to: prioritise access to care for those in greatest need; help identify individual risk factors for dental caries and thus support targeted, effective, and efficient disease management (including to manage waitlist and recall time); help those at risk to better understand contributing factors and their likelihood of developing disease; and provide oral health staff a focus for meaningful engagement with their clients that could help informed decision making and actions to reduce risk.

The distinction between a caries risk assessment method (CRAM) and a caries risk assessment tool (CRAT) is important for this research. A CRAM is any method that uses an individual risk or protective factor, as shown in the literature, to independently predict the development or progression of a caries lesion(s), such as the frequency of consumption of sugary drinks or brushing twice daily with a fluoridated toothpaste (17). In an attempt to increase our ability to predict a caries outcome, various combinations of CRAMs (risk/protective factors) are being developed and tested, and these CRAM combinations are called caries risk assessment tools (CRATs) or systems (the term 'tools' will be used in this paper). Various CRATs are currently used in clinical practice and teaching, and common ones include: Caries Management by Risk Assessment (CAMBRA), American Academy of Pediatric Dentistry CRAT and Cariogram (18-20). However, the

evidence to inform the selection of a specific tool is unclear, as CRATs differ in their content and development methodology, mostly due to the absence of a standard reporting and testing guideline (21).

For any health measurement instrument, be it an index test, clinical rating scale, a patient-reported health outcome measure or a combination (as is the case for CRATs), the key measurement properties are accuracy (validity) and precision (reliability). Validity encompasses domains such as content, construct and criterion validity, and reliability includes an assessment of measurement error (22). The majority of CRATs are made up of a combination of self-reported and clinical health indicators/factors (CRAMs) and as such content validity is a critical measurement property for CRATs (234). However, most research on CRATs and CRAMs have focused only on criterion validity (reported as the sensitivity and specificity) in predicting a caries outcome (17, 21, 24). Without evidence for the quality of all relevant measurement properties for a CRAT, making an informed decision on tool selection is impossible.

If new models of ECC management depend on an accurate and precise risk assignment, then evidence is needed to inform each step of the development and selection of CRATs. Without this essential body of work, risk-based models of ECC management run the risk of not delivering the intended positive outcomes, for either the child or the health care organisation.

1.2 Research Context

The research described in this thesis is part of the research portfolio of the VicGeneration birth cohort study on which I am an investigator.

The VicGeneration (VicGen) study is an oral health birth cohort study which began in 2008 to gain an understanding of the multifactorial nature of early childhood caries (ECC) development (25). This study included seven waves of data collection at child ages 1, 6, 12, 18, 36, 48 and 60 months (25). As the main aim of VicGen was to study caries development, the intent was to recruit a cohort

that was broadly representative of the state of Victoria but with a higher risk of caries development. Hence, recruitment focused on socioeconomic disadvantage, cultural diversity and a spread of participants from metropolitan, regional and rural locations (25). The recruited families with newborns were from selected rural, regional and metropolitan Local Government Areas (LGAs) in Victoria, Australia. The selected LGAs were not receiving specific oral health promotion programs (25).

At the time of embarking on this PhD research, public dental services in Victoria were exploring new models of oral health care, particularly in response to the Victorian Auditor General's scathing report on access to public dental services in Victoria (26). Risk-based management of clients in the public dental system was gaining increasing attention as a structured method to: identify those at highest risk of disease, guide treatment decisions, guide discussions with patients (including parents/caregivers) regarding caries risk/protective factors, manage waitlist times and to inform the economics of producing, delivering and using health care (27). Dental Health Services Victoria (DHSV), a State Government entity, administers public dental services in Victoria at the Royal Dental Hospital of Melbourne and by sub-contracting to community health agencies (28). DHSV is driving the transformation of public dental services from the current output-focused (volume) model to one that is structured around improving patient oral health outcomes relative to cost – the value agenda (29). A key operational aspect of DHSV's proposed new model is risk stratification (30). The PhD research is embedded within this context and the new knowledge generated will specifically inform the delivery of public dental services in Victoria.

1.3 Research Aim and Objectives

The aim of this PhD was to generate new knowledge on caries risk assessment tools for young children, to demonstrate their utility in models of oral health care. Specifically, the research objectives were:

- To document and analyse the current state of evidence, including any knowledge gaps, on the quality of current CRATs for young children.
- To assess the quality of a selected CRAT to inform its use among young Victorian children.

1.4 Research Strands

The PhD was structured into three distinct and interrelated research strands (Figure 1) to address the research objectives.

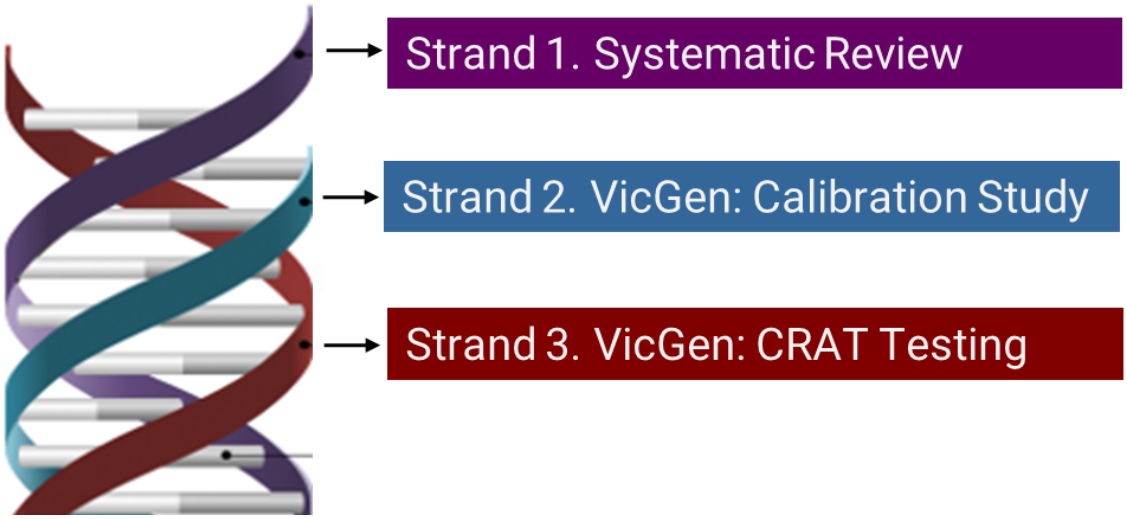


Figure 1. PhD research strands

Strand 1, aligned with the first PhD research objective, provided evidence for the current state of knowledge including knowledge gaps for CRATs for young children, relating to content, development process, and testing methods, risk categorisation and use in practice. Strand 2, aligned with the second PhD research objective, which provided evidence for the quality of the VicGen data to demonstrate it was appropriate to use for Strand 3. It also generated new knowledge related to the process for calibrating clinical examiners in caries diagnosis when conducting research with young children. Strand 3, also aligned with the second PhD research objective, which provided the evidence required to inform the selection of a CRAT for young children in the State of Victoria,

Australia. The method demonstrated in this strand of the PhD could be used as a template to guide research on the topic of CRAT development and testing.

1.5 Overview of PhD Chapters

Chapter 1 introduces the PhD topic by providing a brief description of the study statement of the problem, research context, research aims and objectives, and the research structure. It concludes with a concise overview for each chapter.

Chapter 2 provides the background context to this research. This chapter describes dental caries among young children, its epidemiology and its burden on society. It also critically examines the current model of oral health care, makes a case for why a paradigm shift is required and provides an example of a new model of oral health care in Victoria, which was the stimulus for this PhD research. The content of this chapter was published during the course of the PhD as two papers, which are included in the appendix of this thesis.

Chapter 3 presents the philosophical basis for this PhD research – the ideology of 'value' and the value-based health care agenda. It also discusses the two socio-ecological frameworks underpinning the PhD research – the Fisher-Owens model of child oral health and the National Institute for Health and Clinical Excellence's (NICE) conceptual framework for public health.

Chapter 4 describes the research design for strands 1, 2 and 3 of the PhD research. For strands 2 and 3, the experimental parts of the PhD research, it provides a concise overview of the VicGen study and the rationale for using VicGen data. This chapter also provides overviews of the key methods used in strands 1, 2 and 3 of the PhD research.

Chapter 5 reports on strand 1 of the PhD research, the systematic review, which aimed to answer the question – 'What is the strength of evidence to inform the selection of CRATs for children ages 6 years and less?' The research in this

chapter is presented as a published paper with its own background, methods, results, discussion, conclusion and reference list.

Citation: *Christian B, Armstrong R, Calache H, Carpenter L, Gibbs L & Gussy M (2018). A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. International Journal of Paediatric Dentistry, 29(2): 106-116.*

Chapter 6 reports on strand 2 of the PhD research, the calibration study, which aimed to provide the evidence for the quality of the VicGen data before it could be used in research strand 3. The research in this chapter is presented as a published paper with its own background, methods, results, discussion, conclusion and reference list.

Citation: *Christian B, Amezdroz A, Calache H, Gussy M, Sore R, Waters E (2017). Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical. Community Dental Health. 34(4):248-253.*

Chapter 7 reports on strand 3 of the PhD research, the CRAT testing, which aimed to assess the quality of the CAMBRA CRAT for use among young Victorian children. The research in this chapter is presented as a published paper with its own background, methods, results, discussion, conclusion and reference list.

Citation: *Christian B, Calache H, Adams G, Hall M, Dashper S, Gibbs L and Gussy M (2020). A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children. Journal of Dentistry. 95: 103324.*

Chapter 8, the discussion section of the PhD thesis, provides a thorough examination of the key PhD findings in relation to: its contribution to new knowledge and potential avenues for knowledge translation into practice, the

context of the research and the socio-ecological conceptual frameworks. The strengths and limitations of the PhD research are also presented in this chapter.

Chapter 9 concludes the PhD thesis by highlighting the key findings and how they relate to the PhD research aims and objectives. Recommendations for future research on this topic are also reported.

Chapter 2: Background

This chapter provides the background context to the PhD research and is presented as three sub-sections within this chapter: the disease – dental caries; the setting – models of oral health care; and the intervention – risk assessments using caries risk assessment tools (CRATs). The content of this chapter has been published as two papers that are included in the appendix of this thesis.

Paper 1: Christian B, Hall M, Martin R. A paradigm shift in models of oral health care: An example and a call to action. Family Medicine and Community Health. 2015;3(4):32-37.

Abstract: The consequences of oral disease are wide-ranging and can have a major impact on an individual's and that person's family's quality of life. A range of factors interact to determine a person's oral and general health. Such factors can be biological, social, economic, political, cultural, or environmental, in addition to knowledge, attitudes and behaviours. Traditional models of oral healthcare, however, have generally ignored these factors and instead have focused on the treatment and management of existing pathology (tertiary prevention/downstream approach). This has had no effect on the rate of hospitalization or the inequitable distribution of dental diseases. To reduce the prevalence and severity of oral diseases at the individual and population levels, holistic evidence informed prevention-based health-promoting models of care that focus on upstream determinants of health are required.

Paper 2: Hall M, Christian B. A health-promoting community dental service in Melbourne, Victoria, Australia: protocol for the North Richmond model of oral health care. Australian Journal of Primary Health. 2017;23(5):407-414.

Abstract: Despite the best efforts and commitment of oral health programs, there is no evidence that the current surgical output-based model of oral health care is delivering better oral health outcomes to the community. In fact, Australian evidence indicates the oral health of the community could be getting worse. It is now well-understood that this traditional surgical model of oral health care will

never successfully manage the disease itself. It is proposed that a health-promoting, minimally invasive oral disease management model of care may lead to a sustainable benefit to the oral health status of the individual and community groups. The aim of this paper is to describe such a model of oral health care (MoC) currently being implemented by the North Richmond Community Health Oral Health (NRCH-OH) program in Melbourne, Victoria, Australia; this model may serve as a template for other services to re-orient their healthcare delivery towards health promotion and prevention. The paper describes the guiding principles and theories for the model and also its operational components, which are: pre-engagement while on the waitlist; client engagement at the reception area; the assessment phase; oral health education (high-risk clients only); disease management; and reviews and recall.

I would like to acknowledge my co-authors on these publications – Adjunct Professor Martin Hall and Associate Professor Rachel Martin. My contribution to each paper was at least 50%.

2.1 Dental Caries

2.1.1 What is dental caries?

Dental caries, also referred to as tooth decay is a disease that affects teeth and can cause severe pain (toothache), which if left untreated can lead to tooth loss and more serious systemic complications. It is an ancient disease that is as old as the documented history of humans (31). The term 'caries', a loanword from Latin, means rot or rottenness which has been interpreted in English to mean decay (32). The earliest recorded usage of the word 'caries' is in the 1630s where it was referred to as a 'destructive disease of bone' (32). Pierre Fauchard, popularly referred to as the 'Father of Modern Dentistry', is credited with first using the term 'dental caries' in his seminal book published in 1728 – *Le Chirurgien Dentiste* (The Surgeon Dentist) (33). That Pierre Fauchard would choose the term 'caries', to describe the visible breakdown in tooth structure is in itself very interesting, because knowingly or unknowingly his selection of terminology was accurate and implied the action of bacteria breaking down human tissue. This was at a time when microbiology was very much in its infancy with the first observation of bacteria occurring in 1676 by Antoni van Leeuwenhoek, and the term 'bacterium' being coined in 1828 (34).

In recent times, there have been several definitions for dental caries. In the popular cariology text book (2016 edition), Dental Caries: The Disease and its Clinical Management, dental caries is defined as *"the signs and symptoms – of a localized chemical dissolution of the tooth surface caused by metabolic events taking place in the biofilm (dental plaque) covering the affected area."* (35). In 2009, Longbottom and colleagues with input from key stakeholders published a glossary on the consensus of commonly used terms in cariology and in this document dental caries was defined as *"the localized destruction of susceptible dental hard tissue by acidic by-products from bacterial fermentation of dietary carbohydrates."* (36). In 2018, an expert panel on dental caries met at the International Association of Paediatric Dentistry conference in Bangkok and defined dental caries as *"a biofilm-mediated, sugar-driven, multifactorial, dynamic*

disease that results in the phasic demineralization and remineralization of dental hard tissues, determined by biological, behavioural, and psychosocial factors linked to an individual's environment." (2). Consensus was achieved, in 2019, on a similar definition at a two-day workshop of the European Organisation for Caries Research (ORCA) and the IADR Cariology Research Group (37). At this workshop consensus was achieved on the following definition *"Dental caries is a biofilm-mediated, diet modulated, multifactorial, non-communicable, dynamic disease resulting in net mineral loss of dental hard tissues. It is determined by biological, behavioral, psychosocial, and environmental factors."* (37).

So, it appears that as of 2020, the global cariology community has reached consensus on a definition for dental caries which overall captures the pathogenesis of caries and the associated determinants. However, if we dissect the definition it does have its flaws. **First,** including the term 'multifactorial' in the definition makes the inclusion of any other factor in the definition redundant. Multifactorial is too broad a term to be used in the definition of a disease and implies that there may be a myriad of other factors not captured in the definition which caused caries, when this is not the case. The pathogenesis of dental caries is straightforward – the metabolic activity in the tooth biofilm generates more acid than can be neutralised, which causes a shift in the chemical equilibrium at the tooth-biofilm interface, and if sustained for prolonged periods results in a net tooth mineral loss. While the metabolic activity at the tooth-biofilm interface is ubiquitous, the observed disease patterning is a result of the influences of the world we live in and our place in society, which in turn determines our health-related behaviours. So, in terms of the actual pathological process it is not multifactorial.

Second, is the inclusion of 'diet modulated' in the definition. It is obvious that this term has been added to the definition in recognition of the role that diet plays in dental caries (38). However, diet is a modifiable lifestyle factor and a health-related behaviour, of which there are others that also have an influence on dental caries, and which have generally had a lower visibility than diet in caries research.

These health-related behaviours (including diet) are not part of the direct pathogenic process for dental caries, but their action creates the microenvironments, at the biofilm-tooth interface that are conducive for the caries process. While, diet no doubt has an important influence on the caries outcome, its elevated status in the caries literature most likely stems from the well-known and historic Keyes triad (39).

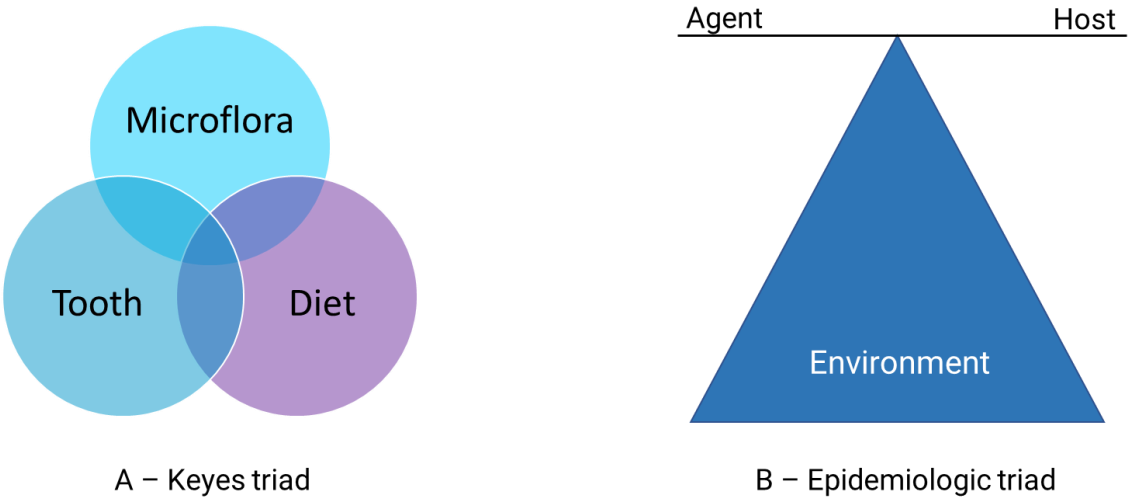


Figure 2. The Keyes triad and the classic epidemiologic triad

It is obvious from Figure 2 that the Keyes triad is a modification of the classic epidemiologic triad (40) with the microflora as the agent, tooth as the host and diet replaced environment. In the epidemiologic triad, the environmental conditions in which the agent and host operate, it either maintains the agent-host equilibrium or tips the balance towards an adverse health outcome (40). In the Keyes triad, diet is used instead of environment and this is incorrect because diet is not an environment. Diet is one of several modifiable lifestyle factors that influences the agent-host environmental conditions and others include the use of fluorides and plaque removal. At the time of Keyes triad in the 1960s, the evidence to support these other factors were just emerging. For example, evidence from the Grand Rapids-Muskegon studies to support water fluoridation was new at the time (41). We know from the evidence that it takes at least 20 years for new evidence to diffuse through the dental profession and be

adopted into practice (42). Hence, 'diet modulated' can either be removed from the definition of dental caries or replaced with 'environment modulated'.

Third, the use of the term 'dynamic disease' in the definition does not provide any added value to understanding/classifying the disease. Activity at micro-environment level (i.e., at the biofilm-tooth interface) is a dynamic and ubiquitous process. However, when the equilibrium in this micro-environment is tipped towards one that promotes a net mineral loss (i.e. caries), it is not dynamic anymore. **Fourth,** the inclusion of the last sentence of the definition "*It is determined by biological, behavioral, psychosocial, and environmental factors"* is probably unnecessary for two reasons – the profession is attempting to define a disease as opposed to a health state and the term 'multifactorial' is already included in the first part of the definition.

Our[†] revised definition for dental caries by taking into consideration the afore discussed issues and keeping with the ethos of the classic epidemiologic triad, would be *"Dental caries is a non-communicable, biofilm-mediated, and microenvironment modulated disease, which causes a net mineral loss of dental hard tissues."* This definition was used for the purposes of this thesis.[†]

2.1.2 Early childhood caries

This PhD research focused on very young children (less than 6 years of age) for the simple reason that oral health research in this age group is very limited. It also built on my research interest, work, experience and expertise on this subject. While the pathogenesis for dental caries is the same in children and adults, early childhood caries (ECC) was defined by Drury et al. (1999) as "*the presence of one or more decayed (non-cavitated or cavitated lesions), missing (due to caries), or filled surfaces, in any primary tooth of a child under age six"* (43). Consensus was again achieved for this definition at the recent ECC expert meeting in Bangkok in November 2019 (2). Other, now obsolete, terms for ECC were rampant caries, nursing caries, baby bottle caries and nursing bottle caries.

[†] Developed in discussion with my PhD supervisor – Professor Hanny Calache.

ECC is of concern for a number of reasons including: the age of the child puts them at risk for more serious systemic complications as a result of untreated ECC, the deciduous dentition's role in the eruption of the permanent dentition, the thinner dental hard tissue in the deciduous dentition makes the tooth more susceptible to demineralisation with relatively fast progression of the disease through the tooth structure and concern about the hospitalisation of very young children and the use of general anaesthetic to extract teeth for a disease that should have been prevented (4-7). At the expert panel meeting on ECC in 2018, it was estimated that, globally, the mean caries prevalence among 1-, 2-, 3-, 4- and 5-year-olds were 17%, 36%, 43%, 55% and 63%, respectively (2). Caries prevalence estimates (not yet published) from the VicGen birth cohort study (used in this PhD) at child ages 12, 18, 36, 48 and 60 months were 5%, 7%, 23%, 40% and 63%, respectively. This high prevalence of disease in such young children and the limited action to improve their oral health outcomes is very concerning.

In Australia, the general lack of action to improve the oral health of very young children is highlighted by the fact that the latest national survey, the National Child Oral Health Study 2012-14, did not collect oral health information on pre-school aged children (3). Whilst some Australian States and Territories do have data for this age group as a result of research studies, the data is seldom representative of all pre-school children for that State/Territory. An example, of state level data that was collected from research is the VicGen birth cohort study which collected data from across the State of Victoria for children from birth to 5 years (26). A search of the Australian literature on ECC revealed that in the past 10 years there were only a handful of studies on dental caries in children less than 6 years of age. The identified studies varied in their methods and aims and as a result are not comparable and do not generate generalisable estimates of the disease (44-49). Hence, at the Australian national level there is much to be done in this space. Particularly, there is the urgent need to generate data to inform programs and policies on ECC.

2.1.3 What causes early childhood caries?

The causation of ECC should be contextualised to the definition of dental caries that was used for this PhD – *"Dental caries is a non-communicable, biofilm-mediated, and microenvironment modulated disease, which causes a net mineral loss of dental hard tissues."* While the general process by which caries occur in teeth is the same across all ages, the factors responsible for the caries process in the deciduous teeth of young children are shown to be different (50, 51). These factors are broadly referred to in the caries literature as risk factors. Burt and colleagues (52), identified the lack of consistency in the use of the term 'risk factor' in the literature on the epidemiology of oral diseases and their proposed definition which used to date is: *"an environmental, behavioral, or biologic factor confirmed by temporal sequence, usually in longitudinal studies, which if present directly increases the probability of a disease occurring, and if absent or removed reduces the probability. Risk factors are part of the causal chain, or expose the host to the causal chain. Once disease occurs, removal of a risk factor may not result in a cure"* (52, p. 1008).

Two key features of this definition are: First, *"…confirmed by temporal sequence.."*, which means that a factor can only be called as a 'risk factor' if it has been shown to be an independent predictor of the outcome in a longitudinal study, i.e. temporal demonstration of the exposure occurring before the outcome. Second, *"…part of the causal chain..."*, a factor can only be called as 'risk factor' if it is directly in the causal pathway for a particular outcome of interest – ECC in this case. According to Burt and colleagues, if a temporal relationship between the exposure and the outcome cannot be established due the study design (for example a cross-sectional study) then such factors that are associated with the outcome should be referred to as 'risk indicators' (52).

Two systematic reviews on risk factors for dental caries in young children (under six years of age) identified a relatively long list of factors as being associated with ECC. Harris and colleagues (2004), identified 106 risk factors for ECC, however, they did not differentiate between risk indicators and factors as per the

definition of Burt et al. (2001) (50, 52). Most of the papers included in their systematic review were cross-sectional in study design, and as such, the factors reported in these included cross-sectional study design papers that should have technically been labelled as risk indicators rather than factors. This systematic review also included studies with longitudinal and case-control study designs, both of which can be used to study temporal relationships between an exposure and an outcome; and in these studies (50), ECC specific risk factors were identified and included: use of sweetened milk in a bottle, breast feeding at 6-12 months, nocturnal breast feeding, enamel hypoplasia, presence of *Streptococcus mutans* at 12-36 months of age, and duration of breast feeding (50). The systematic review by Leong and colleagues (51), documented additional factors that may cause ECC such as: consuming pre-tasted foods at nine months of age, sharing eating utensils more than three times per day at nine months of age, habitually kissing on baby's lips at nine months of age, on demand formula feeding at 12 months of age, maternal and child *S. mutans* levels, and mother's education level (51). These documented risk factors cause caries by disrupting the dynamic equilibrium of the microenvironment conditions at the tooth-biofilm interface in favour of more caries promoting conditions including change in the composition of the microbiome (53).

It is evident from the literature that ECC has relatively specific risk factors which need to be addressed in any intervention, program or policy to reduce the ECC burden on the individual or the community.

2.1.4 Burden of early childhood caries

The global prevalence of ECC is very high and includes high income countries (2). It appears that the global reduction in caries estimates, observed over the past 20 to 30 years, has not occurred for early childhood caries and will need further investigation.

ECC can cause toothache, tooth loss, lost school days and parent's workdays, hospitalisation under general anaesthetic for tooth extraction and significant

cost of treatment, all of which can lead to reduced quality of life for the child and the family (54, 55). The ECC morbidity and mortality pyramid shown in Figure 3, is a comprehensive illustration of the situation with the most common consequences of ECC at the bottom of the pyramid (56).

Figure 3. The ECC burden pyramid

Source: Casamassimo PS, Thikkurissy S, Edelstein BL, Maiorini E. Beyond the dmft: the human and economic cost of early childhood caries. Journal of the American Dental Association. 2009;140(6):650-7.

Toothache and tooth loss, which are probably the most serious consequences from ECC (57), can have serious implications on the chewing process, which in turn can impact on the development of the dental arches, the eruption of future permanent teeth; and as a result can lay the foundation for a lifetime of poor oral health (58). A long term consequence which can be a lifelong fear of dental care

which could be prevented by early nontraumatic interventions. For example, pain or tooth loss can cause the child to focus their chewing on one side of the mouth and this can cause several issues on the side not used in chewing such as: loss of occlusal contact which can lead to malocclusion, and further plaque build-up (increased predisposition to caries for teeth in that area) due to the absence of cleansing that the chewing action produces (59). In addition, premature deciduous tooth loss will cause the adjacent deciduous teeth to move in to fill the space created and as a result there is the loss of space for the underlying permanent teeth to erupt into correct position. These sequalae from premature tooth loss due to ECC can result in developmental anomalies such as reduced dental arch size, overcrowding and malocclusion (60, 61). All of which have the potential to negatively impact on oral health across the lifespan at a great cost to both the individual, families and communities involved.

The 2017 Global Burden of Disease study showed that in the last 30 years, for both disease prevalence and incidence (Figures 4A and 4B), dental caries, in both the permanent and deciduous dentitions, features in the top 30. (1). For disease incidence, in Figure 4B, caries in the deciduous dentition has been consistently in the top 5 for the past 30 years. Unlike other non-communicable diseases such as cardiovascular diseases and cancers, where disease burden is measured by both mortality and morbidity, the oral disease burden is estimated based on morbidity only. The economic burden from oral diseases, based on their morbidity and co-morbidity cost, is very high; and because oral services are mostly delivered in the private sector, the cost burden is directly borne by the patient. Moreover, public dental services face considerable challenges in improving oral health outcomes with limited resources and rising health care costs.

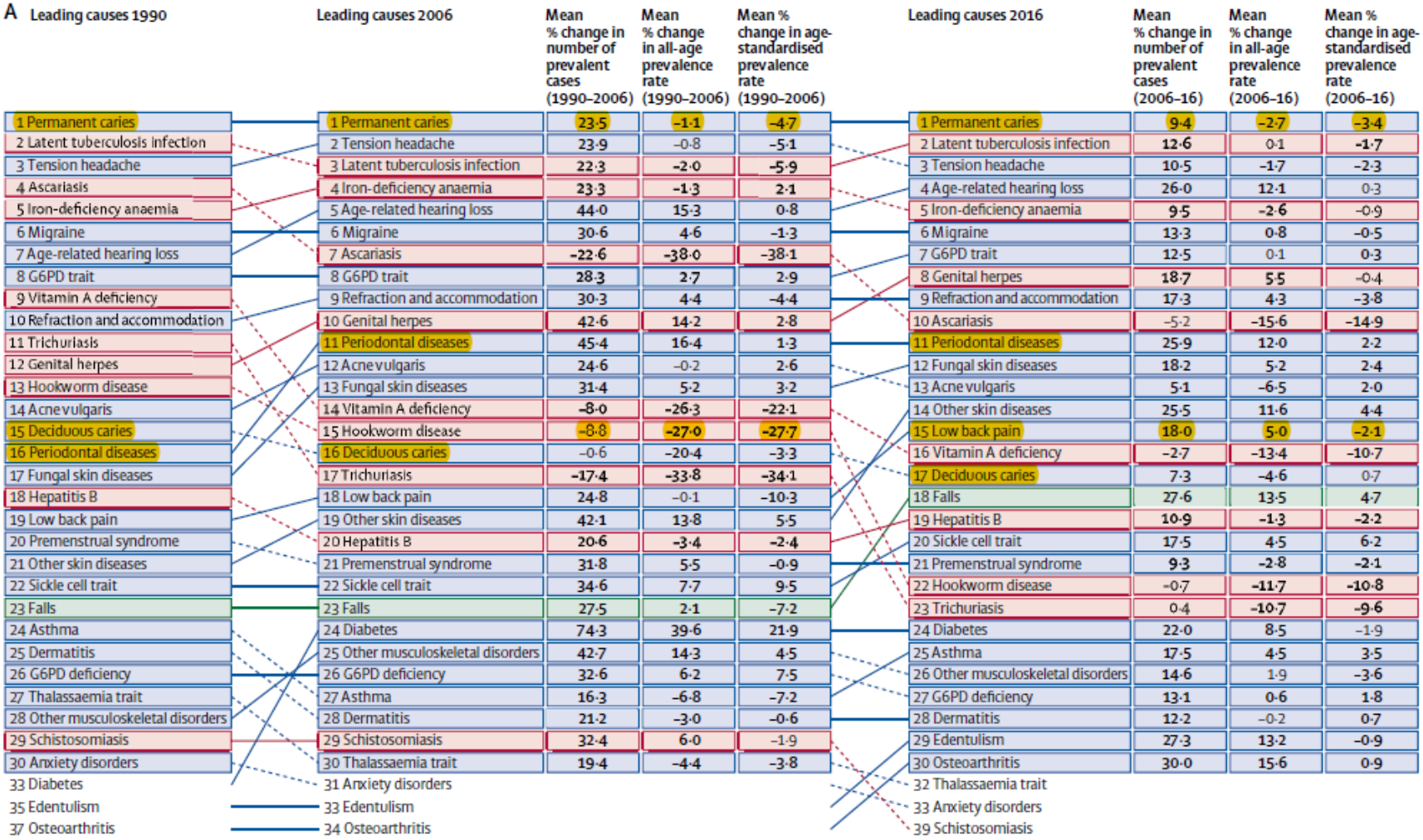


Figure 4A. Prevalence trends for the 30 leading causes of global disease burden with mean percentage change of counts, and all-age and age-standardised rates.

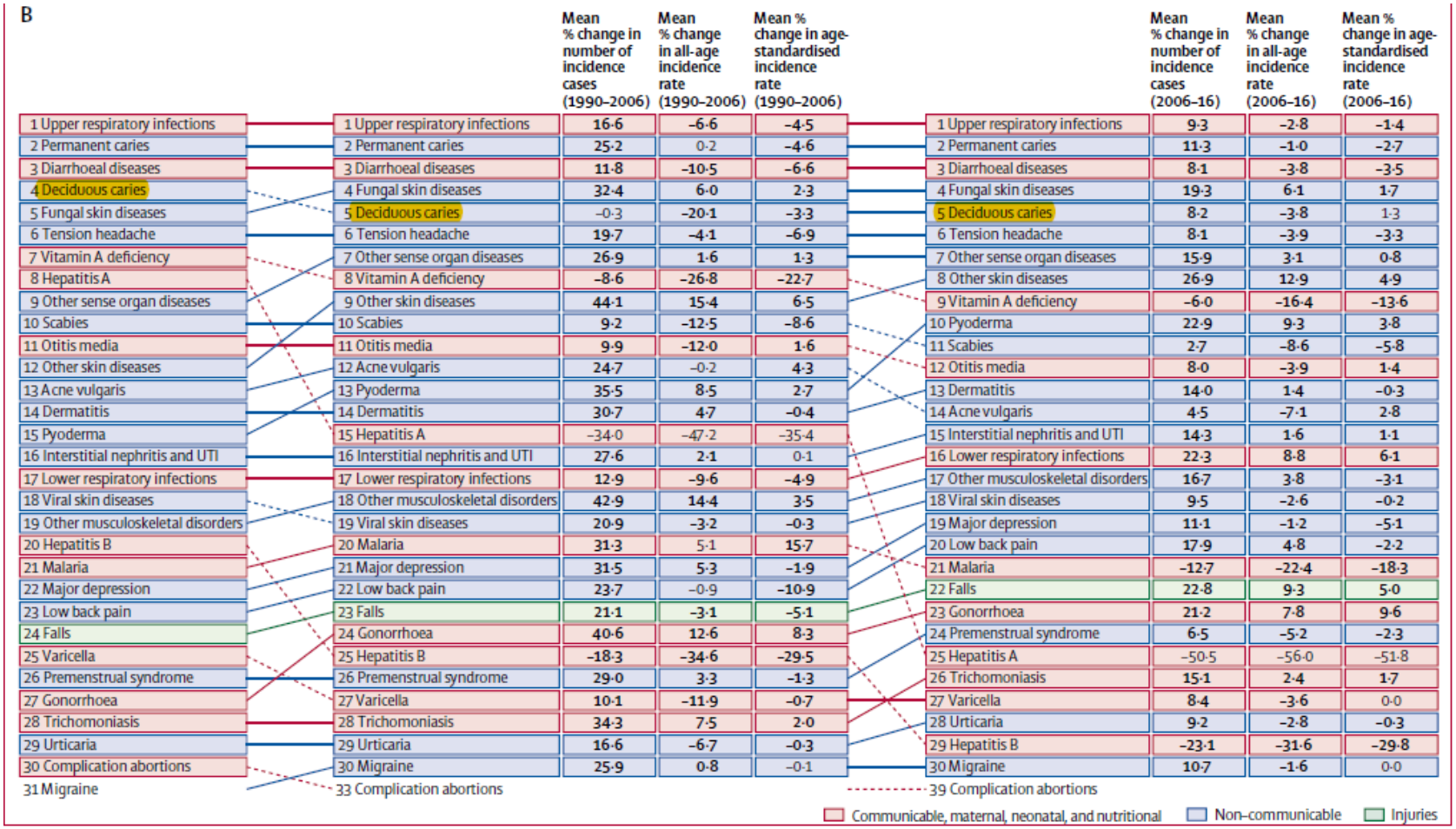


Figure 4B. Incidence trends for the 30 leading causes of global disease burden with mean percentage change of counts, and all-age and age-standardised rates.

Note: Solid lines connecting causes indicate an increase. *Source: GBD 2016 Disease and Injury Incidence and Prevalence Collaborators. Global, regional, and national incidence, prevalence, and years lived with disability for 328 diseases and injuries for 195 countries, 1990-2016: a systematic analysis for the Global Burden of Disease Study 2016. Lancet. 2017;390(10100):1211-59.*

In Figure 5, Listl et al. (2019) shows the direct treatment costs for several non-communicable diseases, including oral diseases across the European Union countries (62). This pattern is similar for Australia where oral diseases account for 7.0% (approximately $8.2 Billion) of health expenditures compared with cardiovascular at 8.9% ($10.4 Billion), cancers at 7.2% ($8.4 Billion) and respiratory diseases at 3.4% ($4.0 Billion) (63).

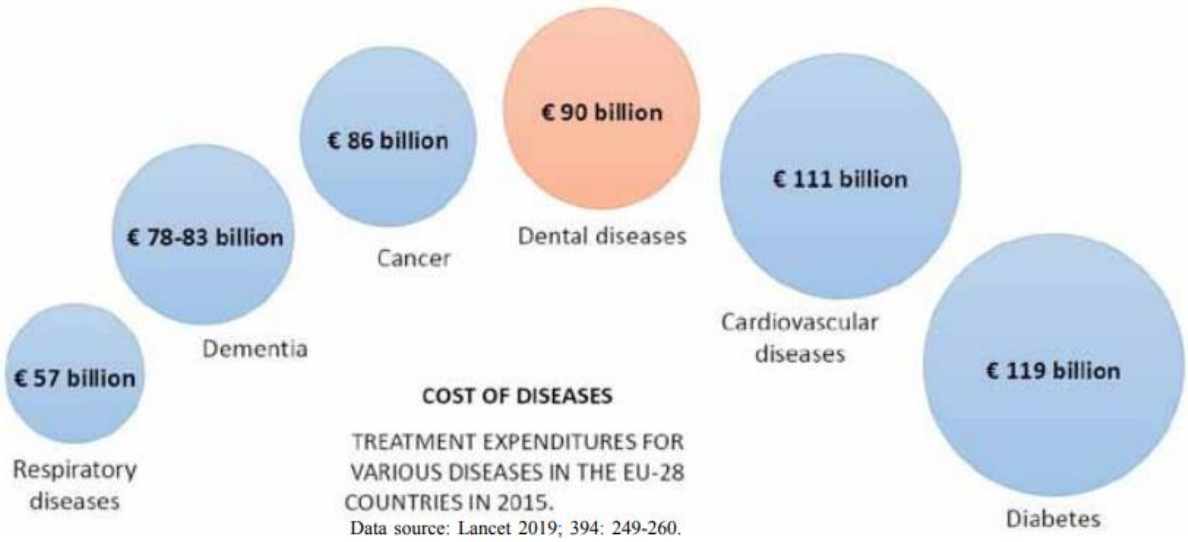


Figure 5. Economic burden of oral diseases

Source: Listl S, Grytten JI, Birch S. What is health economics? Community Dental Health. 2019;36(4):262-74.

It is interesting to note that the cost of treating oral diseases is right up there with the big non-communicable diseases (NCDs) of cancer, cardiovascular disease and diabetes, yet oral diseases do not receive as much attention as these other NCDs, due to the low mortality burden. This is a major flaw in the health system and among decision influencers, globally, because the burden of oral diseases (from morbidity and co-morbidity) can be greatly reduced by relatively simple lifestyle changes and policy level intervention. The money saved can be redirected towards other areas of public health where mortality is the main consequence of the disease.

The economic burden on public dental services should also be highlighted. With increasing health care costs, limited funding allocated to public dental services and the pressure to continue to provide the same volume of services, the current model

of oral health care at public dental services, which is driven by volume (outputs) is not sustainable.

ECC, a preventable disease, continues to be a major public health issue, globally. Urgent action is required at every level to reduce the disease burden and new innovative models of ECC management are required to make a real difference to the ongoing disease burden.

2.2 Models of Care

For this thesis, a 'model of care' (MoC) broadly defines the way health services are delivered and includes best-practice care and services for a person, population group, or patient cohort as they progress through the stages of a condition, injury, or event (13). The particular model of care (MoC) at a health care service can significantly impact on outcomes for both the client accessing the services and the service provider. A model of care's outcomes can be related to domains such as health outcomes, service accessibility, service quality, service economics, organisation policy, workforce structure, and sustainability. Given this range of outcomes, a best-practice model of care will need a multipronged approach to positively address the various domains. Models of care which encompass prevention and seek to address the broader determinants of oral health outcomes will have the most potential to reduce the inequitable social gradient in oral health (9). Such models need to be continually evaluated, evolving, adapting to circumstances and new evidence, and be able to operate in the various domains, stated previously. Evaluations of models of care should be ongoing, as there is often very limited evidence on which to make decisions, and as such policy and economics heavily influence the direction of new approaches.

2.2.1 Traditional model of oral health care

A range of factors interact to determine a person's oral and general health. Such factors can be biological, social, economic, political, cultural, or environmental, in addition to knowledge, attitudes, and behaviours (64, 65). Traditional models of oral health care, however, have generally ignored the above-stated determinants and

instead have focused on the treatment and management of existing pathology (tertiary prevention/downstream approach). This approach has had no effect on the rate of hospitalisation or the inequitable distribution of dental diseases (6, 66-68). An example in the oral health sphere is the conventional practice for caries management, where a reliance on surgical intervention (drilling, filling and extractions) is informed more by opinion derived from subjective experience among practicing clinicians than by current evidence on the caries process (69). Australian data shows that restorative treatment continues to be common practice for managing caries including for non-cavitated lesions (70). Nadanovsky and Sheiham (67) showed that dental services explained only about 3% of the differences in the change in 12-year-old caries levels across several countries. Socioeconomic factors had the largest impact independent of the use of fluoridated toothpaste. Thus, with the traditional model of oral health care, the high prevalence of oral diseases and the associated inequalities in health will continue to be prevalent and may even increase.

2.2.2 New models of oral health care

In contemporary times, better understanding of the caries disease process, changing service needs, workforce deficiencies, the move to an outcome-based (value) approach and the continuing high disease burden, have created an urgent need for new approaches to both the delivery and the types of oral health care provided in the community. For example, this better understanding has renewed interest in the use of Silver Diamine Fluoride (SDF) as a high-value, minimally invasive treatment to manage caries lesions (71-73). SDF arrests the progress of active caries lesions on the tooth, including large open cavities, thus preventing pain and pulpal involvement, providing a window of opportunity to manage the disease process at the person level (74). Also, the current (2020) COVID 19 pandemic has restricted the use of aerosol generating procedures, such as drilling with high speed turbine handpieces, to manage dental caries; and again, there may be the opportunity to use SDF as an effective and minimally invasive caries management treatment.

The urgent need for flexible evidence-informed models of care has been identified by some health services as being important to improving oral health, and the efficiency and effectiveness of service delivery. As a result, innovative, evidence-informed models of care are being developed and tested in response to the broader understanding of the social determinants of health and wellbeing, and the critical importance of prevention and oral health promotion to health outcomes (75, 76). These new models will require efficient, effective, and sustainable integration of population-based health promotion and disease prevention strategies along with, but with a reduced emphasis on, treatment-oriented clinical interventions. They will also need to include strategies for risk assessments, early detection and disease identification and be based on the principles of minimally invasive dentistry.

The concept of minimally invasive dentistry came about in the late 1980s, for several reasons: the dramatic decline in caries rates; the evidence to support fluoridation and widespread use of fluoride toothpaste and other fluoride vehicles; failure of restorations – repeated restoration cycles; and new evidence on the caries process and associated factors – progression, regression, bacterial biofilm (plaque), and saliva (77). The guiding principles of minimally invasive dentistry are: early caries detection and risk assessment; remineralisation of demineralised enamel and dentine; optimal caries preventive measures; minimally invasive operative interventions; and repair rather than replacement of restorations (78). In its earlier form, minimally invasive dentistry was focused very much on the clinical aspects of disease management; however, in contemporary times, in recognition of the social, cultural, economic, and environmental influences on oral health, the concept of minimally invasive dentistry has become enmeshed in health promotion principles (78).

2.2.3 The North Richmond Model of oral health care

This section provides an overview of the model of oral health care at North Richmond Community Health Ltd (NRCH OH MoC) in the suburbs of Melbourne, Australia, from where this PhD topic originated. The background and protocol for

the NRCH OH MoC has been published and the paper is included in the appendix (Appendix 2) of this thesis.

North Richmond Community Health (NRCH) has been providing quality community health services since 1974. The organisation's vision is to be an innovative organisation, responsive to and supportive of community needs and aspirations: open to change and challenges. NRCH currently operates more than fifteen health service programs, administers an annual budget of $8 million and employs a committed staff group of over 100 people (79). The oral health program at NRCH employs 40 staff and operates with a clinical team consisting of oral health coaches who are dental assistants with an extended scope in oral health promotion, dental and oral health therapists, hygienists, prosthetists, dentists and a specialist dentist. The program operates over 3 sites and 2 mobile outreach programs. Infrastructure at the Richmond site includes a modern seven chair clinic and two portable chairs used for outreach.

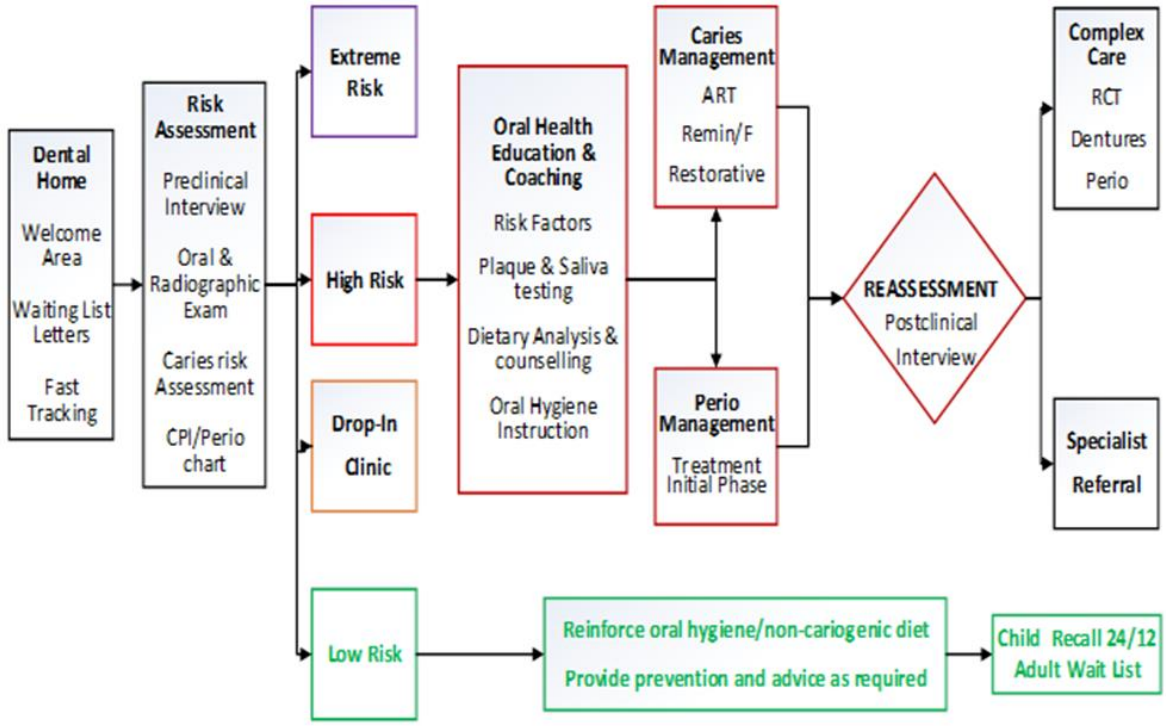


Figure 6. The North Richmond model of oral health care

Source: Hall M, Christian B. A health-promoting community dental service in Melbourne, Victoria, Australia: protocol for the North Richmond model of oral health care. Australian Journal of Primary Health. 2017.

CPI – Community periodontal index; ART – Atraumatic restorative treatment; RCT – Root canal treatment; Remin/F – remineralisation with fluoride; Perio – Periodontal disease.

The development of the NRCH OH MoC (Figure 6) was guided by assumptions conducive to good health. The first assumption was, if clients are empowered to look after their own oral health effectively then this would translate into better oral health outcomes, more successful treatment outcomes and more efficient management of health services. However, changing unhealthy behaviour can be a challenging, time consuming process that requires specialist techniques such as motivation interviewing (80). The second assumption relates to client-identified need, that is, the service providers should cater to the client's need rather than what the clinician determines as being important for the client (client-centred care) (81). It is common practice for dental practitioners to come up with a treatment plan based on their assessment and not focus on the client's identified needs. This plan may not be what the client wants, but due to the perceived clinician-client power relationship, clients agree to take the practitioner's treatment recommendations. The third assumption which relates to the second, is that clients should have input into their oral healthcare plan. This makes them equal partners in the decision process, addresses the power dynamic and gives the client a sense of ownership over their health.

Through the process of developing an evaluation plan for the NRCH OH MoC, the risk-assessment process was identified as being the heart of the model. This PhD study came out of the process to ensure the risk assessment process of the NRCH OH MoC was informed by evidence.

2.3 Caries Risk Assessments

In this section a concise overview of key elements on the topic of caries risk assessments, including assessment tools, is presented. More detail on this topic is given in Chapter 5, the systematic review.

2.3.1 Risk and risk assessments

Risk is defined in epidemiology as the probability that an event will occur following a particular exposure (20). A risk assessment is a method to quantify the risk of future disease using either individual or a combination of risk/protective factors and disease indicators. Caries risk is the probability that the disease will occur in an individual given a particular combination of exposures. More specific definitions for this process are available. For example, Hausen (1997), defines caries risk as *"the probability of an individual developing at least a certain number of caries lesions reaching a given stage of disease progression during a specified time"* (82). The key feature of this definition is a 'certain number of lesions reaching a given stage of disease progression', where the diseased state is determined by the presence of a caries lesion. Given the current evidence on caries aetiology and disease detection methods which capture the lesion development continuum, a more appropriate definition for caries risk informed by the work of Fontana and colleagues (2010) is: "*the probability of an individual developing any sign of caries activity in a specified period in time"* (83).

2.3.2 Caries risk assessment tools

The term 'caries risk assessment tool (CRAT)' refers to any instrument used to determine an individual's risk for developing future disease. These tools are generally a combination of risk and protective factors that together can determine an individual's risk for developing caries (20). Commonly included risk and protective factors for ECC are: diet, child oral health behaviours, fluoride exposure, child dental visits, child saliva properties and clinical indicators such as current caries experience and plaque status (84).

2.3.3 Evidence for current ECC risk assessment tools

At present, there are a variety of CRATs used for ECC management in dentistry. Popular ones include: CAMBRA, American Academy of Pediatric Dentistry Caries Assessment Tool and Cariogram (18, 19, 23). As part of the evidence check for components of the NRCH OH MoC mentioned in section 2.2.4, it was evident that the process and evidence to inform the selection of a risk assessment tool was not clear. A recent review on this topic highlighted the limited evidence on the validity of current CRATs (21). Further ad hoc research on the topic identified other key measurement properties for CRATs including reliability, content validity and the ability of the tool to discriminate between risk levels, for which the evidence was unclear. The literature on this topic also showed that most of the research on CRAT development has been among children ages six and older, with some tools being adapted for use in younger children without being specifically tested among them (21). Hence, the evidence on CRATs for children under six years was very limited.

2.4 Summary

Oral health and healthy behaviour are determined by the complex action and interaction of social, economic, demographic, political, and environmental factors at various levels (individual, family, community, etc.) (85). Traditionally, models of oral health care have focused on downstream drivers of health, without limited success, and as a result, oral diseases continue to be highly prevalent in the population. In addition, most traditional oral health care practices are determined by expert opinion (lowest form of evidence) rather than through scientific enquiry. To reduce the prevalence and severity of oral diseases at the individual and population levels, evidence-informed prevention-based models of care that focus on upstream determinants of health are required.

New minimally invasive models of oral health care use risk-based management for ECC in an attempt to prioritise care for those most at-risk, while at the same time attempting to identify and manage the underlying causes of the disease in a holistic manner. There are numerous CRATs currently in use, however, the lack of

consensus on which of these are the best has made choosing a tool challenging. Existing CRATs differ in their content and quality, which leads to lack of study comparability and the potential for biased results. Information from the literature show that the validity of existing CRATs in predicting future caries is weak. Validity is but one measurement property of a health assessment tool. Another issue identified in the literature is that, most evidence on the performance of CRATs is from cross-sectional studies, which are low in the hierarchy of evidence. Furthermore, most of the evidence generated for CRATs has been in relation to school aged children, i.e., six years and older (21).

Early childhood caries continues to be a major public health problem. If new models of ECC management are based on risk-stratified care, then as a first step we need to: ensure the evidence to support the caries risk assessment process is strong; use evidence-based methods to select a CRA tool; and where required, generate the knowledge to develop an evidence-informed CRA tool. Without this step, risk-based models of ECC management run the risk of not delivering the intended positive outcomes and could instead have the opposite effect and further increase disease burden.

Chapter 3: Conceptual Frameworks

This chapter on methodology describes the philosophy (value-based health care) and conceptual health frameworks that formed the theoretical foundation for my PhD research and within which the research method is embedded. It provides an in-depth examination of the theoretical context (philosophy and framework) of my research, including the background context and rationale for their development; and also, their relevance to my PhD research.

Conceptual frameworks are important because they lay down the theoretical foundations, including the intent, motivation and expectations for the research. They also help illustrate the links between the various components of the PhD research and provide an understanding of the context in which the research is situated. The two conceptual frameworks that steered this PhD were primarily socio-ecological in nature and are well aligned with the focused PhD research topic of oral health as well as the broader public health context within which the research is nested (85, 86). In addition, in the two frameworks that informed this research, the complexities of the various systems that impact on health outcomes are comprehensively presented, in a manner that makes it relatively easy to comprehend. The World Health Organizations Conceptual Framework for Action on the Social Determinants of Health (87), was also considered for this research but the complexity of the conceptualisation, while useful for an in-depth discussion on the determinants of health, was too broad for the scope of this PhD.

3.1 Value in Health Care

At the time of starting my PhD in 2015, the concept of 'value' in health care was gaining increasing attention among leaders of public dental services in the State of Victoria, Australia. Five years later, in 2020, Dental Health Services Victoria (DHSV – the peak body that administers public dental services in Victoria) is well on its way to re-orienting its service model of care towards the value-based health care (VBHC) agenda. A key feature of the VBHC agenda is risk stratified high-value care and my

PhD will generate the evidence to inform this important component of the VBHC agenda. Value and VBHC though, interlinked, are two separate concepts with the former being the ideology that is at the heart of the latter (30). The VBHC agenda is about operationalising the concept of value in health care and is done differently across organisations (88). The philosophical underpinnings of my PhD research were informed by the VBHC efforts by Porter and Teisberg (89, 90), as were DHSV's shift to VBHC.

3.1.1 What is value?

The concept of 'value' one would expect at a supermarket is 'more product at the same or lower price'. In the context of the current oral health delivery system 'more product' can be translated into more or increased volume of 'services provided'. However, with the general ethos of keeping people healthy (that should be embedded at the core of every health practitioner), 'more product' should translate to better health outcomes. Value is not a new concept in health care but a new term for ideology that has guided physicians from time immemorial, such as: "keeping people free of disease", "commits to the best interests of the patient", "the ethics of health care practice and being accountable". The Hippocratic Oath (believed to have been written in 400 BCE), recited by physicians to date, is probably one of the earliest documents of western medicine that has elements of the contemporary term 'value' in its text (91). Even without going into history or evidence, it can be safely assumed that at the core of most people opting for a career in health is the ideology to improve health outcomes. In the context of health care delivery, value has been commonly referred to as the process of achieving the best outcomes at the lowest cost (92, 93). This view of value leaves the term 'outcomes' open to interpretation, which can be outcomes that matter to either the service delivery organisation or the health practitioner or the patient. In a nutshell, 'value' is about outcomes and cost.

3.1.2 The Value-Based Health Care (VBHC) Agenda

As mentioned in the previous section, this PhD research was informed by the value-based health care agenda proposed by Porter and Teisberg (29, 89, 90). Porter (2013) refers to his strategy for implementing VBHC as an 'agenda', because it

provides a clear framework and instruction on re-orienting health care towards value (88). Figure 7 illustrates the key components of the VBHC agenda. This is important to note, because, though the concept of value has been around for a while, there are limited frameworks to support the operationalisation of value (94-96). The delivery of health care services, particularly public/government provided health services, is very complex and influenced by the social, political and economic contexts in which they operate; as well as the training/education and motivations of the individual health practitioners. To re-orient the whole system towards 'value' can be a complicated and lengthy undertaking, requiring action and consensus by stakeholders at every level of the system (30, 76, 88).

Porter and colleagues define value as the health outcomes that matter to the patients relative to the cost of achieving those outcomes (88).

$$Value = \frac{Health\ Outcomes}{Cost}$$

This definition puts very clear parameters around the concept of value. First, the definition puts the focus on outcomes (quality of service provided) and not outputs (quantity). It is well documented that, globally and in Australia, health services focus on delivering quantity (volume/outputs) over quality (value/outcomes) (76, 97). This entrenched health delivery system that focuses on outputs is, as mentioned in the previous section, primarily driven by the current reimbursement system. In addition, in the current health care system 'quality' refers to the process of service delivery (domains such as accessibility, affordability, efficiency, safety and equity) including the patient's experience of the process of receiving care, rather than improving patient health outcomes (76).

Second, Porter's definition of value, places the patient and their outcomes at the centre of the value agenda. Outcomes that matter to the health service provider organisation (e.g. generating income), or the clinician, take the backseat in this definition of value. This does not mean that these outcomes suffer if the system re-orients to focus on patient outcomes. In fact, emerging evidence suggests that

those health services that have adopted the VBHC agenda have seen significant improvements in their performance indicators (98). Finally, cost, the most important driver of the current health care delivery system, is brought into the value equation. Without considering cost and current methods of financing health service delivery, the value agenda will never succeed. This is because the economics of health care drives everything – from the upstream government level action that creates the conditions conducive to health, to the downstream operational models of health care delivery at the service provider and individual patient level.

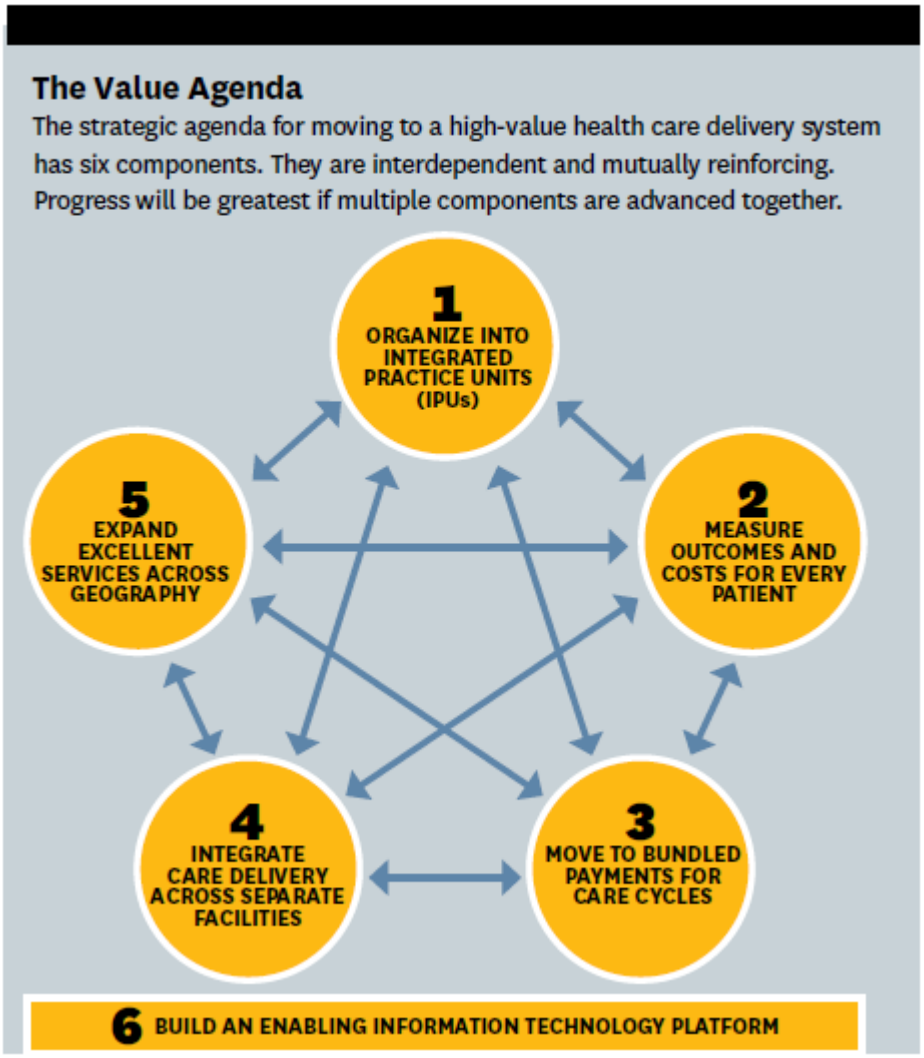


Figure 7. The VBHC Agenda

Source: Porter ME, Lee TH. The strategy that will fix health care. Boston: Harvard Business School, 2013.

3.1.3 Why is value important in health care?

The origins of VBHC – the concept of value in health care arose out of a chain of events that occurred in the United States, with the primary driving force being to contain the increasing cost of health care (99, 100). To increase health care coverage in the United States and reduce the economic burden on households with older and low-income people, the Medicare and Medicaid legislation was enacted in 1965. However, the operational aspects of Medicare and Medicaid were based on the traditional fee for the service payment model (that continues to dominate payment models in the oral health service delivery). As a result of this new legislation, expenditure on health care in the US spiralled out of control and as of 2017 is approximately 18% ($3,492 Billion) of the US Gross Domestic Product (GDP - $19,485 Billion) (101). This trend of increasing health care expenditure is observed across most developed countries. For example, health expenditure in Australia is estimated as being at 10% ($185 Billion) of the GDP (102). As a result, health care expenditure is a topic of continuing debate in the US and in most developed countries and more efficient models of care without compromising quality continue to be explored. From as early as 1984, bundled payment systems have been trialled by private health service providers in the United States, where reimbursements were made for an episode of care and incentivised towards patient outcomes (103). They are known as pay-for-performance models. The effectiveness of these models was recognised in 2008, when the US Centers for Medicare & Medicaid Services (CMS) launched its own version of a pay-for-performance model as a potential transition from the traditional fee-for-service model towards value-based health care – The Evaluation of the Medicare Acute Care Episode (ACE) demonstration (104). Since then value in health care has been gaining global attention and popularity.

There are several reasons why re-orienting the health system towards value is required. First, the concept of value, as defined and discussed by Porter (2010), brings the focus of health services and health care back to the crux of the issue – keeping people healthy (29). This should be the primary measure of quality for any health service provider. It can be confidently stated that, globally, the provision of health care has lost its direction and moved towards increasing the volume of

services delivered and maintaining profit margins (90, 100). This is particularly true for oral health, where historical fee-for-service and output (volume of services) driven models of care continue to thrive, without any consideration for patient health outcomes (15). It is not that all health professionals have lost their direction but the fact that they are operating within an entrenched health system, particularly the payment system, that is incentivised towards volume (95). Hence, a fundamental departure from traditional health care systems is required to ensure successful implementation of the VBHC agenda.

Second, the focus on the patient and their outcomes by making them the centre of care, promotes the integration of various sectors of a presently disjointed system to ensure everyone/every stakeholder shares the same vision/goal – to improve outcomes that matter to the patient (76). This is indeed a key aspect of the VBHC agenda, the integration of care around the patient's particular condition (89). A health system consolidated around a specific agenda, can potentially reduce redundancies in the health service provision process. This integration and unification of services may also help mitigate the widespread variation that currently exists, within and across health service providers, in terms of: payment models, health outcome measures, measures of quality and health system performance, health information systems and interventions (program and policy) to improve health; to name a few (95).

Third, making value the centre piece of the health system enables providers to question or view every service provided from the perspective of value and ask the question – does this service provide value to the patient or population (patient groups with similar needs)? This process triggers a review of services with the aim to eliminate any low-value (high cost and minimal impact of patient health outcomes) services from the health care provider's repertoire of services; which could improve service delivery efficiencies and effectiveness (95).

Fourth, the concept of 'value' provides a reference framework within which stakeholders can engage to re-orient the health system towards better quality as

defined by the VBHC agenda (76). Risk stratified high-value service (lower cost and improved patient health outcomes) provision ensures appropriate resource allocation, which again has implications for improving the efficiencies of service provision (105). All the above stated reasons for re-orienting the health system towards value has the potential to generate significant improvements in overall system efficiencies, which in turn could be associated with economic benefits. Evidence from organisations that have implemented the VBHC agenda demonstrate improvements in patient outcomes and significant economic benefits to the provider (98). However, in terms of actual implementation, the VBHC agenda is still in its infancy and considerably more evidence needs to be generated to demonstrate its usefulness in practice.

3.1.4 My PhD and value-based health care

As mentioned at the start of this chapter, my interest in the topic of VBHC was stimulated through my work in public dental services in Victoria. At the time of embarking on this PhD journey, new models of oral health care, aligned with the VBHC agenda, were being developed and trialled in the State of Victoria. Through my employment with the Oral Health Program at North Richmond Community Health (NRCH) Ltd, I had the opportunity to inform the development of their model of care (16). A key feature of this model was risk-based stratification for caries management. Around this time, Dental Health Services Victoria, in an attempt to re-orient itself away from an output driven system, began to explore the VBHC agenda.

Stratification of patients or populations by risk is a key element of the VBHC agenda (88). Through discussions with my PhD team and the senior leadership at both NRCH and DHSV, we identified the risk assessment component of these new proposed models of care as one of the most crucial components. In order to ensure the risk assessment process was informed by evidence, I searched the literature to get a sense of: the current tools being used to determine risk status; the processes used to develop existing tools; and the quality of evidence to support their selection in practice. Immediately, it was apparent that there was not much in the literature and that there was wide variation between tools in: definitions of risk levels;

definitions of dental caries; the tool content; caries disease prevalence in the population used to test the tool; analytic techniques used, measurement properties used and estimates reported; and study design. This initial scoping exercise showed us a clear knowledge gap and I decided to focus the PhD on investigating and building the evidence on caries risk assessment tools for young children.

All the afore-mentioned reasons to challenge the status quo and re-orient away from an output-based system to a value-based system, holds true for oral health care providers. The application of the VBHC agenda to oral health care is very much in its infancy. In fact, for oral health services, DHSV is a global leader in the implementation of the VBHC agenda (15). Currently, there is very limited published literature on oral health and VBHC, and the publications that do exist are mostly commentaries or opinion pieces on oral health and VBHC (106-110). There is no literature on the outcomes from implementation of the VBHC agenda including evaluations/pilot testing of prototype models of VBHC in the oral health space. Robust evaluations and methodologically sound research will be required to generate the evidence to make the case for a re-orientation of the oral health care system to VBHC. Any attempt to modify/change the current well-entrenched, traditional output-focused oral health care system will be a monumentally complex process; and without scientifically sound peer-reviewed evidence to inform/drive this change, organisations (such as DHSV) run the risk of: (1) Getting siloed in their efforts without local/global recognition or uptake of their learnings; (2) Losing the support from policy makers and decision influencers, who are key to influencing change and creating the economic and political conditions conducive to change; and (3) Failure in their attempt to re-orient towards value.

3.2 National Institute for Health and Clinical Excellence's (NICE) Conceptual Framework for Public Health

The NICE conceptual framework discussed here has a broad public health remit and provides a holistic conceptualisation of the determinants of health outcomes (86). It helped guide the conceptualisation of the PhD research from the public health perspective and provided a broader and deeper insight into the causal mechanisms

of the various determinants of health outcomes. An interesting guiding principle of this framework was that the determinants of health not only cause pathology in the individual person but are also responsible for population level patterns in the pathology, which in turn drives health inequalities. Hence, while most conceptual frameworks and empirical research study the individual and population as separate entities, the NICE framework brings it all together in a continuum, i.e., the individual pathologies and their group patterning have similar determinants.

Another interesting aspect of the NICE framework is that it includes the notion of life course in the conceptual thinking around public health. This makes perfect sense, as health is a continuum in the journey of life, with health gains and losses being fluid throughout the life course. This again is a unique aspect of the NICE conceptual framework, to amalgamate all the key concepts that determine health behaviours and cause health outcomes.

The NICE conceptual framework for public health shown in Figure 8, conceptualises the linkage of the determinants of health to health outcomes using four vectors – population, environmental, organisation and social vectors. The use of the term 'vector' is interesting. In epidemiology, a vector is an agent that is the go-between of a pathogen and the host, e.g. mosquito, the vector for the malaria parasite. The vector is part of the causal chain for malaria and helps explain how the parasite and the host are connected. For effective intervention against malaria, the causal pathway needs to be understood and action taken at all important points along this pathway.

In the context of public health, a vector can be interpreted as the agent that explains the relationship between the determinants of health and the health outcomes. This is an interesting and unique approach to understanding the world we live in and how it relates to human behaviour and health outcomes. While the general relationship between social factors and health is well established, the relationship is not always described in causal terms (111). Vectors help conceptualise this causal relationship. The vectors are not distinct entities, and in reality, the elements

included in these vectors overlap and interact; however, for conceptual purposes on causation they can be considered distinct.

Figure 8. NICE conceptual framework for public health
Source: Kelly MP, Stewart E, Morgan A, et al. A conceptual framework for public health: NICE's emerging approach. Public Health. 2009; 123(1): e14-20.

The 'population vector' is any element that acts at the population level, for example, local, state and federal governments; large corporations; and supra-national formations like the United Nations, European Union and NATO. A very important element of this vector is the economy and its associated aspects such as the Gross Domestic Product as well as market and trade arrangements.

The 'environmental vector' includes all harmful and protective elements that exist in both our macro- and micro-environments, such as: infectious pathogens, asbestos, lead, mercury, radiation and pollution. It also includes elements such as floods, bush fires, climate change and droughts that cannot be controlled by human intervention. This vector is considered to be the traditional area of public health action, since

straight-forward empirical associations may be investigated between the elements in this vector and health outcomes.

The ‘organizational vector’ includes organisations where most human interaction/activity takes place outside of where people live (i.e. their house). It includes education institutions, workplaces such as health care, organisations for social activities such as clubs, and religious organisations. The ‘organizational vector’ is unique in that it acts as the relationship between the population and the environmental vectors. For example, the current COVID-19 situation can be viewed through this concept, where the pandemic caused by the SARS-CoV-2 virus (environmental vector) required governments/supra-national formations such as the World Health Organization (population vector) to implement population level measures to prevent the spread and to reduce morbidity and mortality. The programs and policies (population vector) administered at the population level, provided health care organisations (organizational vector), across the world, with the required supports to prepare for and manage the pandemic situation.

The ‘socio-cultural vector’ includes those elements linked to socio-demographic-economic-cultural circumstances. The commonly studied elements in this vector include income, education, race/ethnic/cultural background, age, sex, social capital and health care card status. It is common practice in research to describe the distribution of health outcomes by these social elements. The social determinants of health and health outcomes including the social gradient in health, as stated previously, are well researched and documented. The socio-cultural vector stimulates thinking beyond the social determinants to the deeper issues underlying social class such as the dynamics of disadvantage, power, conflict and self-interests that occur between and within social classes. These underlying circumstances drive human behaviours, which in turn can cause a particular health outcome. Continuing the example in the previous paragraph regarding the COVID-19 pandemic, all the action (or lack of action) in the three other vectors to manage the pandemic situation, acts via the ‘socio-cultural vector’ to produce the observed individual and population disease patterning.

Risk assessments – the research topic for the PhD, are about predicting the likelihood of developing future disease (early childhood caries). Predictions are made based on our understanding of the factors associated with dental caries. Conceptual frameworks assist in the development of theoretical bases for the causal pathways that lead to the outcome of interest – dental caries in this case. One needs to develop a comprehensive understanding of how the various parts of the system interact with each other, or work in tandem, to produce the outcome. Without this understanding it is impossible to make empirically accurate decisions.

The key question is, why do we want to understand how our world works to produce certain behaviours that in turn cause certain health outcomes? The answer is simple: to keep people healthy. If we know how our world works and where the main touch points are, we can use them to predict what is going to happen or use them as points of intervention to prevent an adverse health outcome. This forms the crux of my PhD research – to develop a deeper understanding of the drivers of oral health outcomes and generate new knowledge on the impact of these drivers on future caries development.

3.3 Fisher-Owens Conceptual Model of Influences on Child Oral Health

The Fisher-Owens conceptual model of influences on child oral health has and continues to be widely referred to in oral health research (85). The framework's popularity is due to the comprehensiveness of the system that it alludes to, yet the simplicity of its portrayal of the determinants of child oral health. Thus, providing a clear, concise and easily understandable frame of reference for epidemiologists, program planners and policy makers to base their work on. Though it has been widely used in research, its utility outside of research to inform health program and policy development is yet to be ascertained.

This framework, shown below in Figure 9, was developed in 2007 out of a need to recognise, stimulate discussion and demonstrate that, influences on oral health outcomes go beyond those that are in the immediate vicinity of the person; which has been the focus on oral health research in the past, such as, position of teeth in

the mouth, saliva flow and composition, diet, oral microbiome and individual oral health behaviours including health seeking behaviour (50, 51). It also arose out of the recognition that the traditional model of oral health care, which is focused on treating existing pathology, has had no effect on the rate of hospitalisation due to dental caries, the inequitable distribution of ECC in the population or reductions in the population estimates of ECC (12). It is now well understood that population level intervention is required to create environments conducive to health, which could support individual action to improve oral health outcomes. The Fisher-Owens framework brings into play two additional levels of influence that are external to the individual person and may not necessarily be within their control – the environments created by their family and the community they live in.

The family-level influences are primarily situated in the realm of the social determinants of health and the community-level influences are at the government/policy level. The Fisher-Owens framework also attempts to stratify the influences by five broad domains of the determinants of health: genetics and biology, social environment, physical environment, health-influencing behaviours, and medical care (112, 113). While such stratification helps illustrate the complexity of the system by placing the multi-level influences within the broad domains of the determinants of health, this process adds additional complexity that has mostly been ignored when using the Fisher-Owens framework; which is probably because it would be empirically challenging to account for these five domains on top of an already complicated multi-level analytic estimation.

The Fisher-Owens conceptual framework was well suited for this research for several reasons: First, the two experimental components of this PhD used data from the VicGeneration (VicGen) study (114). VicGen is an oral health birth cohort study that aimed to gain a deeper understanding of the multifactorial nature of early childhood caries (ECC) development. The theoretical foundation for VicGen, including the theory for causation in the analysis plan, was informed by the Fisher-Owens conceptual framework.

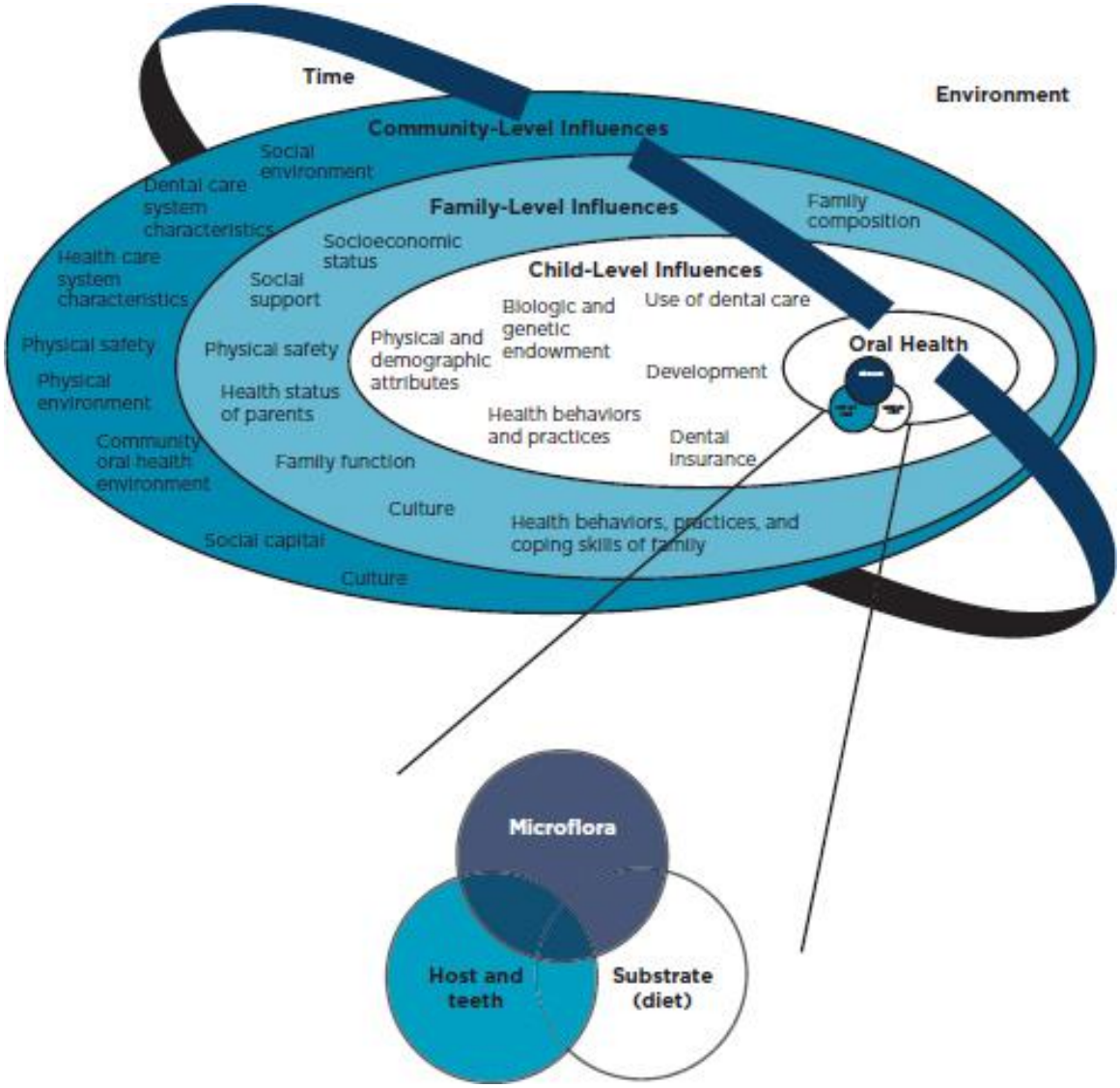


Figure 9. The Fisher-Owens conceptual model of influences on child oral health
Source: Fisher-Owens SA, Gansky SA, Platt LJ, et al. Influences on children's oral health: a conceptual model. Pediatrics. 2007; 120(3): e510-20.

VicGen data included child and caregiver oral health (including saliva microbiology), family socio-demographic-economics; child feeding habits; child and caregivers' oral hygiene-related behaviour; self-reported oral/general health status for child and caregiver; and caregiver's knowledge and attitudes about oral health. Given, that the majority of the PhD research used VicGen data and that VicGen was designed using the Fisher-Owens conceptual model, it made sense that this conceptual framework would be a key influencer of the theoretical foundation for this PhD. Second, the PhD

research was on the topic of risk assessments for dental caries in children, that is, the ability to predict the likelihood of developing future disease (dental caries) based on a current assessment of risk, which is performed by quantifying the current influences on child oral health using various caries risk assessment tools (115). The Fisher-Owens model provided a frame of reference to conceptualise the potential causal mechanisms that could lead to the outcome of dental caries, based on the influences/factors/variables included in the various caries risk assessment tools. In fact, Fisher-Owens states the following in her paper "…., that applying an appropriately complex, yet manageable, model to children's oral health also will result in improvements in predictive power." (85, p. e516). This can bring one to confidently assume that in the development of her conceptual model, Fisher-Owens did envisage its potential use in risk assessments to predict the likelihood of future disease. Third, relates back to the concept of value and the value-based health care (VBHC) agenda discussed earlier in this chapter. Risk stratification is a key component in the implementation of the VBHC agenda. As mentioned in the previous point, the Fisher-Owens model provides a frame of reference to assist in conceptualising the potential causal pathways to a specific outcome, dependent on context.

Although the Fisher-Owens model is very popular in the oral health literature, it can be viewed as over-simplifying complex concepts. In fact, the model in its current form can be viewed as being reductionist in nature and has been used as such in research (116-120), i.e. starting broad with the outermost level and moving inwards to the more specific influences. The model uses the term 'multi-level' to describe the community-, family- and child-level influences. A 'level' can be defined as a position on a scale and as such implies hierarchy in the three levels, whereas in reality this hierarchy may not exist, and it may be that the three levels similarly influence each other. 'Multi-layered' influences on child oral health is probably the more apt term in this context. Regardless of the perceived limitations, the Fisher-Owens model is highly useful in conceptualising and analysing the influences on child oral health.

Chapter 4: Research Design

This chapter provides an overview of the methods used in the three PhD research strands. It presents an overview of and rationale for using data from the VicGeneration (VicGen) birth cohort study. An overview of each research strand and the link between them is also presented.

The systematic review and the two experimental research strands are presented in their respective chapters as published papers which include the detailed methods involved. As this is a thesis by publication, there is unavoidable cross over with the methods reported in the results chapters 5, 6 and 7.

4.1 Research Strand 1, Chapter 5: The systematic review

The purpose of the systematic review was to understand the current state of evidence, including any knowledge gaps, on the quality of current CRATs for young children. It specifically aimed to answer the review question – What is the strength of evidence to inform the selection of CRATs for children ages 6 years and less?

4.1.1 Method

This systematic review was informed by the methods used for a Cochrane review as outlined in the Cochrane Handbook for Systematic Reviews of Interventions (121).

Identification of studies

To achieve adequate breadth of the relevant literature, the following sources were searched: electronic databases, reference lists, existing networks and experts in the field.

MEDLINE was the principal search database for this review. It is the largest subset of PubMed and the primary database for the biomedical and life sciences. Other databases searched included CINAHL and PubMed. Key search terms were

selected in consultation with experts in the field and a University librarian. These keywords were used to generate a Boolean search string (including the truncation function *) which was modified slightly for each database. The base Boolean string was – (caries or dental caries) AND (risk assessment or risk measur*) AND (measur* or tool*). In addition, we also conducted a phrase search for known caries risk assessment tools. The Boolean string for this search was – ("caries management by risk assessment" OR CAMBRA OR cariogram OR "caries management system"). A University of Melbourne, School of Dentistry librarian, familiar with the terminology in this field, helped design and implement the search strategy.

The reference lists of articles selected for inclusion in the review and known cariology literature (including textbooks) were also screened to identify studies that may have been missed in the electronic database search. Finally, known experts (individuals and organisations) in the field were contacted to determine if they had knowledge of publications on caries risk assessment tools that were not picked up by the other search methods.

Several strategies were used at this stage to ensure systematic and thorough documentation of the literature search processes. Each database search was imported and stored in separate EndNote files, named to reflect the database. In addition to this, search logs were created in MS Word for each database, which documented the name of the database, person conducting the search, date of search, time period covered, search strategy and the number of articles retrieved. This process and information are important for transparency and replicability.

Study selection

Decisions on the inclusion and exclusion criteria (Table 1), to determine study selection, were guided by the review question and objectives. These criteria were finalised through periodic meetings of the review team, an iterative process that developed with literature familiarity.

Two members (BC and LC) of the review team independently screened the title, abstract and keywords of selected articles against the inclusion and exclusion criteria. Where these fields lacked clarity the full text article was retrieved and used to make a decision. Differences were resolved via discussion. Following this initial screening, full text articles for the selected papers were retrieved and screened against the inclusion and exclusion criteria, again by two independent reviewers (BC and LC). As previously, disagreement in the screening process was resolved via discussion and this resulted in the final number of papers to be included in this review. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram was used to illustrate the flow of information through the study selection process and is shown in Figure 1 (122).

Data extraction and management

Data from the final list of selected studies were extracted into an evidence table designed specifically for this study and informed by Cochrane's data extraction form (121). Data extraction was performed by one reviewer (BC) and 20% of data extraction forms were checked by a second reviewer (outside of the authorship team) for accuracy. Any discrepancies in data extraction were resolved by discussion. This form captured information such as: author(s) details, study characteristics, population and setting, study objectives and methods, CRA tools used, tool testing methods, and outcomes and key results. Studies were organised chronologically.

Quality assessment

The quality assessment of included studies and measurement properties for identified CRATs was informed by the Consensus-based Standards for the Selection of Health Measurement INstruments (COSMIN) checklist (123). COSMIN provides a standard set of items to inform the selection of health measurement instruments and recognises three main domains for measurement properties – reliability, validity and responsiveness. As the name suggests, COSMIN provides a standard set of items to inform the selection of health measurement instruments and recognises three main domains for measurement properties – reliability, validity

and responsiveness (Chapter 5, Appendix 1). The COSMIN checklist consists of nine boxes, to reflect the measurement properties, with 5–18 items concerning methodological standards for how each measurement property should be assessed (124). The quality assessment included three steps: (A) an assessment of the methodological quality (risk of bias) of included studies; (B) an assessment of the quality of measurement properties for each CRAT in the included studies; and (C) an assessment of the overall strength of evidence for a particular CRAT using information from the previous two steps, a process similar to the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach that is used in reviews of clinical trials (125). For these three steps, the quality assessment was performed by one assessor, 20% independently assessed by a second assessor and discrepancies were resolved by discussion.

(A) Assessing the methodological quality of included studies using the COSMIN checklist

For this review, the methodological quality of a study on measurement properties was evaluated using the COSMIN checklist with a 4-point rating score (excellent, good, fair and poor) and the overall score for a particular study was determined by the lowest rating of any item in the checklist for the particular measurement property (125). The measurement properties internal consistency, structural validity and cross-cultural validity were not included in the quality assessment of included studies on CRA tools, as they were considered not applicable to the construct of caries risk. Internal consistency and structural validity are based on reflective models where all items that make up a particular instrument are a manifestation of the same underlying construct, and hence, are highly correlated. Whereas, for caries risk assessment instruments the items together form the construct risk status and are not necessarily correlated (124).

(B) Assessment of quality of measurement properties for CRA tools

The quality of the measurement properties for each CRA tool was assessed using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (123) and are shown in Chapter 5, Appendix 2. Minor revisions to the

criteria were made to add clarity to the rating methodology and to also reflect criteria used to determine study quality rating in the COSMIN checklist. For example, for criterion validity 'Sens+Spec ≥ 160' was added to the criteria for a '+' rating. This criterion of Sens+Spec ≥ 160 is generally accepted in the literature as being appropriate for a tool to predict caries (126).

(C) Assessment of the overall strength of evidence for per measurement property for a particular CRA tool

The overall strength of evidence to support the selection of a CRA tool was assessed using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (123) and are shown in Table 4. This rating was informed by the two previous steps: (A) assessment of individual study quality; and (B) assessment of the quality of measurement properties for each CRA tool. For example, a study with 'Excellent' rating for criterion validity, in step A and a '-' rating for quality of the measurement property criterion validity, in step B , achieved an overall rating of '---'; which was interpreted as strong evidence of a negative rating in one study of excellent methodological quality.

Data synthesis and analysis

The narrative synthesis was informed by Cochrane Consumers and Communication Review Group's document on data synthesis and analysis (127). The preliminary analysis described and compared study characteristics, was organised chronologically and included author information, year of data of collection, study design, population and setting, child age, number of participants, disease prevalence, geographic location and the CRA tool studied. The primary analysis was an assessment and comparison of the methodological quality of included studies on CRA tools, followed by a comparison of the overall strength of evidence for each identified CRA tool, by the specific measurement property. All comparisons were narratively described and presented in tables in the results section. The narrative description of the results in text was presented by a CRA tool and summarised the key results from each quality assessment table and in the same order for each CRA tool. The primary narrative synthesis was aligned with the research question and

focused on the overall strength of evidence for each CRA tool; specifically studying similarities and differences in quality ratings (including direction and size), and followed by possible explanations for the observed results. Sensitivity, specificity, positive predictive values and negative predictive values were collated (in some cases computed manually from data provided) from all studies. However, this did not warrant a meta-analysis given the different CRA tools and the limited studies on each tool.

4.2 The Experimental PhD Research Strands 2 and 3

The calibration study and the CRAT performance assessment, Chapters 6 and 7, respectively, were the two experimental strands of the PhD. The calibration study involved primary data collection from a subgroup of VicGen children and the CRAT performance assessment was conducted as a secondary data analysis using existing VicGen data. This aimed to assign risk status, using a selected CRAT, at children 18 months of age and to then track caries development at children aged 36 and 48 months. This would provide evidence for the CRAT's methodological quality.

4.2.1 The VicGeneration (VicGen) study: An Australian oral health birth cohort

The VicGeneration (VicGen) study was an oral health birth cohort study conducted to gain a deeper understanding of the multifactorial nature of early childhood caries (ECC) development. VicGen was conducted over eight years from 2008 to 2015 in the State of Victoria, Australia (128). It included seven waves of data collection corresponding to child ages 1, 6, 12, 18, 36, 48 and 60 months (25). For children, a clinical oral health examination was conducted, and saliva collected at all waves. Saliva was collected to study the oral microbiota. For the parent/caregiver, oral health examinations were conducted, and saliva collected at waves 2 (child age 6 months) and waves 6 (child age 48 months). A structured parent/caregiver self-administered questionnaire was used to collect data on: family socio-demographic-economics; child feeding habits; child and caregivers' oral hygiene-related behaviour; self-reported oral/general health status for child and care-giver; and care-giver's knowledge and attitudes about oral health (25).

VicGen was broadly representative of the state of Victoria. Sampled families with newborns were selected from rural, regional and metropolitan Victoria using a two-stage sampling method. In stage one, seven Local Government Areas (LGAs) in Victoria were selected to provide broad representation from across the State. The selected LGAs were not receiving specific oral health promotion programs. The second stage of sampling was families within the selected LGAs. This stage of the sampling focused on: socioeconomic disadvantage and cultural diversity. The metropolitan LGAs included a culturally and linguistically diverse (CALD) sample, and all LGAs had a high representation of socially or economically disadvantaged families. In each LGA, families with newborn children were recruited through the Victorian Government's Maternal and Child Health Service at the child's first health check. Families were excluded if: they intended to move location within the next 12 months; the children required specialist paediatric care; a family member had a severe illness; or the parent had a mental illness.

VicGen started with 466 children and included 465 mother-child and 1 father-child dyads. The Rural, Regional and Metro sample distribution was 24%, 11% and 65%, respectively (25). Selected primary carer reported characteristics were: 65% were born in Australia; 74% spoke English at home; 38% had a bachelor/postgraduate degree and 39% had a health care card (25). Health care cards provide access to cheaper services and medicines for people who meet certain eligibility criteria, generally determined by low income (129).

For the PhD secondary data analysis in research strand 3, I was provided with access to the VicGen data and was not involved in developing or implementing the VicGen study.

4.2.2 Why use the VicGen cohort for the experimental strands of the PhD?

A possible reason for the lack of methodological studies on the measurement properties of CRATs is the rigour, resources and, patient and clinician cooperation required to create the experimental setting necessary to study the phenomenon. For diseases like dental caries, it can take years for the clinical signs of disease to

become evident. Patients need to be regularly followed-up to identify early clinical signs of disease occurrence, which in most cases is not feasible in the health service delivery setting for various reasons including long waitlist backlogs to clear.

Birth cohort studies offer the unique opportunity to study a phenomenon over time and in a controlled setting. The VicGen birth cohort data was a natural choice for this PhD, as it included a comprehensive dataset of child (and primary carer) oral health examination data and corresponding data on the risk/protective factors for caries. In addition, as VicGen already had collected children's data from birth through to 60 months, it was possible to conduct a longitudinal performance assessment of the CRAT.

4.3 Research Strand 2, Chapter 6: The calibration study

This first experimental strand of the PhD involved primary data collection on a subset of children from the VicGen birth cohort. The purpose of this strand of the PhD research was to investigate a key criticism of the quality of the VicGen data before its use in strand 3 of the PhD research – the CRAT performance assessment.

VicGen's clinical oral health examiners were trained and calibrated to diagnose dental caries using intra-oral photographs of caries lesions, without an in vivo calibration strand. Reviewers of early publications using VicGen data noted the lack of an in vivo calibration exercise as a potential source of examiner bias, which would manifest in the inter- and intra-examiner reliability in caries diagnosis. Hence, this calibration study aimed to investigate the potential source of bias in the VicGen data, by determining examiner reliability in caries diagnosis when training and calibration was conducted by only using clinical photographs.

4.3.1 Method

Ethics approval for this study was provided by the Human Research Ethics Committee at the University of Melbourne (Ethics ID 1442648).

The study design was cross-sectional and fully-crossed, which uses the same examiners for all study participants, therefore allowing for systematic bias to be assessed and controlled for in the reliability estimate (130). Ten children from the VicGen birth cohort, ages 3-5 years, were purposively selected (based on caries activity) and invited to participate in this study. The selection process ensured presence of a range of caries lesions.

Sample size and participants

Based on estimates from the VicGen cohort and with the intention to set the minimum acceptable weighted kappa level as 0.61, an estimated sample size of 50 teeth (250 surfaces) was required to detect a weighted kappa score of 0.87 (95% CI: 0.64, 1.00). The sample size calculation was performed by manipulating the estimated distribution of ratings until a sufficiently narrow 95% confidence interval was obtained.

For this study, 880 tooth surfaces from 10 children were analysed. Ten children were purposively selected (based on caries activity) and invited to participate in this study. This selection process ensured presence of caries lesions to reflect the range of caries lesions as per the classification of the International Caries Detection and Assessment System (ICDAS).

Data collection

For this study, data on caries detection at the tooth surface level were collected from three sources: (1) Examiner training and calibration data from the ICDAS e-Learning training programme; (2) data from the in vivo community-based visual examination; and (3) data from examiner detection of caries lesions from the intra-oral digital photographs of the same tooth surfaces from the in vivo visual examination. Caries were detected and scored using the International Caries Detection and Assessment System (ICDAS) (132). ICDAS-II was used due to its ability to measure the progressive nature of the caries process. Air-drying of teeth, using compressed air, was not part of the caries detection process, as this was considered impractical in the community setting. Three dental examiners were used

in this study. The in vivo dental examinations were exclusively visual without the use of a dental probe. The unit of analysis for this study was the tooth surface and the selected ten children had 880 tooth surfaces which were included in the analysis.

ICDAS e-Learning programme training and calibration

The 90 Minute e-Learning programme by the ICDAS Foundation was used to train examiners in the application of ICDAS (131). An experienced clinician and researcher (HC), facilitated the training session. This was followed by a discussion of the scoring criteria until consensus was achieved, and a calibration exercise. For the calibration, examiners scored four sets of eight photographs each (32 photographs in total) of the various ICDAS-II codes, sourced from the e-Learning programme. Examiners were blinded to each other's scoring. Scoring was repeated to measure intra-examiner reliability.

In vivo community-based visual dental assessments

Each participant received two visits one week apart. At each visit the study participant had a dental assessment done by each of the three OHEs in the participant's home. The OHEs were blinded to each other's assessment. Five-minute breaks were provided between examinations for each child. In total each child had six caries assessments.

Children were examined in a community setting with the child lying down on a flat surface with the OHE situated behind the child. The examination involved standardised light sources and infection control protocol. A mouth mirror was used to view the teeth and gauze was used to remove tooth debris where required. The child's teeth were divided into quadrants and examined in a standardised manner. A trained research assistant was on site to record the caries scores onto paper forms, which were then double entered into an electronic database and checked for data entry consistency, to ensure data quality.

Intra-oral photographs

Intra-oral digital photographs of all tooth surfaces in children participating in the community examination were taken for comparison to the corresponding in vivo examination scores. These photographs were taken at either the first or second community visit depending on the participant's convenience. All the photographs were taken by the same OHE trained in the use of the intra-oral camera through a previous research study. The Sopro Acteon 717 intra-oral camera was used to take the photographs. The camera had its own light source and the following settings: intra-oral mode, colour images and output format as JPG. The protocol for taking intra-oral photographs was similar to the in vivo examination. The camera was connected to a computer and the images captured digitally and stored directly to an electronic folder with the ID number corresponding to the particular child. This data was backed up onto an external hard drive at the end of the community day.

For the analysis, 71 photographs were chosen based on clarity and to reflect a range of ICDAS codes. These photographs were scored by the same three examiners who participated in the community in vivo examinations. A standardised environment was provided for scoring of the photographs. One examiner was involved in photograph selection and collation. To avoid bias this examiner's scores were not used.

Data analysis

The unit of analysis for this study was the tooth surface. A child with a complete deciduous dentition of 20 teeth will have 88 tooth surfaces in total. Five tooth surfaces for each posterior tooth (molars – 8 teeth in total) and four tooth surfaces for each anterior tooth (incisors and canines – 12 teeth in total).

Kappa and weighted kappa scores were used to determine inter- and intra-examiner reliability for tooth surface level scoring of caries lesions. Weighted kappa is a recommended analysis technique for determining examiner reliability for ordinal data such as ICDAS caries codes (0 (sound surface) – 6 (extensive decay)). This statistic incorporates the factor of agreement by chance alone and also has a

feature of weighted proportional agreement. The 'kapci' command in STATA was used to generate kappa scores and 95% confidence intervals (95%CI), with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95% CI. A suggested guideline for Kappa scores: Under 0.20=poor; 0.21-0.40=fair; 0.41-0.60=moderate; 0.61-0.80=good and 0.81-1=very good reliability (132).

Clustering effects (surfaces within teeth and teeth within individuals) were explored and found to be minimal and hence, estimates did not require adjustment for clustering (133, 134).

To assess systematic bias in scoring caries severity the Stuart-Maxwell test for marginal homogeneity was conducted. This tests for the marginal homogeneity across all ICDAS categories simultaneously and determined if examiners were systematically scoring differently from other examiners or within themselves. Data were analysed using STATA 13.1 and SAS 9.2.

4.4 Research Strand 3, Chapter 7: The CRAT testing

Research strand 3 involved a methodological study to assess the performance of the CRAT identified in the systematic review (strand 1) as a promising tool for young children.

Though the research in this PhD strand used secondary data, the analyses were conducted as for a prospective study design, i.e. caries risk assessments were made before the cases of disease occurred and examiners were blinded to the caries status of the children. However, from an epidemiological perspective, the study design could be classified as a retrospective cohort, because the subjects were identified only after all data collection for the VicGen study was completed.

4.4.1 Method

The University of Melbourne Human Research Ethics Committee approved this study (Ethics ID 1851421).

Secondary data from the VicGen birth cohort at child ages 18, 36 and 48 months were analysed in this study. Data from the primary carer self-reported questionnaire at child age 18 months (VicGen wave 4) were used to generate responses for the CAMBRA1-5 CRAT's caries risk related questions (23). This process created a caries profile for each child at 18 months of age. Oral health practitioners then applied the CAMBRA CRAT1-5 to the caries profiles of the 233 caries-free 18-month-old children and categorised them as either high, medium or low risk of developing future caries. Caries development at child ages 36 and 48 months was used to validate the risk categorisation. Examiner reliability, a key measurement property of a CRAT, was also assessed as part of this study.

The analysis method for this study was informed by the Consensus-based Standards for the selection of health Measurement Instruments – COSMIN Checklist (124). Though the COSMIN checklist was developed to evaluate the methodological quality of studies on measurement properties of Health-Related Patient-Reported Outcomes (HR-PROs), it can be applied to other health-related measurement instruments, such as CRATs, as the design requirements and measurement properties are similar (123, 125). COSMIN provides assessment standards for three domains of measurement properties – reliability, validity and responsiveness.

Sample size considerations

To achieve an excellent rating for the quality of a measurement property, the COSMIN checklist recommends a sample size of at least 100 cases. This study included 214 cases which had complete data across the three time points – 18, 36 and 48 months of age.

CAMBRA CRAT items and VicGen variables

As CAMBRA CRAT items do not have specific definitions, the author team jointly discussed and agreed on the VicGen variables that best represented the CAMBRA CRAT items. In some cases, two or more VicGen variables were aligned with the CRAT item and in such cases composite variables of the two (or more) variables

were generated. For CRAT items where there was no clearly aligned VicGen variable, logic and evidence-informed assumptions were used to derive the appropriate variables. The CAMBRA CRAT items, the corresponding VicGen variable(s) at 18-months of age and the assumptions used to derive the CAMBRA CRAT item variables are shown in Chapter 7, Table 12.

Data cleaning and management

STATA was used to clean and manage the VicGen data to create the various analysis datasets. Four separate raw datasets were used to create the master dataset – the 18-month questionnaire data and the clinical examination data for 18, 36 and 48 months of age. The master dataset was then used to derive the variables for the CAMBRA CRAT items based on the 18-month questionnaire data. Once the CAMBRA variables were generated, another dataset with only the child unique identification numbers and the CAMBRA CRAT items was created. This CAMBRA dataset was used for the next stage of the research – developing the online caries risk assessment forms and the risk assessment process.

REDCap (Research Electronic Data Capture) hosted at La Trobe University, a secure web platform for building and managing online databases and surveys, was used to create the web-based caries risk assessment forms and manage the risk assessment process and data (135). Two separate surveys were created in REDCap which were then linked for the assessment process. The first survey was created to enable REDCap to directly pull/import the data from the STATA CAMBRA dataset into REDCap. To do this a replica of the STATA CAMBRA CRAT dataset was created in REDCap as a survey, with the variable names of each survey item in REDCap matching the STATA CAMBRA dataset. The completion of this first REDCap survey resulted in a CAMBRA CRA form for each child, which had responses for each CRAT item (Chapter 7, Appendix 3). The second survey was created to enable the examiners to assess each child case, based on their CAMBRA item responses, and categorise the case as high-, medium- or low-risk for future dental caries.

Caries risk assessments

Five examiners with a background in clinical dentistry were trained to apply the CAMBRA CRAT using the online CAMBRA Basic training course available at https://course.mycambra.com/courses/cambra-basic/. This was followed by a quick discussion between the examiners and myself to ensure agreement on assessment method. The caries risk assessments were conducted in REDCap, where the examiner was required to determine whether a child was high-, moderate- or low-risk based on yes/no responses to the CAMBRA CRAT items (which were pre-populated into the CAMBRA CRAT REDCap form).

Two examiners (HC and BC) established the reference standard caries risk status at 18 months of age for each child, by jointly assessing 25 cases to achieve consensus on the scoring method, which was followed by an independent assessment of a further 50 cases where any discrepancies in the assessments were resolved via discussion. Once consensus was achieved, BC assessed the rest of the cases to establish the reference standard risk categorization for the 18-month-old children. This reference standard was used in the validation analysis. To reflect real world conditions in current practice, examiners were not calibrated in using the CAMBRA CRAT.

Data analysis

On completion of the caries risk assessment stage of the research, the data were exported from REDCap into STATA 15.1 for analyses. Eighteen-month-old children who had a clinical examination at 36 and 48 months of age were included in this analysis (n=214). All children were caries free at 18-months of age. In this analysis, caries/caries lesion refers to any clinically apparent sign of tooth demineralisation [non-cavitated and cavitated lesions (ICDAS II scores 1-6)]. A sub-analysis was also conducted to determine which variables were responsible for the increased high-risk classification.

Reliability and measurement error

A fully-crossed design was used, where the same examiners were used for all study participants, therefore allowing systematic bias to be assessed and controlled for in the reliability estimate (130). Both inter- and intra-reliability estimates were calculated. For inter-examiner reliability, three examiners and the reference standard examiner assessed all 214 cases (blinded to each other's assessment) and assigned each case to a risk category (high- medium- or low-risk) based on responses to the CAMBRA CRAT items. For intra-examiner reliability, the first 100 cases were re-assessed by all examiners a few days after the first round of assessments. Reliability estimates were reported as weighted kappa scores and 95% confidence intervals (95% CI) and computed using the 'kapci' command in STATA with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95% CI. To assess systematic bias i.e. measurement error in the caries risk assessments, the Stuart-Maxwell test for marginal homogeneity was conducted to determine if examiners were systematically scoring differently from other examiners or within themselves.

Content validity

The content validity of the CAMBRA CRAT was assessed in our recently published systematic review on the quality of measurement properties for current CRATs for young children (136), and as such was not reported in this paper.

Construct (discriminatory) validity

Correlation estimates were computed between caries risk categorisation/status at 18 months of age and clinical evidence of caries at both 36 and 48 months of age. It was hypothesised that being categorised as high-risk at 18 months of age would be correlated with the greater likelihood of developing caries lesions at the two future time points, compared with being low- or moderate-risk. As the variables used to assess this measurement property were ordinal, Spearman's rank coefficients were used to estimate correlation between them. The distribution of caries lesions and sound tooth surface by risk status were also explored to demonstrate the validity of this measurement property.

Criterion validity

The reference standard for this measurement property was caries development at 36 and 48 months of child age. For the classic CAMBRA CRAT categorisation of low-, medium- and high-risk (no extreme risk was included here as these were very young children) area under the curve (AUC) was calculated. In addition, for comparison with other studies on this topic, the risk categories were dichotomised into high-risk (at-risk) versus low-risk (low and moderate risk categories were combined) and the following measures reported – sensitivity, specificity, positive predictive values and negative predictive values.

Responsiveness

The only difference between criterion (concurrent) validity and responsiveness is that criterion validity refers to the validity of a single score, and responsiveness refers to the validity of a change score. The change score for this analysis was the percentage of disease-free children at 36 months who developed caries lesions at 48 months of age. The analyses conducted were the same as for criterion validity.

4.5 Sequential Stages of the PhD

Figure 10 is a visual representation of the sequential stages of the PhD research and shows the relationship between the PhD research strands. For example, the findings from strands 1 and 2 were used to inform the research method for strand 3, in addition to contributing new knowledge to the overall PhD research.

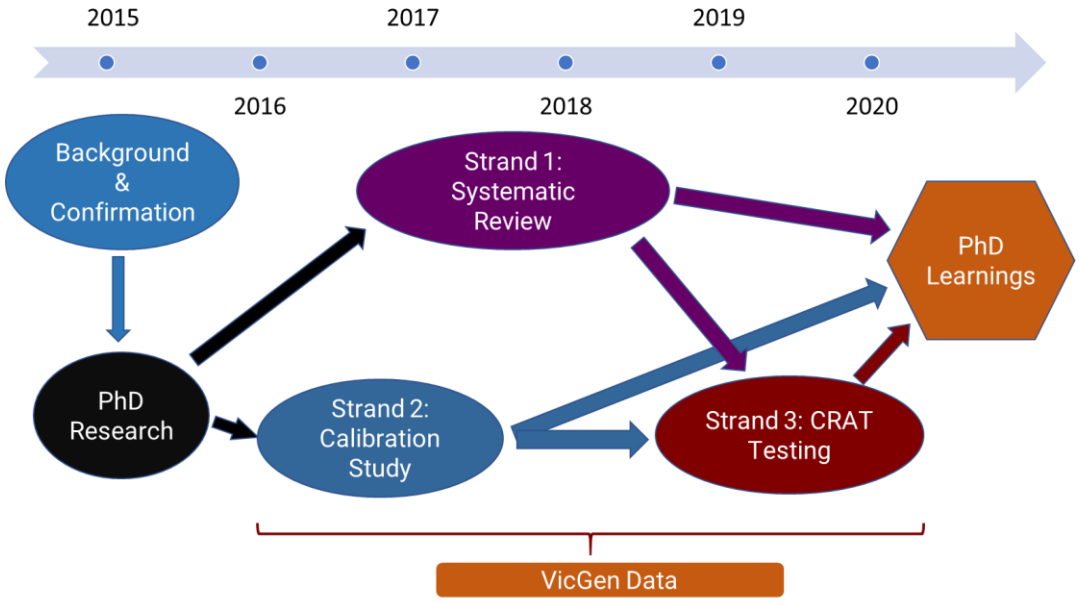


Figure 10. Sequential representation of the PhD stages

Chapter 5: Research Strand 1: Systematic Review

5.1 Preface

This chapter contains the draft manuscript, before peer-review, contributing to research strand 1 of this PhD thesis. The manuscript has been published in *the International Journal of Paediatric Dentistry*. The published paper is included in the appendices (Appendix 3. Research Strand 1). The research in this chapter is a systematic review to assess the strength of evidence to inform the selection of CRATs for children ages 6 years and less. It addresses the first objective of the PhD research study – To document and analyse the current state of evidence, including any knowledge gaps, on the quality of current CRATs for young children.

Citation: *Christian B, Armstrong R, Calache H, Carpenter L, Gibbs L, Gussy M. A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. International Journal of Paediatric Dentistry. 2019;29(2):106-16.*

5.2 The Publication

A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children.

5.2.1 Abstract

Background. At present, there are numerous caries risk assessment tools (CRATs) being promoted for disease management. However, the evidence to inform CRAT selection is unclear.

Aim. This review aimed to assess the strength of evidence to inform the selection of CRATs for children ages 6 years and less.

Design. MEDLINE was the principal search database for this review. Other key databases, the reference lists of included articles, known cariology literature and

experts were also consulted. Peer-reviewed papers describing CRATs and their development methodology were included. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist guided the quality assessment. The reporting of the key measurement properties (reliability, validity and responsiveness) informed the quality assessment.

Results. The search resulted in ten papers, reporting on eight different CRATs. The identified CRATs were: Caries Management by Risk Assessment (CAMBRA), Cariogram, National University of Singapore CRAT (NUS-CRAT), MySmileBuddy, Dundee Caries Risk Assessment Model, University of North Carolina Risk Assessment Models, University of Michigan pediatric dental clinic caries risk assessment sheet and American Academy of Pediatric Dentistry (AAPD) CRAT. Common across all CRATs was the lack of information to determine the levels of evidence for the measurement properties of reliability and construct validity. Studies on tools that were assessed as having strong evidence for content validity, identified the relevant risk factors for caries in the population being studied, before developing and testing their respective CRATs.

Conclusions. The evidence to inform the selection of current CRATs for children is mostly yet to be established. Overall, the NUS-CRAT studies reported the most information to inform the assessment of its measurement properties and as a result this tool attained a higher quality rating than other CRATs studied.

5.2.2 Introduction

A risk assessment is a method to quantify the risk of future disease using a combination of causative factors and disease indicators. Featherstone (137) defines caries risk assessment as a way to formalize and expand upon the patient's caries balance/imbalance in the most predictable fashion to diagnose current caries disease, to help determine the likelihood of future disease, and to determine what factors are out of balance so evidence-based clinical decisions can be made (137). In a broad sense, caries risk is the probability that the disease will occur in an individual given a particular combination of exposures. More specific definitions for this process are available. For example, Hausen defines caries risk as the probability of an individual developing at least a certain number of caries lesions reaching a given stage of disease progression during a specified time (82). The key feature of this definition is 'certain number of lesions reaching a given stage of disease progression', where the diseased state is determined by the presence of a caries lesion. Given the current evidence on caries aetiology and more effective minimally invasive preventive disease management methods, a more appropriate definition for caries risk is as follows: the probability of an individual developing any sign of caries activity in a specified period in time (83).

There are several reasons for the increased interest in objective and standardised caries risk assessments. First, the decline in caries rates, observed pre-dominantly across developed countries, resulted in the majority of disease occurring in a minority of the population. Identifying and managing this high-risk group has the potential to improve health outcomes for those that need it the most, while ensuring that scarce resources are used in the most effective and efficient manner. Second, a risk assessment informs decisions on recall times, which to date has been the arbitrary 6-months recall. Third, it helps inform disease management strategies.

Dental caries is a complex multi-factorial disease. The Fisher-Owens conceptual model illustrates this complexity by showing the various levels and factors that influence child oral health (85). The distinction between caries risk assessment methods (CRAMs) and caries risk assessment tools (CRATs) is important for this

topic. A CRAM describes any method that uses any individual risk or protective factor used to predict caries development or progression. Two recent systematic reviews on CRAMs (risk/protective factors) report on the accuracy of these various methods in caries prediction (17, 24). To further improve caries prediction, various combinations of CRAMs have been developed into so called caries risk assessment tools (CRATs) or systems (the term 'tools' will be used in this paper). At present, there are a variety of CRATs used in dentistry. Popular CRATs include: Caries Management by Risk Assessment (CAMBRA), American Association of Paediatric Dentistry Caries Assessment Tool (AAPD CAT) and Cariogram (18-20). Conceptually, risk assessment seeks to determine an accurate and precise risk level categorisation for the purposes of identifying those at highest risk of disease, to guide treatment decisions, and to determine appropriate recalls and health economics (115).

For any health measurement instrument, be it an index test, clinical rating scale or a self/patient-reported health outcome measure, the key measurement properties that ensure the results of the tool are interpreted properly are validity (accuracy), reliability (precision) and responsiveness. Reliability is the property of the tool to produce similar results under different conditions, whereas validity is the property of the tool to measure what it claims to measure. Responsiveness is the ability of a tool to validly detect change in a measured construct over time. Reliability includes the domains of internal consistency and measurement error. Validity includes domains such as content, construct and criterion validity (124).

Most evidence for CRATs and CRAMs has been generated from cross-sectional studies (17, 24, 50, 51, 138). The issue with cross-sectional studies is that caries experience is associated with past exposure over a lifetime to particular variables and hence, the ability to assess causality at a specific point in time is lost. An exposure is identified as being a risk factor for a particular outcome, in most cases, through longitudinal studies which determine the independent causal relationship between the exposure and the outcome (22). The lack of such studies in cariology is probably the reason for the lack of clarity on disease specific risk factors. Hence,

good quality longitudinal studies are required to establish temporal causal relationships between exposures and dental caries. Also, current research on CRATs and CRAMs has focused only on validity (mostly limited to criterion validity (reported as sensitivity and specificity)) in predicting a caries outcome (17, 21, 24). Without evidence for the quality of all relevant measurement properties for a CRAT, making an informed decision on tool selection is impossible. The authors of this review experienced this issue firsthand, in an attempt to select a CRAT for use in a risk-based caries management model of care at a public dental service in Melbourne, Australia. Contemporary caries management is reliant on valid and reliable risk assignment, making the necessary first step to: use evidence to inform CRAT selection; and where required, generate the knowledge to inform the selection of CRATs. Without this essential body of work, risk-based models of caries management run the risk of not delivering the intended outcomes.

This is not a review of caries risk assessment methods (CRAMs) and is not limited to criterion validity. Rather, this review will focus exclusively on CRATs and the collation and assessment of evidence on the important measurement properties of CRATs for children under 6 years of age. This review offers a critical assessment of the methodological quality of included studies and the measurement properties reported in these studies to inform the selection of CRATs for young children.

The research question guiding this review was: What is the strength of evidence to inform the selection of CRATs for children ages 6 years and less? The objectives were:

1. To identify existing CRATs for use with young children.
2. To assess the methodological quality of studies that assess the measurement properties of existing CRATs, including quality of the reported measurement property.
3. To assess the strength of evidence (overall quality) for a CRAT.
4. To identify gaps in the research on CRATs.

5.2.3 Materials and methods

Identification of studies

MEDLINE was the principal search database for this review. Other databases searched included CINAHL, PubMed and Scopus. Key search terms were selected in consultation with experts in the field and a University librarian. These keywords were used to generate a Boolean search string, modified slightly for each database. The base Boolean string was – (caries or dental caries) AND (risk assessment or risk measur*) AND (measur* or tool*). In addition, we also conducted a phrase search for known caries risk assessment tools. The Boolean string for this search was – ("caries management by risk assessment" OR CAMBRA OR cariogram OR "caries management system"). A University of Melbourne librarian familiar with the terminology in this field helped design and implement the search strategy.

The reference lists of articles selected for inclusion in the review and known cariology literature (including textbooks) were also screened to identify studies that may have been missed in the electronic database search. Finally, experts (individuals and organisations) in the field were contacted to determine if they had knowledge of publications on caries risk assessment tools that were not picked up by the other search methods.

Study selection

Decisions on the inclusion and exclusion criteria (Table 1), to determine study selection, were guided by the review question and objectives. These criteria were finalised through periodic meetings of the review team, an iterative process that developed with literature familiarity. For example, the inclusion criterion 'children ≤ 6 years' was introduced only at the full text screening stage, as the literature showed that this was a sufficiently different age cohort, in terms of having different risk/protective factors to older people (51), to warrant a separate analysis. Several systematic reviews were identified but not included after application of inclusion/exclusion criterion. These excluded systematic reviews were screened for primary research papers that might not have been captured by the search strategy.

A two-step screening process was employed – title/abstract and full text. Two members (BC and LC) of the review team independently screened selected articles against the inclusion and exclusion criteria. Differences were resolved via discussion. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram was used to illustrate the flow of information through the study selection process and is shown in Figure 1.

Quality assessment

Several tools were considered for the quality assessment component of this review. The QUADAS (Quality Assessment for Diagnostic Accuracy Studies) tool has been used in previous systematic reviews on caries risk assessment methods (140). However, as the name indicates QUADAS focuses only on the single measurement of property and validity, with the domains mainly related to criterion validity. For CRATs, content validity and construct validity are equally important. The QUADAS tool also has relatively more subjective questions than the COSMIN checklist and needs to be paired with another relatively subjective method like Grading of Recommendations Assessment, Development and Evaluation (GRADE) to determine overall strength of evidence (140). This review required a more objective method of quality assessment which focused on all the important measurement properties for CRATs. Therefore, the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist was used (125).

COSMIN provides a standard set of items to inform the selection of health measurement instruments. The COSMIN checklist consists of nine boxes, to reflect important measurement properties (Appendix 1), with 5–18 items describing methodological standards for how each measurement property should be assessed (125). For this review the measurement properties of internal consistency, structural validity and cross-cultural validity were not assessed, as they were considered not applicable to CRATs. Internal consistency and structural validity are based on reflective models where all items that make up an instrument are a manifestation of the same underlying construct and hence, are highly correlated (124). Whereas, for CRATs the items together form the construct ‘risk’ status and are not necessarily

correlated. Cross-cultural validity was not assessed, as it relates to the validity of translation of CRATs into other languages and this was not the study focus.

The quality assessment in this review was conducted in three steps. For these three steps, the quality assessment was performed by one assessor (BC) and 20% independently assessed by a second assessor (LC). Discrepancies were resolved by discussion.

Step A. Assessing the methodological quality (risk of bias) of included studies

Firstly, the methodological quality of a study was evaluated using the COSMIN checklist with a 4-point rating score (excellent, good, fair and poor) and the overall score for a particular study was determined by the lowest rating of any item in the checklist for the particular measurement property (125).

Step B. Assessment of quality of measurement properties in each study by CRAT

The quality of the measurement properties in each study was assessed by CRAT using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (Appendix 2) (141). Minor revisions to the criteria were made to add clarity to the rating methodology and to also reflect criteria used to determine study quality rating in the COSMIN checklist. For example, for criterion validity 'Sens+Spec ≥ 160' was added to the criteria for a '+' rating. This criterion of Sens+Spec ≥ 160 is generally accepted in the literature as being appropriate for a tool to predict caries (126).

Step C. Assessment of the overall strength of evidence, by measurement property, for a CRAT

The overall strength of evidence to support the selection of a CRAT was assessed using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (Appendix 3) (141). This rating was informed by the two previous quality assessment steps. For example, an 'Excellent' study rating for criterion validity in Step A and a '-' rating in Step B, for quality of the measurement

property criterion validity, achieved an overall rating of '---' in Step C (this step) of the quality assessment. This was interpreted as strong evidence of a negative rating for criterion validity in one study of excellent methodological quality.

Data synthesis and analysis

The analysis assessed and compared the methodological quality of included studies on CRATs, followed by a comparison of the overall strength of evidence for each identified CRAT. The in-text results were described by CRAT and summarised the key results from each quality assessment step. The narrative synthesis of the results was guided by the Cochrane Consumers and Communication Review Group's document on data synthesis and analysis (127) and was aligned with the research question and focused on the overall strength of evidence for each CRAT.

5.2.4 Results

The search strategy resulted in the identification of 432 unique papers, of which 167 were selected for full-text review based on their title and abstract. On completion of the full text screening, 10 (of the 167) papers, assessing 8 different CRATs, were included in this review (Figure 1). The general characteristics of included studies and their respective CRATs are presented in Table 2.

To align with the research question, the main results are reported below by CRAT. Table 3 presents the methodological quality of each study by measurement property and CRAT. Table 4 presents the quality of each measurement property by CRAT. Table 5 presents the overall strength of evidence for each CRAT by measurement property.

The University of North Carolina Caries Risk Assessment studies (UNCCRA) were conducted to improve methods to identify children prospectively at high risk to dental caries (126, 142, 143). These CRA models have around fifteen items and the assessor determined the caries risk level. The methodological quality of these studies was rated 'Poor' for reliability and measurement error (Table 3). For quality of measurement properties only content validity achieved a positive rating (Table

4). There was strong positive evidence for content validity and strong negative evidence for criterion validity and responsiveness in the overall strength of evidence assessment (Table 5).

The University of Michigan pediatric dental clinic CRA sheet (UMCRA) was developed for initial and recall patient encounters in the Young Patient and Prevention Clinic (YPPC) at the University of Michigan (144). The CRA sheet consists of 14 items, which are used by the assessor to determine the caries risk level. There were no methodologically sound studies found for any of the measurement properties (Table 3). No studies achieved a positive rating for quality of measurement properties (Table 4). For the overall strength of evidence by measurement property, the evidence was mostly unknown due to poor methodological quality (Table 5).

Cariogram is a computer program which graphically represents a caries risk profile for an individual and is expressed as 'chance (%) to avoid caries'. This tool consists of nine items and the risk level is program generated based on a pre-determined algorithm (145). The methodological quality of studies was rated 'good' for criterion validity and responsiveness (Table 3). For quality of measurement properties none achieved a positive rating (Table 4). For the overall strength of evidence for the CRAT, by measurement property, there was strong negative evidence for Cariogram's criterion validity and responsiveness (Table 5).

Similar to the Cariogram, the National University of Singapore caries risk assessment tool (NUS-CRAT) classifies children's caries risk into five risk levels and is expressed as 'chance (%) to avoid caries'. This tool has eleven items (6 items in the community-screening version) and the risk level is program-generated based on a pre-determined algorithm (146, 147). The methodological quality of studies was rated as 'Fair' or higher for all measurement properties with content validity achieving an 'Excellent' rating (Table 3). For quality of measurement properties positive ratings were achieved by responsiveness and content and criterion validity (Table 4). There was strong positive evidence for NUS-CRAT's content, criterion

validity and responsiveness in the overall strength of evidence assessment (Table 5).

The Dundee Caries Risk Assessment Model (DCRAM) is structured in a decision tree format and includes 2–5 items (depending on the particular DCRAM to be used) to identify children at high risk for dental caries (148). The methodological quality of studies was mostly rated as 'Fair' or higher, except for reliability and measurement error which were rated as 'Poor' (Table 3). For quality of measurement properties none achieved a positive rating (Table 4). There was strong positive evidence for DCRAM's content in the overall strength of evidence assessment (Table 5).

The American Academy of Pediatric Dentistry Caries Assessment Tool (AAPD-CAT) designed for use in 0-5-year-olds includes 14 items to classify children as either high, medium or low risk for dental caries (18, 146, 149). The methodological quality of studies was rated as 'Poor' for content validity (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). There was strong negative evidence for AAPD-CAT's criterion validity and responsiveness in the overall strength of evidence assessment (Table 5).

The Caries Management by Risk Assessment (CAMBRA) is an approach that couples risk assessment with tailored preventive care and risk monitoring. The CRAT form for 0-5-year-olds includes 20 items to classify children as either high, moderate or low risk for dental caries (23, 146, 150). The assessor determines the caries risk level using these twenty items. The methodological quality of studies was rated 'Poor' for reliability and measurement error (Table 3). For quality of measurement properties, a positive rating was achieved for content validity (Table 4). There was strong positive evidence for CAMBRA's content validity in the overall strength of evidence assessment (Table 5).

MySmileBuddy (MSB) is a risk assessment, caregiver education, and ECC management tool. MSB consists of a series of five assessment modules containing questions and brief educational units, one each on, feeding practices, attitudes and

beliefs, fluoride, and family history. Risk levels are program-generated and classify children as low (score: 1-3), medium (score: 4-6), and high (score: 7-10) for caries (151, 152). The methodological quality of studies was rated as 'Fair' or lower for measurement properties (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). The overall strength of evidence for the CRAT, by measurement property, was unknown due to the tool being early in its development stages (Table 5).

Narrative synthesis

Common across all tools was a lack of information to determine the levels of evidence for the key measurement properties of reliability (including measurement error) and construct validity. In terms of content validity, UMCRA, Cariogram, AAPD-CAT and MSB achieved 'unknown' ratings. The main reason for this rating was that the risk assessment items in those CRATs were not demonstrated to be relevant to the target population in which the tools were used. Studies on tools that were assessed as having strong evidence for content validity, identified the relevant risk factors for caries in the population being studied, before developing and testing their respective CRATs. For criterion validity and responsiveness, only the NUS-CRAT achieved a strong level of evidence. This was driven mainly by the fact that only the NUS-CRAT achieved a combined sensitivity and specificity score of over 160 and individual sensitivity and specificity scores of at least 75 and 85, respectively. These are the recommended minimum scores for a tool to predict future caries. Overall, based on reported information, the NUS-CRAT attained a higher quality rating than other CRATs studied by achieving strong evidence ratings in 3 out of the 6 measurement properties studied.

5.2.5 Discussion

This review showed that the overall strength of evidence to inform the selection of current caries risk assessment tools, for children under 6 years of age, is mostly unknown due to poor methodological quality of studies assessed using the COSMIN checklist. For example, while criterion validity/responsiveness (determined using sensitivity and specificity estimates) is reported in most studies, properties of a tool

such as reliability in tool application, measurement error, construct validity (including discriminatory validity) and content validity have been, in most cases, poorly studied. This review is in agreement and augments a recent review on the evidence for current caries risk assessment systems (21). While this 2013 review focused on validity of current tools, my PhD review included the range of measurement properties relevant to CRA tools and also focused on a specific age, children ages 6 years and less. The rationale to focus on this age range stemmed from a recent paper, which reported a substantial increase in caries rates from ages 18 to 36 months (45). In addition, as described in the introduction section, very young children are a sufficiently different cohort to warrant sub-group analysis.

The systematic review conducted for this PhD study identified several key issues with studies on current CRA tools for young children. First, reliability (which includes measurement error) is a key measurement property, particularly for reasoning-based user-determined caries risk levels. It is also called precision and can be defined as the ability of the tool to produce the same results when repeated by the same or different assessors, under the same conditions (22). Domejean et al., state the following, in relation to reliability, in their recent review on caries risk assessments “At a practice level, it is suggested that all members of each oral healthcare team are calibrated among each other (using test clinical scenarios and group discussion/consensus), so that risk levels obtained are meaningful across the team and to the patient and any future changes can be monitored and documented over time” (153, p. 195). This statement shows that reliability is an important property for caries risk assessment tools. For CRA tools where the risk levels are algorithm-based program-generated, such as Cariogram and NUS-CRA it is assumable that the same risk level will be generated when the same information is entered for each item by different users. However, until reliability and measurement error estimates are provided for such tools the evidence on reliability remains unknown. Second, the evidence on content validity was unknown for several tools and this rating was mostly due to the tool items not being checked for relevance among population in which the tool was tested. The UNCCRA studies highlighted this issue where caries prevalence rates, geographic location and age

of the population determined the factors to include in the risk prediction models and eventually the items in the risk assessment tool (143, 154). This is a very important consideration for clinical practice as it raises the issue of adopting tools in practice without testing them for relevance to the population being served. Also, for caries risk assessments in young children this could be a major issue as the caries related factors significantly differ from those among older children (51). As such for non-age specific tools, such as the cariogram, incorporating these age-related factors and re-working the algorithms may lead to better performance. Third, discriminatory validity a component of construct validity was another measurement property, particularly relevant to CRA tools, for which the evidence was unknown across all CRA tools for young children. It is the ability of the CRA tool to differentiate among risk levels (155). Without reporting on discriminatory validity, the risk level categorisation is arbitrary and based more on a theoretical intellectual process rather than evidence. The entire concept of a risk assessment is based on accurate and precise risk level categorisation for the purposes of identifying those at highest risk of disease, to guide treatment decisions, to determine appropriate recalls and health economics (27). In fact, oral health services in Australia have begun the process of re-orienting their practice to using risk-based minimally invasive models of care for afore stated reasons (16). If these new models of oral healthcare use risk-based management, then as a first step we need to: ensure the evidence to support the caries risk assessment process is strong; use evidence-based methods to select a CRA tool; and where required, generate new knowledge to support the use of current tools and where required develop new tools. Without this step, risk-based models of oral healthcare run the risk of not delivering the intended positive outcomes. Hence, from a clinical practice perspective it is important that risk categorisations be accurate, precise and supported by evidence.

The items constituting each tool identified in this review is provided in the appendix table. The UNCCRA studies were conducted in the late 80s and early 90s to improve methods to identify children at high risk to future dental caries (142, 143, 154). The studies were mostly methodologically sound, and the risk prediction models developed through a rigorous process, though in terms of sensitivity and specificity

none of the models achieved the recommended combined rating of 160 for these two measures of criterion validity. Cariogram, a popular non-age specific computer-based program, graphically illustrates an individual's caries risk profile and provides an associated risk rating - % chance of avoiding caries; with the 0-20% chance of avoiding caries generally defined as the high risk group (19); though studies have re-defined this high risk grouping to suit their situation (147, 148). The cariogram has been tested relatively widely compared to other CRA tools, however, in young children its testing has been limited (156-158). The evidence to inform the selection of this tool mostly achieved an 'unknown' rating. This was due to cariogram mostly being used as a comparator for other tools in the included studies, rather than specifically assessing its usefulness in the testing population. As a result, measurement properties such as content and construct validity of the cariogram were not investigated in these studies (146, 147). CAMBRA, is another popular caries risk assessment system. It is popular because it also provides clear management guidelines for each risk level (23). CAMBRA is mostly a reasoning-based clinician determined risk level CRA tool, though more recently an algorithm-based version, MyCAMBRA is available for purchase online. For reasoning-based qualitatively determined caries risk levels, reliability in the application of the tool is important to ensure consistency across clinicians in risk assignment as well as the associated management strategies. The evidence for this measurement property of CAMBRA is at present unknown and future studies using this tool are encouraged to measure reliability in risk level categorisation across clinicians, particularly if using the reasoning-based version of CAMBRA. For criterion validity and responsiveness, CAMBRA was assessed as having strong evidence for a negative result. This was mainly due to CAMBRA not achieving the recommended combined sensitivity and specificity score of 160 (143). When individual sensitivity and specificity scores were examined, CAMBRA consistently achieved a high sensitivity score of 0.84 and higher. Specificity scores though were much lower with a maximum of 0.63 in the included studies. AAPD-CAT developed by the American Association for Pediatric Dentistry (18), as with other CRA tools, has an unknown level of evidence for the measurement properties of reliability (including measurement error) and construct validity. In addition, the level of evidence for its

content is also unknown. This could be due to the lack of published literature that describes the process of identifying and including the items that constitute this tool. Of note with the AAPD CAT, was the perfect sensitivity of 1 and almost no (0) specificity and these estimates were similar across two different studies among young children that used this tool (146, 149). The authors of one of these studies state that these estimates could be due to a single item determining the risk level (146). For example, in AAPD-CAT a 'Yes' response to a single item such as 'Child has >3 between meal sugar-containing snacks or beverages per day' could be used to classify the child as high risk. DCRAM is a CRA tool with the least number of items (2-5 items depending on model), has strong evidence to support its content and uses a decision tree format to arrive at a risk level (148). The tool is relatively new compared with other tools and has had limited testing beyond the author group that developed the tool. The NUS-CRA (146, 147), a recently developed and promising risk assessment tool for young children, had the strongest evidence to support its selection, compared with other CRA tools assessed using the COSMIN checklist in this review. The authors of studies on NUS-CRA attribute its positive performance to two main reasons: First, NUS-CRA being algorithm driven allows for a mathematical synthesis of risk factors and indicators and defining their relative contribution to risk using pre-determined weights. This process generates a more objective risk level, compared with reasoning-based tools that subjectively estimate risk levels. The second reason for NUS-CRA's better performance could be the inclusion of age-specific risk factors such as infant feeding practices in the assessment process. To progress the development of this tool its performance will need to be tested in other populations and it will also need to be supported by management strategies appropriate to the calculated risk level. MySmileBuddy (MSB) is the most recently developed CRA tool that also provides oral health education information and caries management protocols (151). The risk assessment component of the tool has a comprehensive dietary component in recognition of the fact that diet and caries have a consistent association in the literature. MSB is an algorithm-based program. Comprehensive and dietary risk scores are algorithm generated as low (score: 1-3), medium (score: 4-6), and high risk (score: 7-10) (151). The tool is in its very early stages of development, hence

the reason for the 'unknown' level of evidence across all measurement properties of the tool. However, being in the early stages of development is an advantage as the tool lends itself to more rigorous testing guided by a standardised checklist for tool development and testing such as COSMIN (124).

The recently developed COSMIN checklist, as the name states, provides a consensus-based standard for the selection of a health measurement instrument (159). Though the COSMIN checklist was developed to evaluate the methodological quality of studies on measurement properties of Health-Related Patient-Reported Outcomes (HR-PROs), it can be applied to other health-related measurement instruments, such as CRA tools, as the design requirements and measurement properties are similar (124, 125). In addition to being used as a standard to assess methodological quality of CRA tools, the suggested methods in the COSMIN checklist can also be used to guide the development and testing protocols for new health-related measurement instruments. It can also be used as a reporting template for studies on health measurement instruments. This systematic review also identified several issues with using the COSMIN checklist. First, the checklist is comprehensive, complex and requires a fair bit of self-learning to familiarise with the measurement properties and rating criteria. Certain measurement properties required more specific author generated definitions for clarity and standardisation in the rating process. For example, a question on content validity is – 'Was there an assessment of whether all items refer to relevant aspects of the construct to be measured?' In this case, a clear definition of the construct and the appropriate assessment is required to ensure consistency in application of the rating criteria. Second, in relation to construct validity the quality criteria require that studies state an a priori hypothesis. None of the studies in this review stated a hypothesis and as such received a lower quality rating. Hence, the rating criteria definitions may need to be revised to better fit an area of research, with the understanding that it will take time for tools such as the COSMIN checklist to become standard practice.

In recent times the debate between algorithm-driven program-generated versus reasoning-based user-determined CRA tools is increasing. Though research

comparing the usability and other measurement properties for these two approaches to caries risk assessments is limited, emerging evidence supports the use program-generated risk tools (146, 160). For example, a recent study comparing six CRA tools reported reliability scores of 94% and 92% for the program-generated Clinical Decision Support (CDS) and MyCAMBRA CRA tools, respectively; compared with 81% and 63% for the user-determined University of Michigan and Consortium for Oral Health Research and Informatics (COHRI) CRA tools, respectively (160). In the same study, program-generated CRA tools took, on average, less than a minute to complete which was significantly lower than user-determined tools. The setting will probably determine which tool is most appropriate. In an education setting, a reasoning-based CRA tool is probably more useful to stimulate intellectual thinking and discussions on the caries disease process, associated factors and best management practices. Generally, in such a setting, time is not a constraint and reliability in application of the tool may not be as important compared to a clinical practice setting. In a non-dental provider setting such as maternal child nurses, paediatricians, mid-wives and health workers, a tool that is quick to administer, does not require much intellectual reasoning and consistent may be more effective, efficient and will probably see more uptake among non-dental providers; and this points towards a program-generated risk assessment.

5.2.6 Conclusions

This systematic review shows that the evidence to inform the selection of existing CRA tools is at present mostly unknown. The findings do not indicate that current CRA tools should not be used, rather it shows that methodologically sound studies are required to provide the evidence on their measurement properties. In addition, caries risk assessments are a valuable intellectual process for the clinician and the process also helps the client understand the disease process and risk factors specific to their situation thus enabling targeted action.

This review also demonstrates that current tools differ: in definitions of risk levels and caries; the tool content; caries disease prevalence in the population used to test the tool; analytic techniques, measurement properties and estimates reported; and

study design. With this level of variation between tools, selecting a particular tool for use in practice is a thought-provoking process. The authors' recommendations for selecting a current CRA tool, based on this review, include: (1) the purpose and setting of the tool – for education, clinical practice or for use in a non-dental provider setting (2) know the disease prevalence in the population the tool is going to be used and select a tool that was tested in a population with similar disease levels. This will provide some indication of how effective the tool will be and is particularly applicable for disease prevalence dependent positive and negative predictive values (important from a clinician's perspective). (3) Choose a tool that was tested in a population with similar demographics particularly age, as it has been shown that risk items in the CRA tool can vary based on socio-demographics and disease levels.

The authors of this review recommend future studies on this topic: (1) Use good research methods and appropriate study designs to identify caries risk factors (causal relationship between exposure and outcome) appropriate to the population in which the tool is to be applied, (2) use the COSMIN checklist or an adapted version to guide the development and testing process of CRA tools. The checklist also provides a standardised reporting framework for comparison across studies on CRA tools, and (3) Go beyond the CRA process and also include management strategies, and study how the system as a whole works to improve outcomes for the client and the service provider.

Table 1. Inclusion and exclusion criteria

Criterion	Inclusion	Exclusion
Population	Children ≤6 years of age (only applied at the full text screening stage)	Special populations: People with a disability and chronic conditions, who are already at high risk and orthodontic patients.
Index tests	Papers that describe CRATs or systems and their development processes – which can include evidence/ theory behind the CRAT and the methodological development including the validity and reliability of the tool.	Original research that only reports on caries risk-based management and do not discuss the application of the CRAT to assign risk and its development.
Target conditions	CRAT to determine an individual's risk – Any instrument to determine an individual's risk of developing future dental caries Coronal caries	No reference to a CRAT Population level CRAT Tooth level caries risk Fluorosis Periodontal disease Root caries
Study and information types	Peer-reviewed publications of original research that report on the application of CRATs to determine caries risk. Non-scientific reports on CRATs Peer-reviewed thesis	Conference presentations and/or abstracts. Commentaries Literature and systematic reviews Non-English publications

Table 2. Characteristics of included studies

Author, publication year	Study year(s)	Study design	Population and setting	Age[a]	N	CP	Predicted caries definition	Country	CRAT(s)
Disney et al., 1992 (143)	1986-1989	Longitudinal	Grade 1 in the geographic areas surrounding 1. Aiken, SC and 2. Portland, Maine	6	4000	72%[b] 40%[c]	dmfs – caries into dentine	USA	University of North Carolina Caries Risk Assessment studies (UNCCRA)
Beck et al., 1992 (142)	1986-1989	Longitudinal	Grade 1 children in the geographic areas surrounding 1. Aiken, SC and 2. Portland, Maine	6	4000	72%[b] 40%[c]	dmfs – caries into dentine	USA	University of North Carolina Caries Risk Assessment studies (UNCCRA)
Wandera et al., 2000 (144)	1994-1997	Longitudinal	Children attending the Young Patient and Prevention pediatric dental Clinic at the University of Michigan	4	140	41%	Not reported Most likely into dentine	USA	University of Michigan Pediatric Dental Clinic tool (UMCRA)
Holgerson et al., 2009 (145)	2000-2007	Longitudinal	Children born between January 2000 and March 2001, attending the Public Dental Clinic in Lycksele, a small municipality in northern Sweden	2	103	71%	Enamel & dentine lesions	Sweden	Cariogram
Gao et al., 2010 (147)	NR	Longitudinal	Children in government kindergartens in Singapore	3-6	1782	40%	dmft – caries into dentine	Singapore	National University of Singapore caries risk assessment tool (NUS-CRA)
MacRitchie et al., 2012 (148)	1993-1997	Longitudinal	The cohort comprised all children born and resident in Dundee between 1 April 1993 and 31 March 1994	0-4	1890	49%	Enamel & dentine lesions	Scotland	Dundee Caries Risk Assessment Model (DCRAM)
Yoon et al., 2012 (149)	2006-2009	Longitudinal	Residents served by the University of Pediatric dental clinic, with fluoridated water in the northern part of the New York City borough Manhattan, which have predominantly low-income and Hispanic populations	≤ 3	471	49%	Enamel & dentine lesions	USA	American Academy of Pediatric Dentistry caries assessment tool (AAPD-CAT)
Gao et al., 2013 (146)	NR	Longitudinal	Children at kindergartens in Hong Kong	3	544	35%	dmft – caries into dentine	Hong Kong	NUS-CRA Cariogram CAMBRA AAPD CAT
Chaffee et al., 2016 (150)	2009-2015	Observational retrospective, longitudinal analysis	Patients at the University of San Francisco Pediatric Dentistry clinic	0-6	1315	67%	Enamel & dentine lesions	USA	Caries Management by Risk Assessment (CAMBRA)
Custodio-Lumsden et al., 2016 (151)	2012-2013	Cross-sectional	Caregiver/child dyads presenting to the Columbia University College of Dental Medicine Pediatric Dental Clinic	2-6	108	35%	caries into dentine – frank cavities	USA	MySmileBuddy (MSB)

Age in years; [b]Caries prevalence for Aiken, South Carolina; [c]Caries prevalence for the Portland, Maine; CP=caries prevalence among population in which tool tested

Table 3. Methodological quality of each study by measurement property and caries risk assessment tool

CRAT & study(ies)	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsiveness
UNCCRA						
Disney et al., 1992	Poor	Poor	Excellent	Fair	Excellent	Excellent
Beck et al., 1992	Poor	Poor	Excellent	Fair	Excellent	Excellent
UMCRA						
Wandera et al., 2000	Poor	Poor	Poor	Fair	Poor	Poor
Cariogram						
Holgerson et al., 2009	NA	NA	Poor	Fair	Good	Good
Gao et al., 2010	NA	NA	Poor	Fair	Good	Good
Gao et al., 2013	NA	NA	Poor	Fair	Good	Good
DCRAM						
MacRitchie et al., 2012	Poor	Poor	Excellent	Fair	Good	Good
AAPD CAT						
Yoon et al., 2012	Poor	Poor	Poor	Fair	Good	Good
Gao et al., 2013	Poor	Poor	Poor	Fair	Good	Good
NUS CRA						
Gao et al., 2010	NA	NA	Excellent	Fair	Good	Good
Gao et al., 2013	NA	NA	Excellent	Fair	Good	Good
CAMBRA						
Gao et al., 2013	Poor	Poor	Poor	Fair	Good	Good
Chaffee et al., 2016	Poor	Poor	Excellent	Fair	Excellent	Excellent
MSB						
Custodio-Lumsden et al., 2016	NA	NA	Poor	Fair	Poor	NA

NA – Not Applicable

Table 4. Quality of the measurement properties by caries risk assessment tool and study

Author, year	CRAT(s)	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsive-ness
Disney, 1992	UNCCRA	?	?	+	?	-	-
Beck, 1992	UNCCRA	?	?	+	?	-	-
Wandera, 2000	UMCRA	?	?	-	?	-	-
Holgerson, 2009	Cariogram	?	?	-	?	-	-
Gao, 2010	NUS-CRA^{s}	?	?	+	?	+	+
	NUS-CRA^{C}	?	?	+	?	+	+
	Cariogram	?	?	-	?	-	-
MacRitchie, 2012	DCRAM1	?	?	+	?	-	-
	DCRAM2	?	?	+	?	-	-
	DCRAM3	?	?	+	?	-	-
	DCRAM4	?	?	+	?	-	-
Yoon, 2012[a]	AAPD CAT	?	?	-	?	-	-
Gao, 2013	NUS-CRA^{S}	?	?	+	?	-	-
	NUS-CRA^{C}	?	?	+	?	+	+
	Cariogram^{S}	?	?	-	?	-	-
	Cariogram^{C}	?	?	-	?	-	-
	CAMBRA^{S}	?	?	-	?	-	-
	CAMBRA^{C}	?	?	-	?	-	-
	AAPD CAT^{S}	?	?	-	?	-	-
	AAPD CAT^{C}	?	?	-	?	-	-
Chaffee, 2016	CAMBRA	?	?	+	?	-	-
Custodio-Lumsden, 2016	MSB	?	?	-	?	?	?

NUS-CRA S = screening tool, reduced # risk factors assessed – excludes microbiology and saliva analysis
NUS CRA C = comprehensive, includes all risk factors
DCRAM1 – predicting d1mft>0 (All caries lesions and predicting any caries)
DCRAM2 – predicting d3mft>0 (Dentine caries lesions and predicting any caries)
DCRAM3 – predicting d1mft≥3 (All caries lesions and predicting more than 2 caries lesions)
DCRAM4 – predicting d3mft≥3 (Dentine caries lesions and predicting more than 2 caries lesions)
[a]Only overall AAPD CAT used
+ = positive rating? = indeterminate (Unknown) rating, – = negative rating

Table 5. Overall strength of evidence for each caries risk assessment tool, by measurement property.

CRAT	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsiveness
UNCCRA	?	?	+++	?	---	---
UMCRA	?	?	?	?	?	?
Cariogram	?	?	?	?	---	---
NUS-CRA	?	?	+++	?	+++	+++
DCRAM	?	?	+++	?	--	--
CAMBRA	?	?	+++	?	---	---
AAPD-CAT	?	?	?	?	---	---
MSB	?	?	?	?	?	?

+++ or --- strong evidence positive/negative result, ++ or -- moderate evidence positive/negative result, + or - limited evidence
positive/negative result, ± conflicting evidence? unknown, due to poor methodological quality

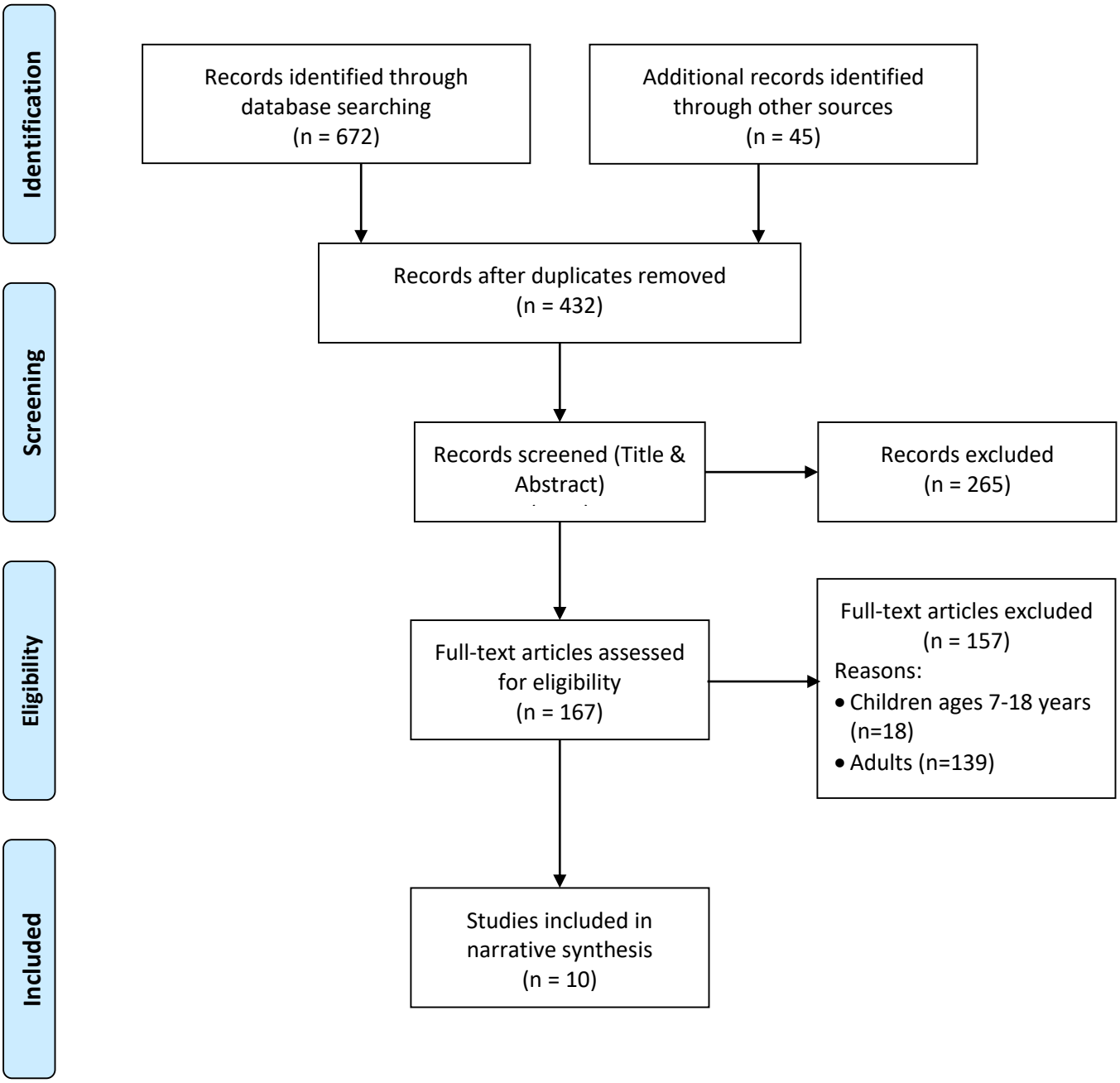


Figure 11. PRISMA flow diagram

Adapted from: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Medicine. 2009, 6(6): e1000097. doi:10.1371/journal.pmed1000097

5.3 Chapter 5 Appendices

5.3.1 Appendix 1. COSMIN measurement properties and domains

Measurement Properties		Definition
Reliability	Reliability	The extent to which scores for patients who have not changed are the same for repeated measurement under several conditions:
	Internal consistency	The degree of interrelatedness among the items
	Measurement error	The systematic and random error of a patient's score that is not attributed to true changes in the construct to be measured
Validity – The degree to which an instrument measures the concept(s) it purports to measure	Content validity	The degree to which the content of an instrument is an adequate reflection of the construct to be measured
	Construct validity	The extent to which scores on a particular instrument relate to other measures in a manner that is consistent with a theoretically derived hypothesis concerning the concepts that are being measured
	Criterion validity	The degree to which the scores of an instrument are an adequate reflection of a 'gold standard'
	Structural validity	The degree to which the scores of an instrument are an adequate reflection of the dimensionality of the construct to be measured.
	Cross-cultural validity	The degree to which the performance of the items on a translated or culturally adapted instrument is an adequate reflection of the performance of the items of the original version of the instrument
Responsiveness		The ability of an instrument to detect change over time in the construct to be measured

Adapted from COSMIN manual

5.3.2 Appendix 2. Quality criteria for measurement properties

Property	Rating	Criteria
Reliability		
Reliability	+	ICC / weighted Kappa ≥ 0.70 OR Pearson's r ≥ 0.80
	?	Neither ICC / weighted Kappa, nor Pearson's r determined
	-	Criteria for '+' not met
Measurement error	+	MIC > SDC OR MIC outside the LOA
	?	MIC not defined
	-	Criteria for '+' not met
Validity		
Content	+	All items are considered to be relevant for the construct to be measured, for the target population, and for the purpose of the measurement AND the questionnaire is considered to be comprehensive
	?	Not enough information available
	-	Criteria for '+' not met
Construct	+	At least 75% of the results are in accordance with the hypotheses
	?	No correlations with changes in instrument(s) measuring related construct(s) AND no differences between changes in relevant groups reported
	-	Criteria for '+' not met
Criterion	+	Convincing arguments that gold standard is "gold" AND correlation with gold standard ≥ 0.70 OR Sens+Spec ≥ 160[a]
	?	Not all information for '+' reported
	-	Criteria for '+' not met
Responsiveness		
	+	Correlation with changes on instruments measuring the same construct ≥ 0.50 OR at least 75% of the results are in accordance with the hypotheses OR AUC ≥ 0.70 AND correlations with changes in related constructs are higher than with unrelated constructs OR Sens+Spec ≥ 160[1]
	?	No correlations with changes in instrument(s) measuring related construct(s) AND no differences between changes in relevant groups reported
	-	Criteria for '+' not met

Modified from Terwee CB, Bot SD, de Boer MR, et al. Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007;60: 34-42
AUC = area under the curve; ICC = intraclass correlation coefficient; IRT = item response theory; LoA = limits of agreement; MIC = minimal important change; SDC = smallest detectable change; [a]not part of original COSMIN criteria; + = positive rating? = indeterminate rating, – = negative rating

5.3.3 Appendix 3. Criteria for overall strength of evidence for a particular CRAT

Level of Evidence	Rating	Criteria
Strong	+++ or ---	Consistent findings in multiple studies of good methodological quality or in one study of excellent methodological quality
Moderate	++ or --	Consistent findings in multiple studies of fair methodological quality or in one study of good methodological quality
Limited	+ or -	One study of fair methodological quality
Conflicting	±	Conflicting findings
Unknown	?	Only studies of poor methodological quality

5.3.4 Appendix 4. Sensitivity, specificity, positive predictive values and negative predictive values for current CRA tools for children, by study

Author, year	CRA tool	Sens	Spec	Sens+ Spec	PPV	NPV
Disney, 1992[a]	UNCCRA	0.59	0.83	142	0.48	0.88
		0.59	0.84	143	0.47	0.89
Beck, 1992[a]	UNCCRA	0.80	0.61	141	0.73	0.69
		0.66	0.78	144	0.57	0.84
Wandera, 2000[b]	UMCRA	0.76	0.75	151	0.59	0.86
Holgerson, 2009	Cariogram[c]	0.46	0.88	134	0.90	0.40
Gao, 2010	NUS-CRA^{S}	0.82	0.81	163	0.74	0.87
	NUS-CRA^{C}	0.90	0.90	180	0.85	0.93
	Cariogram	0.70	0.65	135	0.57	0.76
MacRitchie, 2012[24]	DCRAM1	0.67	0.57	124	0.59	0.64
	DCRAM2	0.53	0.77	130	0.68	0.63
	DCRAM3	0.69	0.60	129	0.62	0.66
	DCRAM4	0.65	0.69	134	0.66	0.67
Yoon, 2012	AAPD CAT	1	0.03	103	0.49	1
Gao, 2013	NUS-CRA^{S}	0.74	0.85	159	0.72	0.85
	NUS-CRA^{C}	0.78	0.85	163	0.73	0.87
	Cariogram^{S}	0.63	0.78	141	0.60	0.79
	Cariogram^{C}	0.64	0.79	143	0.62	0.80
	CAMBRA^{S}	0.94	0.44	138	0.47	0.93
	CAMBRA^{C}	0.84	0.63	147	0.55	0.87
	AAPD CAT^{S}	0.99	0.05	104	0.35	0.90
	AAPD CAT^{C}	1	0.04	104	0.35	1
Chaffee, 2016	CAMBRA	0.87	0.55	142	0.69	73.9
Custodio-Lumsden, 2016	MSB	NR	NR			

[a]top row estimates for Aiken, SC and bottom row for Portland, Maine the study sites for UNCCRA
[b]Re-categorised into 2 risk levels for sens and Spec computation – Moderate and High levels combined
[c]Modified cariogram for pre-school children
NUS-CRA^{S} = screening tool; NUS-CRA^{c} = comprehensive tool
DCRAM1 – predicting d1mft>0 (All caries lesions and predicting any caries)
DCRAM2 – predicting d3mft>0 (Dentine caries lesions and predicting any caries)
DCRAM3 – predicting d1mft≥3 (All caries lesions and predicting more than 2 caries lesions)
DCRAM4 – predicting d3mft≥3 (Dentine caries lesions and predicting more than 2 caries lesions)

5.3.5 Appendix 5. Content for CRA tools identified in this review

CRA tools	Factors/variables
CAMBRA, 2007 0-5 years old	**Risk indicators (parent interview)**
	Mother/caregiver: active caries in the past 12 months
	Child: recent restorations
	Parent/caregiver: low socioeconomic status and/or low health literacy
	Child: developmental problems
	Child: no dental home/episodic dental care
	Risk factors (biological) (parent interview)
	Child: frequent (> three times daily) between-meal snacks of sugars/cooked starch/sugared beverages
	Child: saliva-reducing factors (medications and medical)
	Child: continually uses bottle – contains fluids other than water
	Child: sleeps with a bottle or nurses on demand
	Non-biological protective factors (parent interview)
	Mother/caregiver: decay-free last three years
	Child: dental home and regular dental care
	Biological protective factors (parent interview)
	Child: lives in a fluoridated community or takes fluoride supplements by slowly dissolving or as chewable tablets
	Child: fluoridated toothpaste (pea-size) daily
	Mother/caregiver: xylitol chewing-gum/lozenges 2-4× daily
	Risk indicators/factors (child clinical examination)
	Obvious white spots, decalcifications, or obvious decay
	Restorations in the last 2 years
	Obvious plaque on teeth and/or gums bleed easily
	Dental or orthodontic appliances (fixed or removable)
	Visually inadequate saliva flow (dry mouth)
Cariogram	**Caries experience**
	DMFT, DMFS
	New caries experience in the past 1 year
	Related diseases
	Medical history
	Medications
	Diet, contents
	Diet history
	Lactobacillus test count
	Diet, frequency

CRA tools	Factors/variables
DCRAM, 2012	**Data collection at age 1 for prediction at age 4:**
	d1mft >0 – 'any caries–risk' model
	Health visitor opinion of risk
	Deprivation category
	Parental smoking
	Breastfeeding
	Use of a dummy
	d3mft >0 – 'any caries–risk' model
	Health visitor opinion of risk
	Parental smoking
	Food or drink at night
	d1mft – 'high caries–risk' model
	Type of housing
	Use of a feeder cup
	d3mft – 'high caries–risk' model
	Type of housing
	Health visitor opinion of risk
	Use of vitamins
AAPD CAT, 2015	**0-5 year- olds**
	Biological factors
	Mother/caregiver: active cavities
	Parent/caregiver: low socioeconomic status
	Child: >3 between meal sugar-containing snacks or beverages per day
	Child: put to bed with a bottle containing natural or added sugar
	Child: special healthcare needs
	Child: recent immigrant
	Protective factors
	Child: optimally-fluoridated drinking water or fluoride supplements
	Child: teeth brushed daily with fluoridated toothpaste
	Child: topical fluoride from health professional
	Child: dental home/regular dental care
	Clinical findings
	Child: active white spot lesions or enamel defects
	Child: visible cavities or fillings

	Questionnaire results, 24 h recall or dietary recall (3 days)		Child: presence of plaque
	Plaque amount (plaque index)		Child: >1 decayed/missing/filled surfaces
	Mutans streptococci		**Child: elevated mutans streptococci levels**
	Fluoride programme		
	Fluoride exposure		
CRA tools	Factors/variables	**CRA tools**	**Factors/variables**
NUS-CRA	Age	MySmileBuddy	Diet (for example, what did your child eat yesterday?) Feeding practices (for example, how often do you prechew your child's food?) Caregiver attitudes and beliefs (for example, how confident are you in reducing your child's risk for tooth decay) Fluoride use (for example, what type of toothpaste does your child most routinely use?) Family history (for example, have you ever had an abscessed tooth?)
	Ethnicity		
	Family socioeconomic status		
	Infant feeding history		
	Diet		
	Fluoride		
	Dental attendance		
	Oral hygiene		
	Past caries		
	Systemic health		
	Mutans streptococci		
	Lactobacilli		
UNCCRA	Race		
	Education – head of household		
	Brushing frequency		
	Fluoride mouth rinse		
	Dental visit		
	Fluoride tablets		
	dmfs/DMFS		
	Morphology score		
	White spot lesions		
	Mutans streptococci score		
	Lactobacilli score		
	Predicted caries score		
	Examiner reliability in scoring caries		

Adapted: *from Domejean S, Banerjee A, Featherstone JB, Caries risk/susceptibility assessment: its value in minimum intervention oral healthcare. British Dental Journal. 2017; 223(3):191-197*.

Chapter 6: Research Strand 2: The Calibration Study

6.1 Preface

This chapter contains the manuscript contributing to research strand 2 of this PhD thesis. The manuscript presented here is a draft of the paper before it went through the peer-review process. The final version has been published in the journal *Community Dental Health* and a pdf version of the published paper is included in the appendices (Appendix 4. Research Strand 2). The research presented in this chapter is the first of the two experimental parts of this PhD study. Chronologically, this research study was required to determine the quality of the VicGen data before it could be used to test the CAMBRA CRAT in research strand 3 (Chapter 7). It is aligned with the second objective of the PhD research study – to demonstrate the methodological process required to generate the evidence to inform the selection of a CRAT for use in practice.

Citation: *Christian B, Amezdroz E, Calache H, Gussy M, Sore R, Waters E. Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical. Community dental health. 2017;34(4):248-53.*

6.2 The Publication

Title: Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical.

6.2.1 Abstract

Aim: To study caries detection using different methods for the purpose of examiner calibration in populations and settings where clinical (in vivo) calibration is not practical.

Methods: Study design was cross-sectional and fully-crossed. The units of analysis were 880 tooth surfaces, from ten children aged 3 to 4 years. The study

had three data components: (1) Examiner training and calibration using the ICDAS e-Learning programme; (2) In vivo community-based visual examination; and (3) Intra-oral digital photographs of the same tooth surfaces from the in vivo visual examination. Dental caries was assessed using the International Caries Detection and Assessment System. Kappa and weighted kappa scores were used to study reliability estimates. Systematic differences in caries assessments were determined using the Stuart Maxwell test. Data were analysed using STATA 13.1 and SAS 9.2.

Results: Weighted kappa scores for examiner reliability estimates in this in vivo component ranged from 0.50 to 0.66 and from 0.64-0.74, for inter- and intra-examiner reliability, respectively. Comparing caries detection for selected tooth surfaces between the two methods, it was observed that caries lesions detected in vivo were also detected on photographs, albeit with increased severity when using photographs. For example, of 46 tooth surfaces assessed as being sound in the in vivo examination, 22 (48%) of these were assessed as having caries when photographs were used as the diagnostic method.

Conclusions: From this research it appears that good quality photographs alone may be used for training and calibration among challenging populations or settings without adversely affecting data quality.

6.2.2 Introduction

Data quality in oral health research, through reliable and valid measurement of disease levels, is essential to ensure unbiased estimates are obtained (161). Validity (accuracy) is the ability of a test to distinguish between those with and without disease, commonly measured using sensitivity, specificity, positive predictive and negative predictive values. Reliability (precision) is the ability of the test to produce the same results when repeated (22). Standardization in caries assessments can be achieved by training and calibrating examiners (OHE). Training is where examiners are trained in examination protocols and; calibration is a process that allows measurements of validity and reliability to be determined by, comparing to reference standards (validity), and repeated scoring by the same and different examiners (intra- and inter-examiner reliability).

It is generally recommended that in vivo training and calibration exercises be conducted following photograph slide-based training to give trainee examiners the opportunity to apply their training to the in vivo setting where the clarity of a visual examination may not be as good as when compared to the photograph of a caries lesion. This step also provides the opportunity to study, discuss and evaluate their scoring of the in vivo presentation of caries lesions before data collection. An in vivo calibration exercise entails several examinations on the same subject by different examiners, at different time points, making it a rigorous process requiring maximum co-operation from study participants. Hence, while in vivo training and calibration is good practice, it may not always be possible for reasons such as timing of data collection, lack of resources, specific population limitations (very young children, people with a disability and the elderly). For example, calibration exercises and dental assessments among pre-school aged children can be demanding on the child, parents/caregivers and examiners. Children, particularly under five years, are more temperamental, unpredictable and reactive to their environment, thus making examinations more difficult to conduct (10). To ensure data quality in dental research, when using in-vitro methods for training and calibration of examiners, it is imperative that these methods be studied to provide evidence justifying their use. Demonstrating validity and reliability of such methods would benefit future dental research by making the training and

calibration process more efficient. For example, if intra-oral photographs demonstrate good reliability and validity in caries detection, future studies may need only use this method for dental examiner training and calibration, thus, eliminating the need for multiple examinations on the same subject.

Several studies have investigated the use of intra-oral photographs as a caries diagnostic tool and fewer its validity (162, 163). A series of studies comparing intra-oral photographs with visual in vivo clinical assessments for caries detection demonstrated good comparability between the two methods (164-166). While these studies demonstrated good reliability, the unit of analysis was overall caries experience score (dmft) per study participant, which does not lend itself to interpretation of the actual caries diagnosis at the tooth or surface level. In addition, most research in this space has focused on caries diagnosis at the level of cavitation (dentine lesions) which does not recognize early non-cavitated stages of the disease process, which are particularly important from a prevention and health promotion perspective (167). Other gaps in this literature include: most studies being in vitro; caries assessments at the dentine level; and limited research in pre-school aged children and to our knowledge no comparative studies in a community-based setting (168).

The purpose of this research was to study caries diagnosis using different diagnostic methods for the purpose of examiner calibration in populations and settings where clinical (in vivo) calibration is not practical. Objectives:

1. The purpose of this objective was to answer the question: Can photographs alone (without an in vivo training component) be used in the training and calibration of examiners without adversely affecting data quality? This was achieved by assessing inter- and intra-examiner reliability in diagnosing the severity of caries lesions at the surface level, using: (a) Photographs of caries lesions sourced from the ICDAS e-Learning training programme; and (b) In vivo community-based visual examination of children's teeth.
2. To assess systematic bias in in vivo caries diagnosis.

3. To compare caries diagnosis using intra-oral photographs of selected tooth surfaces with their corresponding in vivo community examination scores; to assess systematic differences in caries diagnosis.

6.2.3 Materials and methods

Ethics approval for this study was provided by the Human Research Ethics Committee at the University of Melbourne (Ethics ID 1442648).

Study design and setting

This study was cross-sectional and fully-crossed in design. A fully-crossed design uses the same examiners for all study participants, therefore allowing for systematic bias to be assessed and controlled for in the reliability estimate (130).

Study participants were a sub-sample of 3- to 4-year-olds from the VicGen cohort of children residing in Melbourne, Australia. This age range was chosen for the full complement of primary teeth. The VicGeneration (VicGen) study is an oral health birth cohort study which began in 2008 to gain an understanding of the multifactorial nature of early childhood caries (ECC) development (128).

Sample size and participants

Based on estimates from the VicGen cohort and with the intention to set the minimum acceptable weighted kappa level as 0.61, an estimated sample size of 50 teeth (250 surfaces) were required to detect a weighted kappa score of 0.87 (95% CI: 0.64, 1.00). The sample size calculation was performed by manipulating the estimated distribution of ratings until a sufficiently narrow 95% confidence interval was obtained. For this study, 880 tooth surfaces from 10 children were analysed. Ten children were purposively selected (based on caries activity) and invited to participate in this study. This selection process ensured presence of caries lesions to reflect a range of ICDAS caries codes.

Data collection

Data collection involved three components: (1) Examiner training and calibration using the ICDAS e-Learning programme; (2) In vivo community-based visual

examination; and (3) Intra-oral digital photographs of the same tooth surfaces from the in vivo visual examination.

Components (1) and (2) relate to objective one, which was to determine whether sufficient reliability was achieved in the in vivo assessments, when training and calibration was conducted using photographs from the ICDAS e-Learning programme (169). Components (2) and (3), relate to objective three (Tables 5 & 6), where the in vivo assessments for selected surfaces were compared with their corresponding intra-oral photograph scores to determine whether caries lesions detected in vivo would also be detected on photographs; because if this was the case, it means that photographs are as sensitive as the visual in vivo examination. The argument for conducting in vivo calibration exercises following training is that lesion presentation is different between in vivo and in vitro caries detection; and that in vitro calibration alone can negatively affect data quality (unreliable caries detection) of the in vivo assessments (data collection) for a particular study.

Three dental examiners were used in this study. Caries was diagnosed and scored using the International Caries Detection and Assessment System (ICDAS) (131). ICDAS codes (Table 6) were used due to their ability to measure the progressive nature of the caries process as compared with other systems such as World Health Organization (WHO) and BASCD methods, which only record dentinal lesions (cavities) (170-172). Air-drying of teeth, using compressed air, was not part of the diagnostic process, as this was considered impractical in the community setting. This lack of air drying prevented the use of ICDAS caries code 1 on smooth surfaces. The in vivo dental examinations were exclusively visual without the use of a dental probe.

ICDAS e-Learning programme training and calibration

The 90 Minute e-Learning programme by the ICDAS Foundation was used to train examiners in the application of ICDAS (169). An experienced clinician and researcher (HC), facilitated the training session. This was followed by a discussion of the scoring criteria, until agreement on the reference standard ICDAS code was achieved, and a calibration exercise. For the calibration, examiners scored four

sets of eight photographs each (32 photographs in total) of the various ICDAS codes, sourced from the e-Learning programme. Examiners were blinded to each other's scoring. Scoring was repeated to measure intra-examiner reliability.

In vivo community-based visual dental assessments

Each participant received two visits one week apart. At each visit the study participant had a dental assessment done by each of the three OHEs in the participant's home. The OHEs were blinded to each other's assessment. Five-minute breaks were provided between examinations for each child. In total each child had six assessments.

Children were examined in a community setting with the child lying down on a flat surface with the OHE situated behind the child. The examination involved standardised light sources (Henry Schein MirrorLites) and infection control protocol. A mouth mirror was used to view the teeth and gauze was used to remove tooth debris where required. The child's teeth were divided into quadrants and examined in a standardised manner. A trained research assistant was on site to record the caries scores onto paper forms which were then double entered into an electronic database and checked for data entry consistency, to ensure data quality.

Intra-oral photographs

Intra-oral digital photographs of all tooth surfaces in children participating in the community examination were taken for comparison to the corresponding in vivo examination scores. These photographs were taken at either the first or second community visit depending on the participant's convenience. All the photographs were taken by the same OHE experienced in the use of the intra-oral camera through a previous research study and trained in the examination protocol. The Sopro Acteon 717 intra-oral camera was used to take the photographs. The camera had its own light source and the following settings: intra-oral mode, colour images and output format as JPG. The protocol for taking intra-oral photographs was similar to the in vivo examination. The camera was connected to a computer and the images captured digitally and stored directly to an electronic folder with

the ID number corresponding to the particular child. This data was backed up onto an external hard drive at the end of the community day.

For the analysis, 71 photographs were chosen based on clarity and to reflect a range of ICDAS codes. These photographs were scored by the same three examiners who participated in the community in vivo examinations. A standardised environment was provided for examiners to score the photographs, which included a PowerPoint presentation of the photographs projected onto a screen with the examiners seated approximately 3 meters from the screen. The room lighting was dimmed for the duration of the photo presentation and each photograph was viewed for approximately 20 seconds. One examiner was involved in photograph selection and collation. To avoid bias this examiner's scores were not used.

Data analysis

The unit of analysis for this study was the tooth surface. A child with a complete primary dentition of 20 teeth will have 88 tooth surfaces in total. Five tooth surfaces for each posterior tooth (molars – 8 teeth in total) and four tooth surfaces for each anterior tooth (incisors and canines – 12 teeth in total). Though both ICDAS codes were recorded, the focus for this analysis were the ICDAS caries codes.

Kappa and weighted kappa scores were used to determine inter- and intra-examiner reliability for tooth surface level scoring of caries lesions. Weighted kappa is the recommended analysis technique for determining examiner reliability for ordinal data such as ICDAS caries codes (0 (sound surface) – 6 (extensive decay)). This statistic incorporates the factor of agreement by chance alone and also has a feature of weighted proportional agreement (173, 174). The 'kapci' command in STATA was used to generate kappa scores and 95% confidence intervals (95% CI), with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95% CI. A suggested guideline for Kappa scores: Under 0.20=poor; 0.21-0.40=fair; 0.41-0.60=moderate; 0.61-0.80=good and 0.81-1=very good reliability (132).

Clustering effects (surfaces within teeth and teeth within individuals) were explored and found to be minimal and hence, estimates did not require adjustment for clustering (133, 134).

To assess systematic bias in scoring caries severity the Stuart-Maxwell test for marginal homogeneity was conducted. This tests for the marginal homogeneity across all ICDAS categories simultaneously and determined if examiners were systematically scoring differently from other examiners or within themselves. Data were analysed using STATA 13.1 and SAS 9.2.

6.2.4 Results

Reliability estimates for the two caries diagnostic methods are provided in Table 7. Using the e-Learning photographs, Kappa scores for inter-examiner reliability ranged from 0.56 to 0.70 (moderate to good). In the in vivo community examination, using ICDAS caries codes only, weighted Kappa scores for inter-examiner reliability ranged from 0.50 to 0.64 (moderate to good).

Systematic differences in scoring caries lesions, in the in vivo community examination, were assessed in Tables 8 and 9. Testing for marginal homogeneity between the three examiners showed that in some instances statistically significant ($p<0.05$) differences were observed, both between and within examiners. From Table 8, it appears that examiner 2 was scoring differently to examiners 1 and 3, as determined by the non-significant p-value of 0.330 for examiner 1 against 3. This difference in caries diagnosis is shown in the marginal distribution of ICDAS scores by examiners in Table 9.

Tables 10 and 11, show results comparing caries diagnosis of the same tooth surfaces using two methods: In vivo community-based dental examination and intra-oral photographs of the same teeth. To assess systematic differences in caries diagnosis between the in vivo visual examination and intra-oral photograph methods, the scores for the two methods from one examiner were cross-tabulated (Table 10). A statistically significant difference was observed between the two methods (Stuart-Maxwell $p=0.003$), where more caries lesions were diagnosed

from photographs. For example, of the 46 tooth surfaces diagnosed as being sound (ICDAS code 00) in the in vivo examination, only 24 (52%) of those were diagnosed as sound using photographs. The remaining 22 experienced caries, of which 16 were non-cavitated caries lesions (ICDAS code 02). Studying the distribution of these 22 tooth surfaces (Table 11), it was observed that the majority of diagnostic differences between methods occurred on posterior teeth (17/22).

6.2.5 Discussion

This methodological paper explored issues with examiner calibration in settings where in vivo calibration is not practical. The findings from this study present an interesting scenario. On the one hand, moderate to good reliability, in in vivo caries diagnosis, was achieved when training and calibration was conducted using photographs. On the other hand, more caries lesions were detected from intra-oral photographs when compared with in vivo diagnosis on the same tooth surfaces. Two important lines of discussion arise from these observations: (1) the training & calibration of dental examiners; and (2) caries detection, both of which are key to collecting quality epidemiological data.

As stated earlier, calibration using participants in epidemiological studies is a rigorous process and may not always be possible because of the timing of data collection, a lack of resources and/or specific population limitations. In such cases, alternative methods such as photographs and extracted teeth have been used, without an in vivo component, which may raise the issue of data quality. In vivo data quality (reliability and systematic bias), which is affected by factors such as: magnification, presence of saliva and debris on the tooth surfaces, lack of appropriate equipment and behaviour of the participant, is rarely assessed following the use of in vitro calibration methods. Similarly, quality of intra-oral photographs is affected by factors such as: inability to move camera head to a different angle to confirm diagnosis, image size, resolution of the computer screen, distance of camera head from tooth, number of teeth per photograph and difficulty in accessing particular tooth surfaces, such as the lower lingual surfaces of posterior teeth.

This study found that moderate to good reliability can be achieved in in vivo caries detection in the community setting when training and calibration was conducted using photographs only. However, when the in vivo scores were assessed for systematic bias, it was observed that one examiner scored differently to the others. This may have been the result of the presentation of caries lesions in vivo. Though the difference was statistically significant, whether it was clinically significant is questionable. This is because the difference was mainly observed in the scoring of non-cavitated lesions and sound surfaces, ICDAS caries codes 0-2.

Assessing systematic bias was a unique feature of this study and it is recommended that this becomes common practice when reporting calibration estimates. From this preliminary research into calibration methods for community-based epidemiological studies it appears that photographs alone may be used for training and calibration, without adversely affecting the data quality.

Though there is no standard classification for the Kappa score, it is generally accepted in the statistics literature that Kappa scores of over 0.5 are an indication of moderate to good reliability (132). The kappa estimates in this study were similar to those estimates reported in the paper on the development of ICDAS (131), in spite of being conducted in community conditions and in a challenging population.

Reliability estimates are influenced by the diagnostic method used as well as by disease prevalence. The more stages of disease you have the more chances you have for disagreement. A system like ICDAS, which has 7 stages of caries disease process is more prone to examiner disagreement compared with the WHO dmf/DMF index which has only 1 disease stage – cavitation. Hence, the reason for WHO dmf/DMF achieving Kappa scores of 0.8. This paper utilises ICDAS and not the WHO DMFT, in light of evidence that (1) the caries disease process is characterised by several distinct stages of progression not captured by the WHO dmf/DMF assessment of caries lesions; (2) non-cavitated lesions are reversible; and (3) disease management should be based in primary prevention of the disease and not mechanical repair of existing pathology (53). Hence, in modern times, the

WHO DMF/dmf is becoming obsolete. It should be noted that caries prevalence/~90% caries free in this paper (Table 9) refers to the proportion of tooth surfaces without any previous caries experience and among a sample of pre-school aged children it can be expected that the majority of surfaces will be caries free. Similar caries prevalence scores for tooth surfaces were also reported among Brazilian preschool children (175).

Comparing the in vivo caries assessments with their corresponding intra-oral photograph assessments, this study showed that caries lesions detected in vivo were also detected using photographs, by the same examiners, with a trend towards increased caries severity when using photographs. From a calibration perspective, it means that photographs are not missing out those lesions detected using the in vivo method; which shows that while in vivo is the ideal calibration method, photographs may also be used without adversely affecting data quality. The photographs used in this analysis component were selected for their clarity and re-examined, following the analysis, to ensure the absence of introduced artefacts such as the intra-oral camera light being reflected off the tooth surface. In the absence of artefacts and assuming that the photographs were valid, the better examination conditions and magnification when using photographs could be the reason for more caries being detected using this method. This finding raises the issue of validity of caries detection methods, particularly clinical (visual) versus photographs. From a calibration point, it means that photographs are not missing those lesions detected using the in vivo method, which indicates that while in vivo is the ideal calibration method, photographs may also be used without adversely affecting data quality. The 71 photographs used in this component of the analysis were selected for their clarity and again examined following the analysis result to ensure the absence of introduced artefacts such as the intra-oral camera light being reflected off the tooth surface. In the absence of artefacts and assuming that the photographs were valid, the better examination conditions when using photographs could be the reason for more caries being detected using this method.

The visual and visual-tactile methods continue to be the standard for caries detection in the clinic. These have also been used as the gold standard in validation studies (166, 167). However, the strength of evidence to support the validity of this method is poor, calling into question its use as the gold standard (176). When studying the validity of caries detection methods, histological sectioning of a tooth for visual evidence of caries activity is considered the most appropriate gold standard, even though it has limitations (176). A recent study, comparing the validity of occlusal caries assessments using in vivo visual and intra-oral photographs, with histology as the reference standard, demonstrated that the use of intra-oral photographs was the more sensitive method (162). Median sensitivity was 81.3% for the photographs compared with 65.6% for the in vivo visual method. Specificity for both methods was similar at 81.3% and 82.4%, respectively. However, these validity assessments may be overestimated since the visual assessment and photographs were performed on extracted teeth without the challenges of the in vivo oral examination. In addition, caries was assessed at the dentine level, which excluded early non-cavitated caries lesions and this may have also contributed to the high specificity estimates (162). Nonetheless, the evidence shows that good quality photographs are a more valid method for caries detection and further research is required to improve the strength of evidence for this method, particularly in relation to non-cavitated caries lesions.

Our findings of intra-oral photographs detecting more caries lesions compared to in vivo visual examination are consistent with results reported by Boye and colleagues (162). This was particularly the case for non-cavitated enamel lesions in the posterior primary molars, where the in vivo visual examination failed to detect 50% of enamel lesions. The in vivo examination is influenced by factors such as presence of saliva and debris on the tooth surfaces, accessibility to tooth surfaces and participant behavior, which in the case of very young children can be challenging. In addition, the collection of epidemiological community data is affected by lack of appropriate equipment and lighting. These factors together probably contributed to the in vivo examination detecting fewer carious lesions.

For epidemiological caries studies and oral health programs focused on prevention, it is important to detect lesions before they become cavitated (45). In such cases, it appears that the use of intra-oral photographs to detect caries offers several advantages over the commonly used visual-tactile method for caries detection: (1) more sensitive caries detection method, where intra-oral cameras are used in the community setting for caries detection; (2) cost savings, a trained research assistant could take the photographs and have the disease diagnosed and assessed at a later time by a dental practitioner; (3) multiple examinations can be performed on the photographs without burdening the participants; (4) reduced anxiety for participant as a result of the absence of the power dynamic a dental practitioner brings to the examination; and (5) a database of intra-oral photographs being available to verify any discrepancies in the assessment, which can also be used for training and calibration of new examiners. These advantages will have to be weighed up against disadvantages such as: cost of the cameras, training and standardisation of personnel, participant compliance, accessibility to the tooth surfaces, photographs providing only a two-dimensional picture and electronic data management.

A limitation of this study was the number of examiners. More than three examiners are generally recommended to achieve the highest score on a quality rating scale (166). It should be noted that the use of the modified ICDAS method (no air drying) eliminates ICDAS caries code 1 on the smooth surface and those in the pits and fissures that would have only been visible after drying. As such, this would cause an underestimation of the true population caries estimate. However, for this study on reliability estimates (kappa scores) and systematic differences (Stuart-Maxwell tests), the results would remain unchanged as the caries assessment protocol used was similar across detection methods.

This research highlights the important role of clinical photographs as a tool for calibration of examiners in caries detection and assessment for dental research. It could be particularly useful for future epidemiological surveys where access to appropriate equipment and lighting may not be available. It also highlights the relative lack of research on calibration methods in oral health. More research is

required to determine best practice for examiner calibration in oral health, such as: larger studies testing different combinations of calibration techniques to determine the most effective and efficient method.

Recommendations from this research: (1) Photographs may be used to calibrate examiners, particularly in challenging population sub-groups, (2) Systematic bias should be assessed in all calibration exercises before data collection commences and where required further training and discussion be conducted to eliminate the bias, (3) Intra-oral photographs be used in caries detection for studies/programs where prevention is the primary aim, and (4) Further research be undertaken to determine best practice methods of calibration.

6.2.6 Conclusions

To conclude, it appears that good quality photographs alone may be used for training and calibration, without adversely affecting data quality. Also, intra-oral photography could be a more sensitive method for caries detection over in vivo visual examination, with promising application in epidemiological studies. More research is required to determine best practice for examiner calibration in oral health, such as: larger studies testing different combinations of calibration techniques to determine the most effective and efficient method.

Table 6. ICDAS Codes

Treatment Codes		Caries Codes	
0	Sound	0	Sound surface
1	Sealant, partial	1	First visual change in enamel
2	Sealant, full	2	Distinct visual change in enamel
3	Tooth coloured restoration	3	Enamel breakdown: no dentine visible
4	Amalgam restoration	4	Dentinal shadow (not cavitated into dentine)
5	Stainless steel crown	5	Distinct cavity with visible dentine
6	Porcelain or gold or PFM crown or veneer	6	Extensive distinct cavity with visible dentine
7	Lost or broken restoration		
8	Temporary restoration		

Table 7. Estimates (95% CI) for inter- (at time 1) and intra-examiner reliability

Examiners	1	2	3
1	0.96 (0.88-1.00)[1] 0.60 (0.54-0.66)[2] 0.74 (0.65-0.81)[3]	0.60 (0.41-0.80) [1] 0.55 (0.48-0.61) [2] 0.52 (0.40-0.63) [3]	0.70 (0.54-0.89) [1] 0.54 (0.47-0.60) [2] 0.64 (0.51-0.74) [3]
2		1.00[1] 0.65 (0.58-0.71) [2] 0.64 (0.51-0.74) [3]	0.56 (0.35-0.76) [1] 0.54 (0.48-0.61) [2] 0.50 (0.37-0.63) [3]
3			0.96 (0.88-1.00) [1] 0.56 (0.49-0.63) [2] 0.66 (0.52-0.77) [3]

[1] Kappa scores from the ICDAS e-Learning training photographs (n=32 photographs)
[2] Kappa scores (all ICDAS codes) in vivo community dental examinations (n=880 tooth surfaces)
[3] Weighted Kappa scores (ICDAS caries codes only) in vivo community dental examinations (n=880 tooth surfaces)

Table 8. Stuart-Maxwell test for marginal homogeneity to assess systematic differences in the in vivo assessment of caries lesions, both between and within examiners

Examiners	1	2	3
1	P<0.001	P=0.030	P=0.330
2		P=0.093	P=0.020
3			P=0.444

Table 9. Marginal distribution of ICDAS caries codes by examiner from the in vivo community-based dental examination

ICDAS Caries Codes (n=880)	Examiners		
	1	2	3
00	89.5%	89.2%	91.0%
01	0.5%	0.1%	0.1%
02	6.4%	8.0%	6.2%
03	1.1%	0.9%	0.5%
04	0.9%	1.3%	0.6%
05	0.8%	0.2%	0.8%
06	0.5%	0.1%	0.5%
Column totals	100%	100%	100%

Table 10. Cross tabulation (marginal frequencies) of caries diagnosis (ICDAS codes) using in vivo visual examinations and intra-oral photographs of the same tooth surfaces for one examiner

In vivo ICDAS Scores	Intra-oral Photograph ICDAS Scores												
	00	01	02	03	04	05	06	30	32	34	36	84	Total
00	24	0	16	0	3	2	0	1	0	0	0	0	46
01	0	0	2	0	0	0	0	0	0	0	0	0	2
02	1	0	5	1	0	0	0	0	1	0	0	1	9
03	0	0	3	1	0	0	1	0	1	0	0	0	6
04	0	0	0	0	0	0	0	0	0	0	0	0	0
05	0	0	0	0	0	2	0	0	0	0	0	0	2
06	1	0	0	0	0	1	0	0	0	0	0	0	2
30	0	0	1	0	0	0	0	0	0	0	0	0	1
32	0	0	0	0	0	0	0	0	0	2	0	0	2
34	0	0	0	0	0	0	0	0	0	0	0	0	0
36	0	0	0	0	0	0	0	0	0	0	1	0	1
84	0	0	0	0	0	0	0	0	0	0	0	0	0
Total	26	0	27	2	3	5	1	1	2	2	1	1	71

Stuart Maxwell test for marginal homogeneity: p=0.003

Table 11. Distribution of 22 (of the 46) tooth surfaces that were diagnosed as sound (ICDAS code 00) in the in vivo examination but carious when using photographs in Table 5, by tooth location and surface

Tooth location	Tooth Surfaces					Total
	Distal	Occlusal[1]	Mesial	Buccal	Lingual	
Anterior	0	n/a	1	2	2	5
Posterior	2	6	0	5	4	17
Total	2	6	1	7	6	22

[1] Anterior teeth did not have an option to score occlusal surfaces and hence not applicable (n/a)

Chapter 7: Research Strand 3: CRAT Testing

7.1 Preface

This chapter contains the draft manuscript, prior to peer-review, contributing to research strand 3 of this PhD thesis. The manuscript has been published in the *Journal of Dentistry*. The published paper is included in the appendices (Appendix 5. Research Strand 3). The research presented in this chapter is the second of the two experimental parts of this PhD study. The systematic review on CRATs for young children (Chapter 5 of this PhD thesis), identified the lack of methodological studies to provide evidence on the quality of the measurement properties (reliability and validity) for CRATs as a major knowledge gap. This knowledge gap was addressed in this chapter through the second objective of the PhD research study – to demonstrate the methodological process required to generate the evidence to inform the selection of a CRAT for use in practice.

Citation: *Christian B, Calache H, Adams G, Hall M, Dashper S, Gibbs L, Gussy M. A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children. Journal of Dentistry. 2020;95:103324.*

7.2 The Publication

Title: A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children.

7.2.1 Abstract

Objectives: To assess the CAMBRA caries risk assessment tool (CRAT) in terms of its measurement properties: reliability, validity and responsiveness to change in caries estimates over time.

Methods: Secondary data from the VicGeneration (VicGen) birth cohort study were used. Caries risk status for child at 18 months of age was compared against caries development at 36 and 48 months. Questionnaire data for 18-month-old children,

were used to generate the CRAT item responses. Five examiners assessed the caries risk profile for each 18-month-old child and assigned a risk status. The analytic method was informed by the COnsensus-based Standards for the selection of health Measurement Instruments Checklist.

Results: Each examiner applied the CAMBRA CRAT to 214 cases from the VicGen study. At 18 months of age, the proportion of low-, moderate- and high-risk children were 17%, 17% and 66%, respectively. Caries prevalence at 36 and 48 months of age was 18% and 35%, respectively. Almost perfect reliability (both inter- and intra-reliability) was achieved in caries risk categorisation. For criterion validity and responsiveness, the area under the curve was approximately 0.50. For the dichotomised risk, the sensitivity of the CAMBRA CRAT to predict caries at 36 and 48 months, was 74% and 70%, respectively. The corresponding specificity scores were 35% and 36%, respectively.

Conclusions: The results from this study show that the CAMBRA CRAT, in its current form, is not a good predictor of future caries among young Victorian children and in fact overestimates the proportion of children at high-risk of future disease. As such, it may not be ideal for use among young Victorian children and it is highly likely its use could be driving over treatment.

Clinical Significance: This study provides evidence for the applicability of the CAMBRA CRAT with young Victorian children to inform decision making on caries risk-based management. It also highlights the important measurement properties and related analytics (applicable to any CRAT) that should be considered when selecting a CRAT for use in clinical practice.

7.2.2 Introduction

Dental caries in children is common. In the deciduous dentition the disease can manifest as early as the first year of life and continue until exfoliation (177). The factors contributing to caries risk in the early years (0-5 years) have been shown to differ from those in older children (51). This is understandable given the rapid changes in diet, tooth development and oral hygiene practices as a child progresses through developmental stages from infant to preschool age. The well substantiated influences on caries in young children are: parenting behaviours, knowledge, beliefs and attitudes; infant feeding practices; maternal circumstances and oral health; and infant-related oral health behaviours (5, 51). Understanding the age specific influences on child oral health and using this knowledge to inform the design and implementation of interventions (including service delivery), has greater potential to translate to effective action, compared with blanket (broad spectrum) interventions to improve oral health or service delivery across all ages.

A recent study showed that caries prevalence, among children in Victoria, Australia, increased from 8% to 23% between 18 and 36 months of age, with the development of new lesions on previously sound tooth surfaces (45). It is evident from the study by Gussy et al. (45), that the time period between 18 and 36 months is a window of opportunity for early intervention and disease prevention. However, to act effectively during this window of opportunity, at-risk children need to be accurately and precisely identified. Valid and reliable tools that discriminate children truly at risk of dental caries from those not at risk are required.

Risk-based management using CRATs is promoted as a way of: identifying those at highest risk of disease, guiding treatment decisions, guiding discussions with patients (including parents/caregivers) regarding caries risk/protective factors, determining appropriate recalls and to inform the economics of producing, delivering and using health care (27). However, whether risk-based management is achieving these outcomes for either the patient or the health service organisation is unclear. From the information/evidence in two recent systematic reviews of the measurement properties (reliability and validity) of CRATs (21, 136), it can be extrapolated that current CRATs used in risk-based management may in

fact result in overtreatment and as such are wasteful of resources. This has major implications for public/community dental services with limited resources and where risk-based management is used as a mechanism to ensure appropriate resource allocation, so that people with the most need get the required care. Hence, research is urgently required to provide the evidence to demonstrate that risk-based management and CRATs are an efficient and effective method to improve oral health and health service outcomes.

Numerous caries risk assessment tools (CRATs) are currently being used in clinical dental practice with the popular ones being: Caries Management by Risk Assessment (CAMBRA), American Academy of Pediatric Dentistry CRAT and Cariogram (18-20). These CRATs are a collation of various caries risk assessment methods (CRAMs), that have been compiled into prediction models, with the assumption that together the CRAMs will be able to predict future disease better than any single method alone. As stated previously, the evidence that CRATs accurately predict future disease is unclear and in fact may be resulting in overtreatment and wastage of resources. Commonly used CRAMs include socio-economic status, dental service use, diet and oral health behaviours.

The ability of a CRAT to perform well by correctly and consistently identifying those individuals at risk of future disease is affected by the quality of two core measurement properties – reliability and validity. Reliability is the property of the CRAT to produce similar results under different conditions, whereas validity relates to the degree to which the CRAT actually measures what it was designed to measure. Reliability includes the domains of internal consistency and measurement error. Validity includes the domains – content, construct, and criterion validity. Responsiveness, a relatively new addition to these classic measurement properties, is the ability of an instrument to detect change in the construct over time (124).

Evidence for the quality of measurement properties of CRATs is required to inform tool selection for use in practice, however, current research on the topic is very limited (2, 21). To investigate this topic in-depth, we conducted a comprehensive

systematic review on the quality of measurement properties for current CRATs for young children (136). This review identified a major knowledge gap by demonstrating the lack of methodological studies to provide the evidence to assess the quality of the measurement properties for the identified CRATs.

The authors of this study aimed to address the knowledge gap by conducting a methodological study to assess the measurement properties for the CAMBRA CRAT. Specifically, the objectives were to assess the CAMBRA CRAT in terms of its: reliability in application, validity in predicting future caries in young children, and responsiveness to change in caries estimates over time. The CAMBRA CRAT for children under 6 years of age was selected for use in this study, as it showed strong evidence for content validity (136); and although not the best performing CRAT in our recent review, is currently (in its original or modified versions) the most commonly used CRAT across many public dental services in Victoria, Australia. The CAMBRA CRAT was used as a case study and the method demonstrated in this study could be used as a road map for testing other CRATs.

7.2.3 Materials and methods

The University of Melbourne Human Research Ethics Committee approved this study (Ethics ID 1851421).

The CAMBRA CRAT was assessed using a secondary analysis of data from the VicGeneration (VicGen) study, an oral health birth cohort study to understand the multifactorial nature of early childhood caries development [13]. Seven waves of data collection occurred in VicGen, at child ages (months) 1, 6, 12, 18, 36, 48 and 60; and each wave included a clinical dental examination, saliva collection and a self-administered questionnaire (25). Dental caries was assessed visually (no probe) and recorded using the International Caries Detection and Assessment System (ICDAS-II) (131). The ICDAS-II protocol was modified to suit the field setting by omitting air-drying of teeth and as such ICDAS-II caries code 1 on smooth surfaces of examined teeth could not be detected. All clinical examiners were trained and calibrated with good examiner reliability in caries diagnosis (178). Structured self-administered questionnaires were used to collect data

(caregiver self-reported) on: family socio-demographic-economics; child feeding habits; child and caregivers' oral hygiene-related behaviour; self-reported oral/general health status for child and care-giver; care-giver's knowledge and attitudes about oral health. A more comprehensive description of the VicGen study methods has been published previously (25).

Based on the data collected in the VicGen study, the premise of this study was to assign a caries risk level to the 18-month-old children prior to the onset of the caries disease process (clinically apparent demineralisation of the dental hard tissue) and validate this risk assignment against future caries lesion development in these children. Questionnaire data for 18-month-old children were used to generate the CAMBRA CRAT item responses. This process gave each 18-month-old child a completed CAMBRA caries risk profile where each CRAT item had a response. Five examiners with a background in clinical dentistry assessed the caries risk profile for each 18-month-old child and assigned a risk status (high-, moderate-, or low-risk). For the reliability analysis, risk assessment data from all the clinical examiners were used. The risk assessments were a desktop exercise. Caries risk status of child 18 months of age was compared against actual caries development of child 36 and 48 months of age, as reported in the VicGen data.

The analysis method for this study was informed by the COnsensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist (124). Though the COSMIN checklist was developed to evaluate the methodological quality of studies on measurement properties of Health-Related Patient-Reported Outcomes (HR-PROs), it can be applied to other health-related measurement instruments, such as CRATs, as the design requirements and measurement properties are similar (124).

Sample size considerations

To achieve an excellent rating for the quality of a measurement property, the COSMIN checklist recommends a sample size of at least 100 cases. This study included 214 cases which had complete data across the three time points – 18, 36 and 48 months of age.

CAMBRA CRAT items and VicGen variables

As the CAMBRA CRAT items did not have specific definitions, the author team defined, jointly discussed and agreed on the VicGen variables that best represented the CAMBRA CRAT items. In some cases, two or more VicGen variables were aligned with the CRAT item and in such cases composite variables of the two (or more) variables were generated. For CRAT items where there wasn't a clearly aligned VicGen variable, logic and evidence-informed assumptions were used to derive the appropriate variables. The CAMBRA CRAT items, the corresponding VicGen variable(s) at 18-months of age and the assumptions used to derive the CAMBRA CRAT item variables are shown in Table 12.

Data cleaning and management

STATA was used to clean and manage the VicGen data, and in creating the analysis datasets. Four separate datasets were used to create the master dataset – the 18-month questionnaire data and the clinical data for 18, 36 and 48 months of age. The master dataset was then used to derive the variables for the CAMBRA CRAT items based on the 18-month questionnaire data. Once the CAMBRA variables were generated, another dataset with only the child unique identification numbers and the CAMBRA CRAT items was created. This CAMBRA dataset was used for the next stage of the research – developing the online caries risk assessment forms and the risk assessment process.

REDCap (Research Electronic Data Capture Version 9.1.21) hosted at the author's institution, a secure web platform for building and managing online databases and surveys, was used to create the web-based caries risk assessment forms and manage the risk assessment process and data (135). VicGen data were imported into RedCap and a CAMBRA CRA form was created for each child, which had responses for each CRAT item. The examiners then completed the risk assessments in REDCap, based on the CAMBRA item responses, and categorised each case as high-, moderate- or low-risk for future dental caries.

Caries risk assessments

Five examiners (four examinations as BC + HC jointly examined to establish the reference standard) with a background in clinical dentistry were trained to apply the CAMBRA CRAT using the online CAMBRA Basic training course available at https://course.mycambra.com/courses/cambra-basic/. The caries risk assessments were conducted in REDCap, where the examiner was required to determine whether a child was high-, moderate- or low-risk based on yes/no responses to the CAMBRA CRAT items (which were pre-populated into the CAMBRA CRAT REDCap form). Caries risk was summated based on the balance of risk vs protective factors – high, if more risk than protective factors; moderate, if equal numbers of both risk and protective; and low, if more protective than risk factors.

Two examiners (HC and BC) established the reference standard caries risk status at 18 months of age for each child, by jointly assessing 25 cases to achieve consensus on the scoring method, which was followed by an independent assessment of a further 50 cases where any discrepancies in the assessments were resolved via discussion. Once consensus was achieved, BC assessed the rest of the cases to establish the reference standard risk categorisation for the 18-month-old children. This reference standard was used in the validation analysis. To reflect real world conditions in current practice, examiners were not calibrated in using the CAMBRA CRAT.

Data analysis

On completion of the caries risk assessment stage of the research, the data were exported from REDCap into STATA (version 15.1, StataCorp, College Station, TX, USA) for analyses. Eighteen-month-old children who had a clinical examination at 36 and 48 months of age were included in this analysis (n=214). All children were caries free at 18-months of age. In this analysis, caries/caries lesion refers to any clinically apparent sign of tooth demineralisation [non-cavitated and cavitated lesions (ICDAS-II scores 1-6)]. A sub-analysis was also conducted to determine which CAMBRA CRAT items were responsible for the increased high-risk classification.

Reliability and measurement error

A fully-crossed design was used, where the same examiners were used for all study participants, therefore allowing systematic bias to be assessed and controlled for in the reliability estimate (130). Both inter- and intra-reliability estimates were calculated. For inter-examiner reliability, three examiners and the reference standard examination (BC + HC joint examination) assessed all 214 cases (blinded to each other's assessment) and assigned each case to a risk category (high-, moderate- or low-risk) based on responses to the CAMBRA CRAT items. For intra-examiner reliability, the first 100 cases were re-assessed by all examiners a few days after the first round of assessments. Reliability estimates were reported as weighted kappa scores and 95% confidence intervals (95% CI) and computed using the 'kapci' command in STATA with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95% CI. To assess systematic bias i.e. measurement error in the caries risk assessments, the Stuart-Maxwell test for marginal homogeneity was conducted to determine if examiners were systematically scoring differently from other examiners or within themselves.

Content validity

The content validity of the CAMBRA CRAT was assessed in our recently published systematic review on the quality of measurement properties for current CRATs for young children.

Construct (discriminatory) validity

Correlation estimates were computed between caries risk status at 18 months of age and clinical evidence of caries at both 36 and 48 months of age. It was hypothesised that being categorised as high-risk at 18 months of age would be correlated with the greater likelihood of developing caries lesions at the two future time points, compared with being low- or moderate-risk. As the variables used to assess this measurement property were ordinal, Spearman's rank coefficients were used to estimate correlation between variables. The distribution of children with and without caries by risk status was also explored as part of the assessment for this measurement property.

Criterion validity

The reference standard for this measurement property was caries development at 36 and 48 months of child age. For the classic CAMBRA CRAT categorisation of low-, moderate- and high-risk (no extreme risk), the area under the curve (AUC) was estimated. In addition, for comparison with other studies on this topic, the risk categories were dichotomised into high-risk versus low-risk (low and moderate risk categories were combined) and the following measures reported – sensitivity, specificity, positive predictive values and negative predictive values.

Responsiveness

The only difference between criterion (concurrent) validity and responsiveness is that criterion validity refers to the validity of a single score, and responsiveness refers to the validity of a change score. The change score for this analysis was the percentage of disease-free children at 36 months who developed caries lesions at 48 months of age. The analyses conducted were the same as for criterion validity.

7.2.4 Results

Five examiners applied the CAMBRA CRAT to 214 cases from the VicGen birth cohort study. At 18 months of age, the proportion of children categorised as low-, moderate- and high-risk were 17%, 17% and 66%, respectively. The prevalence of caries at 36 and 48 months of age was 18% and 35%, respectively. Results of the sub-analysis showed that the CRAT items responsible for the higher proportion of high-risk categorisations were related to the child's dental service utilisation – 'Child: no dental home/episodic dental care' and 'Child: dental home and regular dental care'. The following results are presented by measurement property.

Reliability and measurement error

The trained examiners achieved almost perfect reliability (both inter- and intra-reliability) in caries risk categorisation using the CAMBRA CRAT (Table 13). There was no evidence of a measurement error either between or within examiners.

Discriminatory validity

Figure 1 shows the proportions of children with caries by risk status at 36 and 48 months, respectively. There was some evidence of a correlation between the caries risk assessment at 18 months and actual caries development at 36 months of age (Spearman correlation coefficient = 0.13, P = 0.06), but not at 48 months of age (Spearman correlation coefficient = 0.08, P = 0.27).

Criterion validity

For the CAMBRA three-level risk categorisation (low, moderate and high), at 36 and 48 months of age, respectively, the AUC is close to 0.50, which indicates a low ability of the CRAT to predict future caries development. Table 14 shows the measures of criterion validity for the dichotomised risk categorisation (high-risk vs low-risk) and caries development at 36 and 48 months. The sensitivity (95% CI) of the CAMBRA CRAT to predict caries at 36 and 48 months, was 74% (58%, 87%) and 70% (58%, 80%), respectively. The corresponding specificity scores were 35% (28%, 43%) and 36% (28%, 44%), respectively.

Responsiveness

The measures to determine the responsiveness of the CAMBRA CRAT to the change in caries development between 36 and 48 months (Table 14), followed a similar pattern to those for criterion validity with sensitivity and negative predictive values achieving the highest scores, 64.8% and 73.6%, respectively. The AUC was 0.48.

7.2.5 Discussion

The results of this study show that the CAMBRA CRAT in its current form may not be useful in this population. An obvious observation from the analysis was the higher proportion of children (66%) designated as high-risk at baseline (18-months of age) compared with the proportion that developed caries lesions at 36 (18%) and 48 (35%) months of age. Gao et al. (146), made a similar observation in their validation study and reported that the majority of children were considered high-risk when the CAMBRA CRAT was used. The sub-analysis on this issue revealed that the CRAT items related to child's dental service utilisation were driving the

higher proportion of high-risk categorisations – 'Child: no dental home/episodic dental care' and 'Child: dental home and regular dental care'. To investigate whether the omission of these two items improved validity estimates, the full analyses were replicated on a dataset where these two items were removed (not reported in this paper because the modification did not improve criterion validity). Though this modification to the CAMBRA CRAT reduced the proportion of 18-month-old children assessed as being at high-risk (dropped from 66% to 51%), it did not improve estimates of validity. We therefore report results based on the original CAMBRA CRAT with the child dental service utilisation variables included. However, in very young children the utility of 'child dental service use' items as a risk indicator should be further investigated. Systematic reviews of risk factors in the early years do not show child dental service use as being associated with early childhood caries (50, 51). The recent paper by Chaffee et al. (150), validating the CAMBRA 0-5 CRAT and its individual items did not include the CRAT items for dental service use (dental home) and as such the use of this indicator as a CRAT item for very young children is not clear.

Evidence for the reliability and measurement error in the application of CRATs is limited in the literature (2). The examiners in this study demonstrated excellent reliability in the application of the CAMBRA CRAT to assign caries risk status to individual cases. During the training the examiners were instructed to be very objective in the application of the CRAT, i.e. rather than making subjective decisions on risk based on particular risk/protective factors/indicators, the examiners were instructed to assign risk based on the balance of 'yes' and 'no' responses to the CRAT items, as recommended in the CAMBRA training course. For example, three 'yes' responses to risk factors/indicators versus two 'yes' to protective factors/indicators tipped the balance towards being high-risk for future caries. Such objectivity in the risk assessments contributed to the excellent reliability scores. Reporting on the examiner's reliability in the application of CRATs has generally been ignored in the literature on CRAT development, and also in studies where CRATs were used in practice.

The reliability in risk assessments affects the reliability in allocating patients to care pathways and disease management, which in turn has implications for resources and cost. In public and community dental services, which operate on a tight budget, risk-based management is used as a method to control demand and waiting lists (using risk-based recall protocols) and to ensure the most urgent cases are prioritised for care. A lack of consistency among clinicians in applying the risk-based disease management protocols, particularly low-risk patients being incorrectly recalled too soon, will have significant implications on the services' ability to deliver positive outcomes and to ensure efficient use of its resources. Hence, one of the first steps in a risk-based model of service delivery is to ensure consistency in the risk-assessments and associated risk-based management pathways.

CRATs claim to be able to determine the likelihood of an individual developing future caries, i.e., the individual's risk (179). Future caries could be defined as either any sign of the caries disease process (non-cavitated or cavitated lesions) or more specifically related to the progression to cavitation. In this study the former definition was used. As mentioned in the methods section, the content validity for the CAMBRA CRAT was assessed in our previous systematic review (136). The CAMBRA CRAT achieved a strong rating for its content based on the information reported in the papers on CAMBRA's development (23, 150). However, it may be the case that the CAMBRA CRAT items are only valid for the particular population it was developed in and whether all the items are generalisable needs further investigation.

Studying the correlation of risk categories to future lesion development, a positive (non-significant) correlation was observed between the three risk categories at 18 months and caries lesion development at 36 months, but not at 48 months of age. Chaffee et al. (150), also reported a positive correlation for the CAMBRA CRAT, where increasing risk was strongly associated with more caries lesion development at follow-up. When reporting on the distribution of caries lesion development by risk categories, it is also important to simultaneously report on the distribution of healthy/disease free individuals by risk categories. It may be the

case that while the distribution of those people with caries increases with increasing risk, the distribution of healthy people does not decrease with increasing risk, as was observed in this study. The observed trend of more children with caries at 36 months of age in the high-risk category could be an artefact of the over categorisation as high-risk at 18-months-of age. That is, this finding could be mirroring the distribution of the high-risk categorisation rather than the CRAT being truly discriminative in identifying children with caries. The discriminant property of a CRAT is dependent on testing and reporting of such estimates, however; the literature on this measurement property for CRATs is relatively non-existent.

Criterion validity is the most studied and commonly reported measurement property for CRATs (2, 21, 136). A possible reason for the focus on criterion validity (while ignoring other measurement properties) is the view that CRATs are similar to medical diagnostic tests for which the validation method is well established, where a new test is validated against an established reference/gold standard; and tools developed to research diagnostic tests (such as the Quality Assessment for Diagnostic Accuracy Studies – QUADAS) have been adopted in CRAT research (24, 139). However, CRATs are made up of multiple caries risk/protective factors/indicators, with the rationale being that together these factors/indicators will improve outcome prediction compared with only using a single factor/indicator. As such, in the case of CRATs all the domains of validity (content, construct and criterion) require careful investigation. However, the methods to adequately test content and construct validity are relatively new and yet to be widely adopted in CRAT research.

In this study, for the CAMBRA CRAT risk levels (low-, moderate- and high-risk) the criterion validity analysis showed that the ability of the CAMBRA CRAT to predict caries, at 36 and 48 months among disease/lesion free 18-month-old children, was very low (the AUC was close to 0.50 for both ages). When the risk categorisation was dichotomised to high- and low-risk (moderate and low were combined) sensitivity the CAMBRA CRAT was between 70%-74%, while specificity was low at

approximately 35%. Similar findings of acceptable sensitivity but low specificity scores for the CAMBRA CRAT have been reported previously (146, 150).

Several issues need to be explored to help explain the low validity for the CAMBRA CRAT among these children. First, was the CAMBRA 0-5 CRAT developed with the intention that it would be predictive of caries lesion development in young children who currently had no sign of the disease process (i.e. sound tooth surface)? The answer to this specific question is probably 'yes' as the main paper on the CAMBRA CRAT for young children, states the following "To achieve the best management and outcomes for good dental health, an appropriate caries risk assessment screening must be executed as early as possible and preferably prior to the onset of the disease process." [23, p. 688]. The original consensus statement on CAMBRA in 2002, clearly states that CAMBRA was developed out of a clear need to manage the underlying causes (risk factors) for dental caries "rather than just the results of the disease" [20, p. 258], in an attempt to maintain the balance between the protective and pathologic factors (137, 180).

Second, the definitions for each of the CRAT items are not explicitly stated in the CAMBRA background papers. For example, how was the CAMBRA item 'Parent/caregiver: low socioeconomic status and/or low health literacy' to be determined? The reason could be that the CAMBRA developers wanted to be adaptable to any situation or population. However, in the absence of definitions, the authors of this study used their own interpretation and evidence to derive the CRAT items from the VicGen data, as shown in Table 12. It is possible that the authors' item definitions were different to those used in the development of CAMBRA and as such could be a reason for the low validity estimates in this study.

Third, the use of the three risk-levels (no extreme risk as special needs and medically compromised children were excluded from the VicGen study) in very young children who have no signs of the disease process, may be diluting the predictive capacity of the CRAT. A better option, as shown in this study, could be to have only two risk-levels – high/at-risk and low-risk. Interestingly, the two other studies that report on criterion validity for the CAMBRA CRAT both report a similar

dichotomised risk (146, 150), with the emphasis being on predicting caries among high-risk children. Another reason for dichotomising risk in this study was our observation of clinical practice data showing that clinicians tend to categorise most people as moderate risk (the safe option), which in terms of caries management triggers shorter recall periods and more intense preventive services being provided, when they were not required i.e. no effect on oral health outcomes. This again goes back to the crux of the problem, that current CRATs result in over-treatment and wastage of resources. By dichotomising the risk assessment scores we are likely to reduce the possibility of over treatment and wastage of resources.

Fourth, the predictors of dental caries among young children in Victoria, Australia may not be the same for children in San Francisco, USA, where CAMBRA was developed and tested. In fact, a recent cross-sectional study among migrant pre-school aged children in metropolitan Melbourne, Australia, reported length of residence in Australia as an independent predictor of caries (181). Results from our recently published longitudinal analysis of predictors for early childhood caries among Victorian children, also using data from the VicGen birth cohort, shows a relatively different mix of independent risk and protective factors, compared with those currently in the CAMBRA CRAT (120). The University of North Carolina Caries Risk Assessment studies (UNCCRA) also clearly demonstrated the variation in independent predictors of dental caries, by population (142, 143). Understanding the drivers of disease in a specific population is very important, as it highlights the potential risk of adopting CRATs without testing them for relevance to the population being served.

Finally, the time between the risk assessment and first signs of the disease process may be long enough to allow for the risk/protective factors from the original assessment to have changed. For example, it is well-known that feeding practices are quite dynamic in the early years (182, 183), so a risk assessment at 18-months of age may only be valid up to that age where the risk/protective factors remain the same. Future research should study the time-based nature of caries risk profiles. It should also be noted that the reliability in diagnosing the clinical

outcome can be a potential source of variability in the validation of risk categorisation and as such should be reported in studies on caries risk assessments. The clinical examiners in VicGen were shown to have moderate to good reliability in diagnosing dental caries using ICDAS and as such we do not believe this was a major source of variability in the CRAT assessment outcomes for this study.

Responsiveness is a relatively new measurement property that has not been explored in previous studies on this topic. Technically it is measured and assessed like criterion validity, however the main difference between the two is that criterion validity refers to the validity of a single score, and responsiveness refers to the validity of a change score (124). In this study, the ability of the CRAT to predict the number of new cases of caries between 36 and 48 months, was used to assess responsiveness. This is an important measurement property that attests to the tool's ability to detect change overtime and not just at a specific point in time. The results and reasons for the results were similar to that discussed for criterion validity and will not be elaborated on further.

An obvious limitation of our analysis was the use of data that were not collected specifically for the CAMBRA CRAT items. As a result, the created variables may not be the same as those used in the development of CAMBRA. However, given that the assumptions were extensively discussed with experts in the field and where possible validated using evidence, the authors believe that the generated variables were the best representation of CAMBRA items for the Victorian child population. The ideal methodological study would have collected data specific to the CRAT of interest and then followed the cohort of children prospectively to measure caries lesion development. This method, however, is resource and time intensive and in most cases may not be feasible. The use of secondary data is the more viable option, and, in such cases, variables derivation should be guided by discussion and evidence-informed assumptions.

7.2.6 Conclusions

Caries risk assessment tools are being increasingly used in new models of health care for various reasons including (but not limited) to: identify those at highest risk of disease, inform disease management plans, support patient self-management regarding caries, determine appropriate recall periods (and thus manage waitlists) and inform the economics of health A major knowledge gap in the literature on CRATS was the lack of methodological studies to provide the evidence on the quality of their measurement properties, which in turn would inform their selection in practice. This study provides the evidence to show that the CAMBRA CRAT, in its current form, may not be ideal for use in risk-based disease management among young Victorian children. Due to its low specificity scores it is highly likely that the use of this CRAT in Victoria could be driving over treatment, which is a major issue, particularly in public dental services where risk-based service delivery is used as a resource allocation and waitlist management method, to prioritise care for those most at risk of future disease. An important next step on this topic of research is to investigate, at the practice/service delivery level, whether caries risk assessments are aligned with the appropriate management strategies.

Future research on the topic of CRATs should follow a similar methodological approach and endeavour to assess all relevant measurement properties, which will assist clinicians and decision makers in CRAT selection for use in practice.

Table 12. CRAT caries indicators/factors and the corresponding VicGen questions

CAMBRA CRAT Items	VicGen variables at child 18 months of age	CRAT item derivation and assumptions
Risk indicators (parent interview)		
Mother/caregiver: active caries in the past 12 months	Mother/caregiver caries estimates at child 6 months of age.	Caries lesion activity status was not recorded in VicGen. *Assumption:* A parent was assumed to have active caries in the past 12 months, if they were in the top one third (66^{th} percentile) of parents with the highest DMFT scores ('D' defined as caries into dentine). For this sample of parents, the DMFT score at the 66^{th} percentile was 9.
Child: recent restorations	Not applicable as no restorations at this age.	This CRAT item was excluded from the caries risk assessment.
Parent/caregiver: low socioeconomic status and/or low health literacy	What is the highest post school education that you have completed? Do you or your partner have a health care card?	For this CRAT item, a composite variable was created from the two VicGen variables. *Assumption:* Any respondent with less than an advanced diploma qualification was considered as having low health literacy. *Composite variable:* Any respondent with less than an advanced diploma qualification or who had a health care card.
Child: developmental problems	Has the doctor ever told you that your child has developmental problems?	*Derived variable:* Same as the VicGen variable.
Child: no dental home/episodic dental care	How often does your child have a dental check-up?	*Derived variable:* Same as the VicGen variable. *Assumption:* A response of 'never' or 'less than 1/year' was assumed to be evidence of 'no dental home/episodic dental care'.
Risk factors (biological) (parent interview)		
Child: frequent (> three times daily) between-meal snacks of sugars/cooked starch*/sugared beverages	Does this child drink any of the following? In a usual week how often does the child have the following foods?	For this CRAT item, a composite variable was created from the two VicGen questions. *Composite variable:* Child reported to consume >3 cariogenic foods or drinks per day. Decisions on which foods and drinks to include were informed by the published VicGen cariogenic scale paper [31]. Only food and drink classified as moderately cariogenic and higher were included for this composite variable.

Child: saliva-reducing factors (medications and medical)	Medically compromised children were not included in VicGen.	This CRAT item was excluded from the caries risk assessment.
Child: continually uses bottle – contains fluids other than water	Does the child sip from bottle on and off during the day? What is usually in the bottle?	For this CRAT item, a composite variable was created from the two relevant VicGen questions. *Composite variable:* Child sips from bottle on and off during the day, and it usually contains fluids other than water.
Child: sleeps with a bottle or nurses on demand	Does this child usually take a bottle to bed?	*Derived variable:* Same as the VicGen variable.
Non-biological protective factors (parent interview)		
Mother/caregiver: decay-free last three years	Mother/caregiver caries estimates at child 6 months of age.	*Assumption:* The mother/caregiver was assumed to be decay free the last three years if they had no decayed teeth ('D' component of the DMFT score) at child age 6 months. The 'D' was defined as an ICDAS caries score of 4-6. The evidence shows that it takes approximately 77 months for enamel lesions to progress to dentine [32]. Hence, those parents with large cavities were most likely not decay free in the past 3 years.
Child: dental home and regular dental care	How often does your child have a dental check-up?	*Derived variable:* Same as the VicGen variable. *Assumption:* A response of '1/year or more' was assumed to be evidence of 'dental home and regular dental care'.
Biological protective factors (parent interview)		
Child: lives in a fluoridated community or takes fluoride supplements by slowly dissolving or as chewable tablets	Water fluoridation status at the postcode where the child lived.	*Derived variable:* Water fluoridation status for the postcode of each case, approximated to the time of data collection when the child was 18 months of age.
Child: fluoridated toothpaste (pea-size) daily.	What type of toothpaste do you use for child? How much toothpaste do you use for child?	For this CRAT item, a composite variable was created from the two relevant VicGen questions. *Composite variable:* Child reported to use fluoridated toothpaste at least 1/day and a pea size amount of toothpaste.
Mother/caregiver: xylitol chewing-gum/lozenges 2-4 × daily	In the last week, how many times did you chew sugar-free gum for at least 10 minutes continuously?	*Derived variable:* Same as the VicGen variable.
Risk indicators/factors (child clinical examination)		

Obvious white spots, decalcifications, or obvious decay	Child ICDAS caries codes at 18 months of age.	This CRAT item was excluded from the caries risk assessment as all children were caries lesion free at 18 months of age.
Restorations in the last 2 years	Not applicable as no restorations at this age.	This CRAT item was excluded from the caries risk assessment as all children were caries lesion free at 18 months of age.
Obvious plaque on teeth and/or gums bleed easily	Child clinical examination at 18 months of age.	*Derived variable:* Visible plaque on anterior teeth. *Assumption:* If there is evidence of plaque on anterior teeth it was assumed that there was plaque on the posterior teeth too.
Visually inadequate saliva flow (dry mouth)	Not assessed in VicGen	This CRAT item was excluded from the assessment.

Table 13. Weighted Kappa scores (95% Confidence Intervals) for reliability and the Stuart-Maxwell test (*P=*) for marginal homogeneity in risk categorisation using CAMBRA

Examiners	1	2	3	4
1	0.98 (0.93-1.00)	0.97 (0.94-0.99)	0.96 (0.89-0.99)	0.98 (0.97-1.00)
	P=0.61	*P*=0.37	*P*=0.61	*P*=1.00
2		1.00	0.96 (0.88-0.99)	0.98 (0.96-1.00)
		P=1.00	*P*=0.22	*P*=0.37
3			1.00	0.97 (0.91-1.00)
			P=1.00	*P*=0.61
4				0.98 (0.92-1.00)
				P=0.61

Table 14. Criterion validity of the dichotomised (high-risk vs no-risk) caries risk assessment at 18 months of age, in predicting caries development at 36 and 48 months of age. Responsiveness of the CRAT in identifying children with caries at 48 months who were disease free at 36 months.

	Disease prevalence (%)	Sensitivity (%)	Specificity (%)	PPV (%)	NPV (%)
Criterion validity 36 months	18.2(13.2,24.0)	74.3(57.8,86.9)	35.4(28.3,43.0)	20.4(14.1,28.0)	86.1(75.9,93.1)
Criterion validity 48 months	34.5(28.2,41.3)	70.3(58.5,80.3)	35.7(27.8,44.2)	36.6(28.7,45.1)	69.4(57.4,79.7)
Responsiveness	**% of new cases at 48 months**	**Sensitivity (%)**	**Specificity (%)**	**PPV (%)**	**NPV (%)**
	25.2(19.5,31.6)	64.8(50.6,77.3)	33.1(25.9,40.9)	24.6(17.8,32.5)	73.6(61.9,83.3)

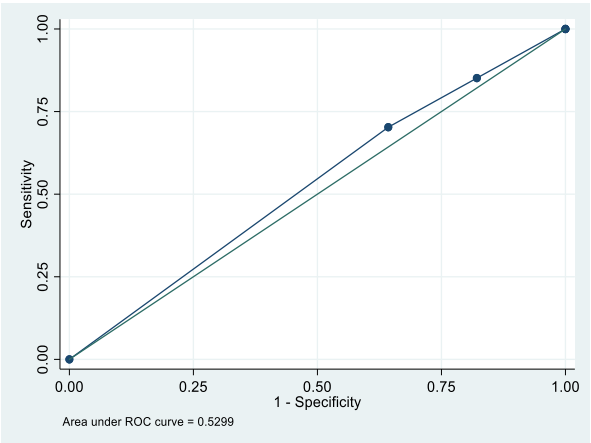
PPV-positive predictive value; NPV-negative predictive value

7.3 Chapter 7 Appendices

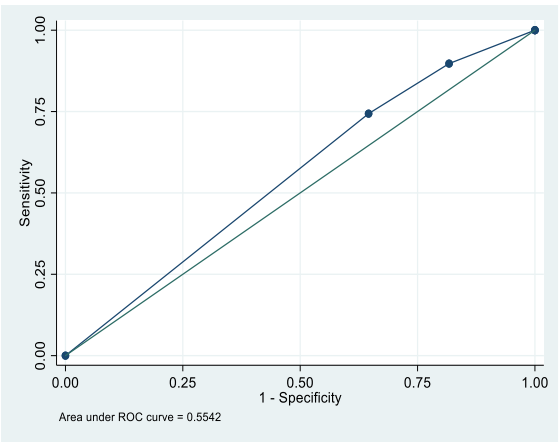
7.3.1 Appendix 1. COSMIN measurement property domains

Measurement Properties	
Reliability	*Reliability* in the application of the CAMBRA CRAT was studied, both within and between examiners.
	Measurement error was assessed as part of the reliability testing and used to quantify the error.
Validity	*Content validity* was assessed, in a systematic review, to determine if the included items in the CAMBRA CRAT were an adequate reflection of the construct being measured.
	Construct validity (Discriminatory validity) was studied to determine whether risk categorisations are discriminatory. This was done by studying correlations between risk categorisation at 18 months of age and actual caries development at 36 and 48 months of age.
	Criterion validity the degree to which the scores of a CRAT reflect a 'gold standard', was studied by comparing risk assessments at 18 months of age to actual caries development at 36 and 48 months of age (the gold standard).
Responsiveness	This is the ability of a CRAT to detect change over time and was assessed similar to criterion validity with the outcome being a change variable.

7.3.2 Appendix 2. Estimates of criterion validity, showing the area under the curve for the CAMBRA CRATs ability to predict caries development at child age 36 (2A) and 48 months (2B) by risk status (low, moderate or high).

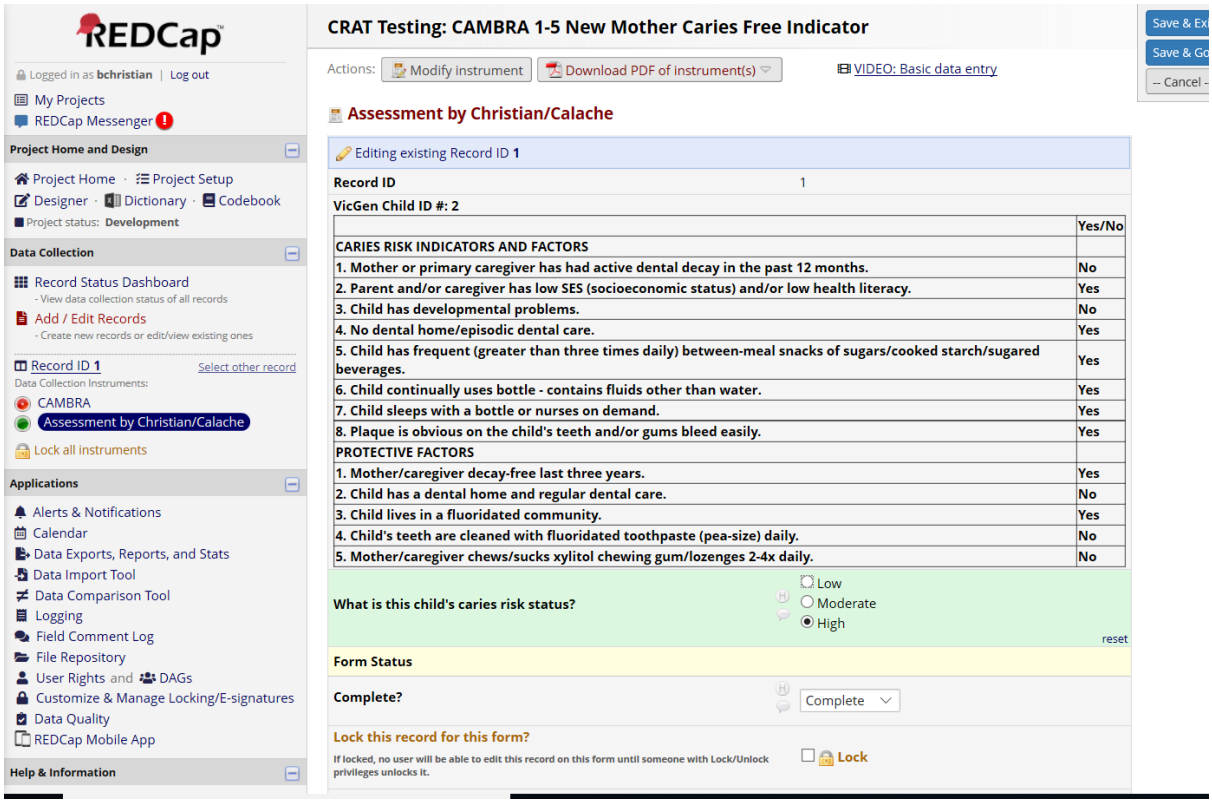


2A



2B

7.3.3 Appendix 3. Image, the CAMBRA caries risk assessment form in REDCap.



Chapter 8: Discussion

The findings from the three separate PhD research strands are brought/pulled together in this PhD thesis chapter as a unified program of research around a specific topic – the use of caries risk assessments tools (CRATs) to manage early childhood caries in young children. Presented in this chapter is a general discussion of the main findings of the PhD, specifically in the context of the conceptual frameworks and the philosophy of value-based health care presented in Chapter 3. The contributions to knowledge, and strengths and limitations are also presented. An in-depth discussion for each PhD research strand (Figure 1) was presented in their respective stand-alone peer-reviewed publications (Chapters 5, 6 and 7).

8.1 General Discussion

This PhD research generated a cohesive body of new knowledge on caries risk assessment tools (CRATs) for young children, to inform their selection and to support their utilisation in new risk-based models of oral health care. The research was framed as two objectives and conducted in three research strands (please see Chapter 1 for details). Data from the VicGeneration (VicGen) birth cohort study were used for the experimental strands (2 and 3) of the PhD (25).

The main findings from this PhD research were: Firstly, there was limited evidence in the scientific literature on the quality of existing CRATs to inform their selection for the management of early childhood caries. This was primarily due to the lack of methodologically sound studies to provide the required evidence. Second, the CAMBRA CRAT, which is widely used across public dental services in the State of Victoria in Australia, was shown to lack the ability to accurately identify/predict children that would develop dental caries. In fact, our research showed that the CAMBRA CRAT over-estimated the proportion of children at risk of caries, which could potentially be driving over-treatment and wastage of resources. Hence, while CRATs are important in caries management and can be used to discuss risk and

protective factors with the patient, which could support behaviour modification activities; given the current state of evidence demonstrated in this PhD, CRATs should not be used in treatment planning or for resource allocation, until their performance has been rigorously assessed in the intended population.

These findings from the PhD research raise some very important considerations for the utilisation of CRATs in ECC management but also for the field of cariology in general. The body of evidence on CRATs for use in ECC management needs to be enriched much further, before we can have confidence in their ability to perform as intended, i.e., predicting future disease accurately and reliably. Particularly, if the performance of the CRAT among a specific population or population sub-group, was not assessed prior to its use. The PhD findings clearly demonstrated that CRATs were reported to perform well only in populations in which they had been developed and tested (142, 143). The PhD research showed considerable variation in methodology used, across studies, to test CRATs. Of note, was the lack of methodologically sound studies to provide the evidence for their use. An observation that was also shared by Jørgensen and Twetman (2019), in their recent paper on caries risk assessment tools for young children (185); which was prepared for the International Association of Paediatric Dentistry Global Summit on Early Childhood Caries in November 2018. A recommendation from this Global Summit on ECC which is aligned with research on CRATs was “Research on ECC inequalities, oral health-related quality of life, interventions, and health economics should be supported to further understand benefits of effective and timely care” (186, p. 385). Thus, it is recognised at a global level that ECC continues to be a major public health issue and that further research is required in several domains (as stated in the previous sentence) to address the issue of ECC. While the findings from this PhD research contributed new knowledge towards the management of ECC as it relates to the use of CRATs, the testing of only one CRAT in one location (Victoria) was within the scope of this PhD. As shown in this PhD systematic review and other recent papers (136, 185, 187), there are a number of other CRATs in use among young children and urgent research is required to demonstrate that they work in their respective populations; using robust methods as demonstrated

in this PhD research. The ideal methodological study would collect data specific to the CRAT of interest and then follow the cohort of children prospectively to measure caries lesion development. This method, however, is resource and time intensive and in most cases may not be feasible. The use of secondary data is the more viable option, and, in such cases, variables derivation, for the CRAT risk indicators, should be guided by discussion and evidence-informed assumptions.

In Australia and globally, there is a major move towards and investment in value-based health systems (15, 76). Value in health care has been defined as the health outcomes that matter to patients relative to the resources or costs required to generate the particular outcome. In this age of big data and freedom of information, health care providers are increasingly being held accountable (by the patient, funders, regulatory orgnisations) for the value their services provide to the individual patients and the community. Besides the overarching ethos of improving health outcomes (rather than the volume/outputs), cost is major factor in the value equation (30). In public and government health systems, with limited resources, the value-based agenda is a mechanism by which the entire system can be re-oriented towards better health outcomes while containing costs (30). Risk-based disease management will form the backbone of value-based care. An important first step on this journey to a value-based system is to ensure the evidence to support the risk assessment process is strong; use evidence-based methods to select risk assessment tools; and where required, generate the knowledge to develop evidence-informed risk assessment tools.

As mentioned in Chapter 3, the philosophical underpinnings of this PhD research were informed by the value-based health care (VBHC) efforts of Porter and Teisberg (89, 90). Porter and colleagues, define value as the health outcomes that matter to the patients relative to the cost of achieving those outcomes (88). The VBHC agenda is a framework to operationalise the shift of the health care system towards value (88). A key aspect of the VBHC agenda is a risk-stratified high-value service provision (88). In an attempt to improve oral health outcomes, including the high burden of early childhood caries, public dental services in Victoria are re-

orienting towards risk-based models of oral health care and utlising the ideology of value (15, 16, 75). CRATs are used in these new models of care to ensure that children at a high-risk of developing dental caries get the necessary care to improve their oral health outcomes and reduce disease burden (in terms of health and cost to the inidividual and the community). However, the PhD findings clearly demonstrate the lack of evidence to support the use of current CRATs, particularly from the perspective of the VBHC agenda, where risk assessments are used as a means to improve operating efficiencies along with patient outcomes. The continued use of CRATs that are not supported by evidence, could potentially set off a perverse chain reaction where there is an increase in low value care without improvement in health outcomes. This creates inefficiency and wastage of resources which increase the costs of care, which in turn reduces the equitable distribution of those resources, especially to those that most need care. This in turn could worsen their oral health (and general health) outcomes of the most vulnerable individuals, families and communities.; which would be measured by increased hospitalisation as an indicator of disease burden and a failure of the system to successfully target those with high risk for oral disease. While a shift to the value ideology is commendable, it needs to be backed by rigorous research and evidence, or else we run the risk of breaking the system rather than improving it.

The complex interaction of various factors (often unmeasurable) at the population or individual level produces a specific health outcome. The two conceptual frameworks discussed in Chapter 3, posit how these various factors interact to cause a health state. The NICE conceptual framework for public health provides a more broad and generic theoretical perspective on the causation for public health outcomes (86); while the Fisher-Owens conceptual model influences and provides more detail on the specific drivers of child oral health (86). What these frameworks are really telling us is that we need to understand the causal pathways to a particular health outcome if we are to effectively intervene.

CRATs are made up of a collation of factors (risk/protective). Based on evidence of associations with oral heath outcomes, which are conceptualised to be on the causal pathway and together predict caries development in the future (2). The PhD finding that the CAMBRA CRAT was not predictive of dental caries among Victorian children clearly shows that we need to better understand the specific causal mechanism for caries development among young children in Victoria. Frameworks, such as the two used in this PhD should guide the development and testing of future CRATs, particulary around understanding the potential causal mechanisms occuring within specific population contexts. For example, in this PhD research the guiding frameworks need to be applied and jointly assessed from the perspective of two contexts: First, the context of the State of Victoria where the children reside and second, the context of very young pre-school aged children. Both contexts will have their own unique and inter-related drivers of tooth decay that need to be understood and acted on for effective intervention to prevent early childhood caries. The conceptual frameworks help theorise how these contexts may interact to produce dental caries, from which research hypotheses can be generated and validated. Going back to the key PhD finding that the CAMBRA CRAT was not useful among young Victorian children, shows that CRATs like any other health-related program or policy should be modified and adapted to the context in which it will be utilised. That is, one size does not fit all.

Our recent publication on predictors of dental caries in young Victorian children (120), which was conducted in parallel to Strand 3 of this PhD research and which also used data from the VicGen study, clearly demonstrated the process for utilising the Fisher-Owens conceptual framework to inform the theoretical foundations, including the intent, motivation and expectations for the research (120).

A key aspect of CRATs not discussed in the published papers is the use of disease indicators such as existing caries lesions to predict future caries. All the CRATs identified through the PhD systematic review (Chapter 5, Appendix 5) included an indicator for existing caries lesions. The evidence in the literature shows that the

presence of caries lesions is the best predictor of future caries lesions (17, 24). In contemporary caries literature where caries is discussed as a disease of the individual (188), then the presence of any caries lesion is an indicator of a diseased state. As such, the presence of any caries lesions should automatically put the indivudal into a high-risk caries management strategy. We do not need a CRAT for the purpose of predicting future disease (including cavitated lesions) when the disease/ lesions already exist. Ideally, CRATs should be able to predict the development of caries lesions in people without visible signs of the disease (depending on the threshold level of defining the disease). While this is achieveable in young children, among older children and adults this may be more challenging as caries lesions are more or less ominpresent. In such cases, it is important to diagnose the activity status of the caries lesion - active or arrested lesion; particulary if using the presence of a caries lesion(s) to determine caries risk. An arrested lesion may be an indication that the person no longer has the disease. For this PhD research we excluded the children that already had signs of ECC, i.e., caries lesions at 18 months of age. In young children, the purpose of using a CRAT should be for primary prevention of disease and as such the use of disease indicators defeats the purpose of a CRAT. That is, to correctly predict those disease free children that are at-risk of developing caries in the future.

8.2 Contributions to Knowledge

The substantive original contribution to knowledge in the subject area of CRATs, made by this PhD body of research, is clearly demonstrated through the five peer-reviewed publications that formed the crux of this PhD. In this section of the chapter, the key contributions are highlighted.

8.2.1 Contribution to oral health practice

The evidence generated from this PhD has the potential for a direct and immediate impact on the delivery of oral health services, particularly for the State of Victoria, Australia. The two published background papers (Appendix 1 and 2) lay down the context for this PhD, make the case for new models of oral health care and propose a new risk-based oral health promoting model of oral health care (12, 16). The PhD

research strands, and associated peer-reviewed papers made a substantial contribution to the evidence on the topic of caries risk assessment tools. From a practice perspective the important contributions are:

- Identification of the CRATs in the published literature that specifically target young children;
- Recognition that the National University of Singapore CRAT achieved the highest quality rating and hence, at this point in time, is recommended as the best CRAT for use in young children;
- Demonstration that intra-oral clinical photographs alone are sufficient to train and calibrate examiners in caries diagnosis without any major impact on data quality, particularly in situations and settings where the recommended in vivo calibration exercise is not practical;
- Recognition that, for clinicians and service providers, the important measurement properties (reliability and validity) and related analytics (applicable to any CRAT) should be considered when selecting a CRAT for use in practice;
- Provision of evidence to show that the CAMBRA CRAT when used among young Victorian children overestimates the proportion of children at-risk for future disease, which can result in over-servicing and wastage of resources (of critical importance to resource constrained public dental services). An appropriate balance of servicing is required (i.e. not 'over-servicing' and not 'under-servicing' either), as under-servicing has its own consequences.

8.2.2 Contribution to research method

The PhD research highlighted the fact that research on caries risk assessments is limited, lacks methodological rigour and methods utilised are not relevant to the subject area. The absence of methodologically sound studies on this topic is particularly concerning. From the CRAT studies identified in Strand 1 of the PhD (the systematic review) it is obvious that there is a clear lack of understanding of the key measurement properties that are relevant to a CRAT; and how to

assess/analyse them (136). A CRAT is unique in that it is a combination of caries risk assessment methods (CRAMs) and includes clinical indicators of disease as well as patient reported items. As such, the application of a CRAT can be very subjective, especially when a CRAT only includes patient reported items, unless clear instruction is provided on how to assign risk. This can cause variability between and within clinicians in assigning risk i.e. reliability, which can cause serious inconsistencies in clinical practice and resource allocation. Interestingly, none of the studies on current CRATs reported on reliability estimates as part of the tool development and testing process (136). This variability in practice due to incorrect risk assessments is starting to be recognised and addressed, particularly across public dental services in Victoria.

Another limitation of current evidence on the topic of CRATs is the reporting of only criterion validity (the sensitivity and specificity of the tool), while ignoring content validity and construct validity, which are very important for a CRAT. The focus on criterion validity and associated analytics was most likely the result of replicating the method for testing and validating a CRAM. However, a CRAM is a single method (risk factor), for example, the frequency of consumption of sugary drinks as the method to predict future caries (189). In this example, there is only a single method and as such content validity is not a major concern. When it comes to CRATs, we have multiple CRAMs and a key component in the selection of the CRAT is to understand whether all the CRAMs (the content) that make up the CRAT are valid.

Hence, a clear knowledge gap was observed around the methods used to develop, test and select CRATs. The PhD research addressed this knowledge gap by identifying and applying a standardised method to assess the quality of current CRATs and to test the CAMBRA CRAT. The method used was informed by the COnsensus-based Standards for the selection of health Measurement Instruments – COSMIN Checklist (123-125, 141, 190). The COSMIN checklist was developed by a multidisciplinary team of experts in medicine, epidemiology, qualitative research, health service research and psychometrics; with the aim to improve the method

and process for the selection of health measurement instruments (159). While it is being widely adopted and popular in health research, its use in the oral health space is very limited. In fact, the COSMIN checklist was most likely used for the first time in oral health research through this PhD. The method demonstrated in this PhD can be used as a template to develop, test and inform the selection of CRATs. The PhD research has set the methods benchmark for any future research on this topic, which will hopefully lead to more methodologically sound studies on CRATs.

8.3 Strengths and Limitations

A strength of this research was the rigorous assessment of the literature on CRATs and testing of a CRAT, which was informed by the use of a standardised and evidence-informed instrument – the COSMIN checklist. This checklist ensured the use of a method that is appropriate to the type of health measurement instrument (CRAT) and which encompassed all aspects of the measurement properties (reliability and validity) of CRATs in assessment of quality. In addition to being used as a standard to assess methodological quality of CRATs, the suggested methods in the COSMIN checklist can also be used to guide the development and testing protocols for new health-related measurement instruments, as was used in research strand 3 to test the CAMBRA CRAT.

Previous research on this topic focused only on criterion validity and used methods which also only focused on criterion validity. For example, the QUADAS (Quality Assessment for Diagnostic Accuracy Studies) tool has been used in previous systematic reviews on caries risk assessment methods (139). However, as the name indicates QUADAS focuses only on the single measurement property of validity, with the domains mainly related to criterion validity. As a result, the quality of the other measurement properties was unknown, which could affect the performance of the CRAT when used in practice. A key strength of this PhD research was a holistic assessment of the all the relevant measurement properties. Another important strength of this research was the use of longitudinal

data to assess the performance of the CRAT, particularly in relation to its ability to predict a caries outcome.

The three main limitations of the PhD research were: First, the research focus was narrow and limited to very young children. While this is an important stage early in the life course for prevention and intervention, caries in the deciduous dentition continue to be highly prevalent; similar research needs to be urgently undertaken for older children and adults because at present the majority of caries risk assessments (particularly in public dental services) are being performed on people older than 6 years, i.e. when they start school. Second, the data used in strand 3, to assess the quality of the CAMBRA CRAT, were not collected specifically for the purposes of a CRAT assessment. As such the CRAT specific indicators had to be derived from the VicGen variables and this may have caused some misclassification error. However, given that the assumptions were extensively discussed with experts in the field and where possible, validated using evidence, the authors believe that the generated variables were the best representation of CAMBRA items for the Victorian child population. Finally, it is possible that participants in longitudinal cohort studies may present a selection bias as they could be more likely to try and prevent disease due to their study participation. However, given that the disease rates in the VicGen birth cohort mirrored both local and global estimates it does not appear that this was an issue in this PhD research.

Chapter 9: Conclusions, Implications and Recommendations

9.1 Conclusions

This PhD study identified a major knowledge gap in the current state of evidence on the subject of caries risk assessment tools (CRATs); which was the lack of methodologically sound studies to provide the evidence to inform decision making on tool selection for use in practice. The PhD study addressed this knowledge gap through a methodologically rigorous research study to demonstrate the analytic process required to generate the evidence to inform the selection of a CRAT. The CAMBRA CRAT which was used as the case study for the PhD research was shown to be ineffective in predicting a caries outcome among young Victorian children. Developing or selecting a CRAT or any risk assessment tool for use in practice needs to be guided by a rigorous research and development method.

9.2 Implications for Practice and Policy

The findings from this PhD research have very serious practice and policy implications for risk-based models of oral health care. These include:

- Risk stratification is at the heart of the new risk-based models of oral health care and also forms a core component of the value-based health care agenda (see PhD chapter 3) that is gaining momentum in Victoria, Australia. The risk assessment determines the care pathway, review and recall intervals. The findings from this PhD clearly demonstrate that the CAMBRA CRAT, which is popular across the world, is not suitable for use with young Victorian children. In fact, the evidence from this PhD shows that its use could be driving over-servicing. For public dental services, who use risk stratification as a strategy to ensure their limited resources are reaching those that need it the most, the use of a tool that has not been through a rigorous research and development process (as

demonstrated in this PhD study) can lead to wastage of resources without a positive impact on oral health outcomes.

- At many public dental services in Victoria, the policy to manage the waitlist is based on risk stratification. As mentioned in the previous point, if the process to select a CRAT for use is not informed by research and evidence, it can reduce the capacity of the service to direct resources to where they are most needed.
- For patients seeking care through private practitioners the use of untested CRATs can lead to increased over-servicing for unnecessary treatment. The issue of over-servicing can also lead to significant out-of-pocket expenditure for patients and unwanted impact on their overall health. Therefore, it is absolutely crucial for a selected CRAT to accurately determine the risk of the patient in developing disease, at the point in time when the risk status is being assessed, as risk status will alter throughout the life span. The issue of dynamic risk status needs further investigating to determine the appropriate stages for risk assessment.
- Dental caries data that is collected routinely at private and public dental practices are generally considered to be low quality and not suitable for research/evaluation purposes. The main reason is that training and calibration to ensure data quality and consistency is generally viewed to be an onerous process. The calibration study, research Strand 2 of this PhD, demonstrated that data quality can be maintained even with a simplified calibration process that uses only clinical photographs. In addition, the calibration study demonstrated the increased sensitivity of using clinical photographs in caries detection, which offers several advantages over the traditional in vivo visual tactile method such as: (1) greater sensitivity, (2) lower cost if a trained research assistant takes the photographs, (3) multiple examinations can be performed on the photographs without burdening the participants, (4) reduced participant anxiety as a result of the absence of the power dynamic a dental

practitioner brings to the examination and, (5) a database of intra-oral photographs can be created to verify any discrepancies in the assessment, which can also be used for training and calibration of new examiners. These advantages will obviously have to be balanced against disadvantages such as: the cost of the cameras, training and standardisation of personnel, participant compliance, accessibility to the tooth surfaces, photographs providing only a 2-dimensional picture and electronic data management.

9.3 Recommendations

1. Good quality research on CRATs is limited. It is recommended that any future research to develop, test, assess the quality of or report on CRATs, use a standard such as the COSMIN checklist to inform the study method and reporting. This is akin to using other standards such as the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) or STrengthening the Reporting of OBservational studies in Epidemiology (STROBE). The use of a standardised method and reporting framework will help strengthen any future research and also allow for comparability between studies and CRATs.
2. It is recommended that priority research and evaluation be undertaken at any organisation where CRATs or other risk assessment tools are being used, to determine if the CRAT(s): are appropriate for the population being served; the content is valid; can be reliably applied by oral health practitioners; can correctly discriminate between risk categories and can accurately predict a health outcome. That is to ensure the tool selection process has been informed by evidence. In the absence of such a rigorous CRAT selection method to demonstrate that it does what it was developed to do, it is recommended that CRATs be limited for use as a tool for discussing risk/protective factors with patients to inform behaviour modification activities.

3. It is also recommended that the quality of routinely collected caries data at public dental services, be improved by incorporating a simplified process for training and calibration of oral health practitioners into their clinical protocols and policies. Good quality, real-time data will be very important to generate evidence on CRAT performance at a particular health service.

References

1. GBD 2016 Disease and Injury Incidence and Prevalence Collaborators. Global, regional, and national incidence, prevalence, and years lived with disability for 328 diseases and injuries for 195 countries, 1990-2016: a systematic analysis for the Global Burden of Disease Study 2016. Lancet. 2017;390(10100):1211-59.

2. Tinanoff N, Baez RJ, Diaz Guillory C, Donly KJ, Feldens CA, McGrath C, et al. Early childhood caries epidemiology, aetiology, risk assessment, societal burden, management, education, and policy: Global Perspectives. 2019;29(3):238-48.

3. Do L, Spencer AJ, editors. Oral health of Australian children: the National Child Oral Health Study 2012–14. Adelaide: University of Adelaide Press; 2016.

4. Hooley M, Skouteris H, Boganin C, Satur J, Kilpatrick N. Parental influence and the development of dental caries in children aged 0-6 years: a systematic review of the literature. Journal of Dentistry. 2012;40(11):873-85.

5. Hooley M, Skouteris H, Millar L. The relationship between childhood weight, dental caries and eating practices in children aged 4-8 years in Australia, 2004-2008. Pediatric obesity. 2012;7(6):461-470.

6. Kruger E, Tennant M. Ten years of hospitalisation for oral health-related conditions in Western Australia: an unjust dichotomy. Australian Journal of Primary Health. 2016. ;22(2):153-158.

7. Seow WK. Early Childhood Caries. Pediatric Clinics of North America. 2018;65(5):941-954.

8. Listl S, Galloway J, Mossey PA, Marcenes W. Global Economic Impact of Dental Diseases. Journal of Dental Research. 2015;94(10):1355-1361.

9. Colak H, Dülgergil CT, Dalli M, Hamidi MM. Early childhood caries update: A review of causes, diagnoses, and treatments. Journal of Nature Science Biology and Medicine. 2013;4(1):29-38.

10. Aminabdi NA, Puralibaba F, Erfanparast L, Najafpour E, Jamali Z, Adhami SE. Impact of Temperament on Child Behavior in the Dental Setting. Journal of Dental Research, Dent Clinics and Dental Prospects. 2011;5(4):119-122.

11. Health Issues Centre. "Was I still on the waiting list?" A study about people waiting for public dental care. Melbourne: Health Issues Centre; 2009.

12. Christian B, Hall M, Martin R. A paradigm shift in models of oral health care: An example and a call to action. Family Medicine and Community Health. 2015;3(4):32-37.
13. Queensland Health. Changing models of care framework. Brisbane: Queensland Health; 2000.
14. Fejerskov O, Nyvad B, Kidd E, editors. Dental Caries: The Disease and Its Clinical Management. 3rd ed. West Sussex: Wiley Blackwell; 2016.
15. Hegde S, Haddock R. Re-orienting funding from volume to value in public dental services. Canberra: Deeble Institute for Health Policy Research; 2019. Contract No.: Deeble Institute Issues Brief no.32.
16. Hall M, Christian B. A health-promoting community dental service in Melbourne, Victoria, Australia: protocol for the North Richmond model of oral health care. Australian Journal of Primary Health. 2017;23(5):407-414.
17. Mejare I, Axelsson S, Dahlen G, Espelid I, Norlund A, Tranaeus S, et al. Caries risk assessment. A systematic review. Acta Odontologica Scandinavica. 2014;72(2):81-91.
18. AAPD. Guideline on caries-risk assessment and management for infants, children, and adolescents. Pediatric Dentistry 2014;35(5):E157-64.
19. Bratthall D, Petersson GH. Cariogram--a multifactorial risk assessment model for a multifactorial disease. Community Dentistry & Oral Epidemiology. 2005;33(4):256-264.
20. Featherstone JDA, S. M.; Anderson, M. H.; Berkowitz, R. J.; Bird, W. F.; Crall, J. J.; Den Besten, P. K.; Donly, K. J.; Glassman, P.; Milgrom, P.; Roth, J. R.; Snow, R.; Stewart, R. E. Caries management by risk assessment: consensus statement, April 2002. Journal of the California Dental Association. 2003;31(3):257-269.
21. Tellez M, Gomez J, Pretty I, Ellwood R, Ismail AI. Evidence on existing caries risk assessment systems: are they predictive of future caries? Community Dentistry & Oral Epidemiology. 2013;41(1):67-78.
22. Chattopadhyay A. Oral Health Epidemiology Principles and Practice. Sudbury, Massachusetts: Jones and Bartlett; 2010.

23. Ramos-Gomez FJ, Crall J, Gansky SA, Slayton RL, Featherstone JD. Caries risk assessment appropriate for the age 1 visit (infants and toddlers). Journal of the California Dental Association. 2007;35(10):687-702.
24. Senneby A, Mejare I, Sahlin NE, Svensater G, Rohlin M. Diagnostic accuracy of different caries risk assessment methods. A systematic review. Journal of Dentistry. 2015;43(12):1385-1393.
25. Johnson S, Carpenter L, Amezdroz E, Dashper S, Gussy M, Calache H, et al. Cohort Profile: The VicGeneration (VicGen) study: An Australian oral health birth cohort. International Journal of Epidemiology. 2017;46(1):29-30.
26. Victorian Auditor-General. Access to public dental services in Victoria. Melbourne; 2016.
27. Twetman S, Fontana M, Featherstone JD. Risk assessment - can we achieve consensus? Community Dentistry & Oral Epidemiology. 2013;41(1):e64-70.
28. Dental Health Services Victoria. Dental Health Services Victoria Melbourne: DHSV; 2016 [Available from: https://www.dhsv.org.au/.
29. Porter ME. What is value in health care? New England Journal of Medicine. 2010;363(26):2477-2481.
30. World Economic Forum. Value in healthcare: Laying the foundation for health system transformation. Geneva: World Economic Forum in collaboration with The Boston Consulting Group; 2017.
31. Oxilia G, Peresani M, Romandini M, Matteucci C, Spiteri CD, Henry AG, et al. Earliest evidence of dental caries manipulation in the Late Upper Palaeolithic. Scientific Reports. 2015;5(1):12150.
32. Online Etymology Dictionary. Caries [Available from: https://www.etymonline.com/word/caries.
33. Lynch CD, O'Sullivan VR, McGillycuddy CT. Pierre Fauchard: the 'father of modern dentistry'. British Dental Journal 2006;201(12):779-781.
34. Gest H. The discovery of microorganisms by Robert Hooke and Antoni Van Leeuwenhoek, fellows of the Royal Society. Notes Records of the Royal Society of London. 2004;58(2):187-201.

35. Fejerskov O, Nyvad B, Kidd E. 2 Dental caries: what is it? In: Fejerskov O, Nyvad B, Kidd E, editors. Dental Caries: The Disease and its Clinical Management. West Sussex: Wiley Blackwell; 2016. p. 1-21.
36. Longbottom CL, Huysmans MC, Pitts NB, Fontana M. Glossary of key terms. Monograms of Oral Science 2009;21:209-216.
37. Machiulskiene V, Campus G, Carvalho JC, Dige I, Ekstrand KR, Jablonski-Momeni A, et al. Terminology of Dental Caries and Dental Caries Management: Consensus Report of a Workshop Organized by ORCA and Cariology Research Group of IADR. Caries Research. 2020;54(1):7-14.
38. Moynihan P, Petersen PE. Diet, nutrition and the prevention of dental diseases. Public Health Nutrition. 2004;7(1a):201-226.
39. Keyes PH. Recent advances in dental Caries Researchearchearch. Bacteriology. Bacteriological findings and biological implications. International Dental Journal. 1962;12:443-464.
40. U.S. Centers for Disease Control and Prevention. Concepts of disease occurence Atlanta: U.S. Department of Health & Human Services; 2012 [Available from: https://www.cdc.gov/csels/dsepd/ss1978/lesson1/section8.html.
41. Lennon MA. One in a million: the first community trial of water fluoridation. Bulletin of the World Health Organization. 2006;84(9):759-760.
42.Chapko MK. Time to adoption of an innovation by dentists in private practice: sealant utilization. Journal of Public Health Dentistry 1991;51(3):144-151.
43. Drury TF, Horowitz AM, Ismail AI, Maertens MP, Rozier RG, Selwitz RH. Diagnosing and reporting early childhood caries for research purposes. A report of a workshop sponsored by the National Institute of Dental and Craniofacial Research, the Health Resources and Services Administration, and the Health Care Financing Administration. Journal of Public Health Dentistry 1999;59(3):192-197.
44. Gibbs L, Waters E, Christian B, Gold L, Young D, de Silva A, et al. Teeth Tales: a community-based child oral health promotion trial with migrant families in Australia. BMJ Open. 2015;5(6):e007321.
45. Gussy M, Ashbolt R, Carpenter L, Virgo-Milton M, Calache H, Dashper S, et al. Natural history of dental caries in very young Australian children. International Journal of Paediatric Dentistry. 2016;26(3):173-183.

46. Lalloo R, Jamieson LM, Ha D, Luzzi L. Inequalities in Tooth Decay in Australian Children by Neighbourhood Characteristics and Indigenous Status. Journal of Health Care Poor Underserved. 2016;27(1 Suppl):161-177.
47. Jamieson LM, Smithers LG, Hedges J, Aldis J, Mills H, Kapellas K, et al. Follow-up of an Intervention to Reduce Dental Caries in Indigenous Australian Children: A Secondary Analysis of a Randomized Clinical Trial. Journal of the American Medical Association. 2019;2(3):e190648.
48. Kroon J, Lalloo R, Tadakamadla SK, Johnson NW. Dental caries experience in children of a remote Australian Indigenous community following passive and active preventive interventions. Community Dentistry & Oral Epidemiology. 2019;47(6):470-476.
49. Roberts-Thomson KF, Ha DH, Wooley S, Meihubers S, Do LG. Community trial of silver fluoride treatment for deciduous dentition caries in remote Indigenous communities. Australian Dental Journal. 2019;64(2):175-180.
50. Harris R, Nicoll AD, Adair PM, Pine CM. Risk factors for dental caries in young children: a systematic review of the literature. Community Dental Health. 2004;21(1 Suppl):71-85.
51. Leong PM, Gussy MG, Barrow SY, de Silva-Sanigorski A, Waters E. A systematic review of risk factors during first year of life for early childhood caries. International Journal of Paediatric Dentistry. 2013;23(4):235-250.
52. Burt BA. Definitions of risk. Journal of Dental Education. 2001;65(10):1007-8.
53. Fejerskov O, Kidd E. Biofilms in caries development. In: Dental Caries the Diseases and its Clinical Management, editor. Oxford: Blackwell Munksgaard Ltd; 2016.
54. Abanto J, Carvalho TS, Mendes FM, Wanderley MT, Bönecker M, Raggio DP. Impact of oral diseases and disorders on oral health-related quality of life of preschool children. Community Dentistry and Oral Epidemiology. 2011;39(2):105-114.
55. Filstrup SL, Briskie D, da Fonseca M, Lawrence L, Wandera A, Inglehart MR. Early childhood caries and quality of life: child and parent perspectives. Pediatric Dentistry. 2003;25(5):431-440.

56. Casamassimo PS, Thikkurissy S, Edelstein BL, Maiorini E. Beyond the dmft: the human and economic cost of early childhood caries. Journal of the American Dental Association. 2009;140(6):650-657.
57. Leake J, Jozzy S, Uswak G. Severe dental caries, impacts and determinants among children 2-6 years of age in Inuvik Region, Northwest Territories, Canada. Journal of the Canadian Dental Association. 2008;74(6):519.
58. Broadbent JM, Foster Page LA, Thomson WM, Poulton R. Permanent dentition caries through the first half of life. British Dental Journal. 2013;215(7):E12.
59. Zou J, Meng M, Law CS, Rao Y, Zhou X. Common dental diseases in children and malocclusion. International Journal of Oral Sciences. 2018;10(1):7.
60. Lindsten R. Secular changes in tooth size and dental arch dimensions in the mixed dentition. Swedish Dental Journal. 2003(157):1-89.
61. Lucas-Rincón SE, Robles-Bermeo NL, Lara-Carrillo E, Scougall-Vilchis RJ, Pontigo-Loyola AP, Rueda-Ibarra V, et al. Interproximal caries and premature tooth loss in primary dentition as risk factors for loss of space in the posterior sector: A cross-sectional study. Medicine. 2019;98(11):e14875.
62. Listl S, Grytten JI, Birch S. What is health economics? Community Dental Health. 2019;36(4):262-74.
63. AIHW. Disease expenditure in Australia Canberra: Australian Institute of Health and Welfare; 2019 [Available from: https://www.aihw.gov.au/reports/health-welfare-expenditure/disease-expenditure-australia/contents/australian-burden-of-disease-groups.
64. Sanders AE. Social determinants of oral health: conditions linked to socioeconomic inequalities in oral health in the Australian population. Canberra: Australian Institute of Health and Welfare; 2007. Contract No.: cat. no. POH 7.
65. World Health Organization. The determinants of health Geneva: World Health Organization; [Available from: http://www.who.int/hia/evidence/doh/en/#.
66. Madan C, Kruger E, Perera I, Tennant M. Trends in demand for general anaesthetic care for paediatric caries in Western Australia: geographic and socio-economic modelling of service utilisation. International Dental Journal. 2010;60(3):190-196.

67. Nadanovsky P, Sheiham A. Relative contribution of dental services to the changes in caries levels of 12-year-old children in 18 industrialized countries in the 1970s and early 1980s. Community Dentistry and Oral Epidemiology. 1995;23(6):331-339.
68. Wadhawan S, Kumar JV, Badner VM, Green EL. Early childhood caries-related visits to hospitals for ambulatory surgery in New York State. Journal of Public Health Dentistry. 2003;63(1):47-51.
69. Sbaraini A, Carter SM, Evans RW, Blinkhorn A. How do dentists and their teams incorporate evidence about preventive care? An empirical study. Community Dentistry and Oral Epidemiology. 2013;41(5):401-414.
70. Ricketts D, Lamont T, Innes NP, Kidd E, Clarkson JE. Operative caries management in adults and children. The Cochrane database of systematic reviews. 2013;3:Cd003808.
71. Chu CH, Lo EC, Lin HC. Effectiveness of silver diamine fluoride and sodium fluoride varnish in arresting dentin caries in Chinese pre-school children. Journal of Dental Research. 2002;81(11):767-770.
72. Crystal YO, Niederman R. Evidence-Based Dentistry Update on Silver Diamine Fluoride. Dental Clinics North America. 2019;63(1):45-68.
73. Oliveira BH, Rajendra A, Veitz-Keenan A, Niederman R. The Effect of Silver Diamine Fluoride in Preventing Caries in the Primary Dentition: A Systematic Review and Meta-Analysis. Caries Research. 2019;53(1):24-32.
74. Seifo N, Robertson M, MacLean J, Blain K, Grosse S, Milne R, et al. The use of silver diamine fluoride (SDF) in dental practice. British Dental Journal. 2020;228(2):75-81.
75. Dental Health Services Victoria. Establishing innovative models of care: A discussion paper. Melbourne: DHSV; 2012.
76. Woolcock K. Value based health care: setting the scene for Australia. Canberra: Deeble Institute for Health Policy Research; 2019. Contract No.: Deeble Institute Issues Brief no.31.
77. Frencken JE, Peters MC, Manton DJ, Leal SC, Gordan VV, Eden E. Minimal intervention dentistry for managing dental caries - a review: report of a FDI task group. International Dental Journal. 2012;62(5):223-243.

78. Calache H, Hopcraft MS, Martin JM. Minimum intervention dentistry--a new horizon in public oral health care. Australian Dental Journal. 2013;58 Suppl 1:17-25.
79. North Richmond Community Health. About us Melbourne: North Richmond Community Health; 2015 [Available from: http://www.nrch.com.au/about.
80. Harvard Womens Health Watch. Why it's hard to change unhealthy behavior--and why you should keep trying. Researchers say successful change comes only in stages. How long it takes is an individual matter. Harvard Women's Health Watch. 2007;14(5):4-5.
81. Australian Commission on Safety and Quality in Health Care. Patient and Consumer Centred Care Sydney: ACSQHC; 2016.
82. Hausen H. Caries prediction--state of the art. Community Dentistry & Oral Epidemiology. 1997;25(1):87-96.
83. Fontana M, Young DA, Wolff MS, Pitts NB, Longbottom C. Defining Dental Caries for 2010 and Beyond. Dental Clinics North America. 2010;54:423-440.
84. Domejean SL, S.; Rechmann, P.; White, J. M.; Featherstone, J. D. How do dental students determine patients' caries risk level using the Caries Management By Risk Assessment (CAMBRA) system? Journal of Dental Education. 2015;79(3):278-285.
85. Fisher-Owens SA, Gansky SA, Platt LJ, Weintraub JA, Soobader MJ, Bramlett MD, et al. Influences on children's oral health: a conceptual model. Pediatrics. 2007;120(3):e510-20.
86. Kelly MP, Stewart E, Morgan A, Killoran A, Fischer A, Threlfall A, et al. A conceptual framework for public health: NICE's emerging approach. Public Health. 2009;123(1):e14-20.
87. Solar O, Irwin A. A Conceptual Framework for Action on the Social Determinants of Health. Geneva; 2010.
88. Porter ME, Lee TH. The strategy that will fix health care. Boston: Harvard Business School; 2013.
89. Porter ME, Teisberg E. Redefining health care: Creating value-based competition on results. Boston; 2006.

90. Teisberg EO, Wallace S. Creating a high-value delivery system for health care. Seminars Thoracic Cardiovascular Surgery. 2009;21(1):35-42.
91. Askitopoulou H, Vgontzas AN. The relevance of the Hippocratic Oath to the ethical and moral values of contemporary medicine. Part I: The Hippocratic Oath from antiquity to modern times. European Spine Journal. 2018;27(7):1481-1490.
92. Wheeler SB, Spencer J, Rotter J. Toward Value in Health Care: Perspectives, Priorities, and Policy. North Carolina Medical Journal. 2018;79(1):62-65.
93. DePinho R, Goozner M. Value-based care is about outcomes, not just cutting cost. Modern Healthcare. 2015;45(12):30-31.
94. Glaser J. IT: key enabler of value-based care: data drives success. Hospital Health Network. 2013;87(5):64.
95. Scott I. Ten clinician-driven strategies for maximising value of Australian health care. Australian Health Review. 2014;38(2):125-133.
96. Spade JS, Strickland SC. Rural hospitals face many challenges in transitioning to value-based care. North Carolina Medical Journal. 2015;76(1):38-39.
97. Lewis S. Value-based healthcare - meeting the evolving needs of our population. Australian Health Review. 2019;43(5):485.
98. Deerberg-Wittram J, editor. What matters most: Patient Outcomes and the Transformation of Health Care. Cambridge: ICHOM International Consortium for Health Outcomes Measurement.
99. Nunlist MM, Uiterwyk S, Nicoletti B. Gambling on the transition from fee-for-service to value-based care. Family Practice Management. 2014;21(5):6-7.
100. Novikov D, Cizmic Z, Feng JE, Iorio R, Meftah M. The Historical Development of Value-Based Care: How We Got Here. Journal of Bone and Joint Surgery. 2018;100(22):e144.
101. National Center for Health Statistics. Gross domestic product, national health expenditures, per capita amounts, percent distribution, and average annual percent change: United States, selected years 1960–2017 Atlanta: US Centers for Disease Control and Prevention; 2018 [Available from: https://www.cdc.gov/nchs/data/hus/2018/042.pdf.

102. Australian Institute of Health and Welfare. Health expenditure Australia 2017–18. Health and welfare expenditure series no.65. Cat. no. HWE 77. Canberra: AIHW; 2019. Contract No.: March.
103. CMS GOV. Bundled Payments for Care Improvement (BPCI) Initiative: General Information Baltimore: The Centers for Medicare & Medicaid Services; 2020 [Available from: https://innovation.cms.gov/innovation-models/bundled-payments.
104. Chen LM, Ryan AM, Shih T, Thumma JR, Dimick JB. Medicare's Acute Care Episode Demonstration: Effects of Bundled Payments on Costs and Quality of Surgical Care. Health Service Research. 2018;53(2):632-648.
105. Gray M, Wells G, Lagerberg T. Optimising allocative value for populations. Journal of the Royal Society of Medicine. 2017;110(4):138-143.
106. Elangovan S, Allareddy V. Value-Based Payment Approaches. Journal of the American Dental Association. 2019;150(6):485.
107. Listl S. Value-Based Oral Health Care: Moving Forward With Dental Patient-Reported Outcomes. Journal of Evidence Based Dental Practice. 2019;19(3):255-259.
108. Panesar SS. The role of oral health in an evolving health care delivery system: An interview with Bruce Donoff, D.M.D., M.D. Healthc (Amst). 2020;8(1):100399.
109. Patel NA, Afshar S. Does Fee-for-Service Medicare Reimbursement Incentivize Volume Over Value Among Oral and Maxillofacial Surgeons? Journal of Oral Maxillofacial Surgery. 2020.
110. Riley W, Doherty M, Love K. A framework for oral health care value-based payment approaches. Journal of the American Dental Association. 2019;150(3):178-185.
111. Marmot M. Social determinants of health inequalities. Lancet. 2005;365(9464):1099-104.
112. Institute of Medicine. The Interplay of Biological, Behavioral and Societal Influences. Washington, D.C.: National Academy Press; 2001.
113. Marmot M, Bell R. Social determinants and dental health. Advances in Dental Research. 2011;23(2):201-206.

114. Johnson E. What We Talk About When We Talk About Value-Based Care. Managed Care. 2019;28(3):37.
115. Twetman S, Fontana M. Patient caries risk assessment. Monograms of Oral Science 2009;21:91-101.
116. Zhang Y, Li KY, Lo ECM, Wong MCM. Structural equation model for parental influence on children's oral health practice and status. BMC Oral Health. 2020;20(1):56.
117. Riggs E, Kilpatrick N, Slack-Smith L, Chadwick B, Yelland J, Muthu MS, et al. Interventions with pregnant women, new mothers and other primary caregivers for preventing early childhood caries. The Cochrane database of systematic reviews. 2019;2019(11).
118. Pierce A, Singh S, Lee J, Grant C, Cruz de Jesus V, Schroth RJ. The Burden of Early Childhood Caries in Canadian Children and Associated Risk Factors. Frontiers in Public Health. 2019;7:328.
119. Martin MA, Zimmerman LJ, Rosales GF, Lee HH, Songthangtham N, Pugach O, et al. Design and sample characteristics of COordinated Oral health Promotion (CO-OP) Chicago: A cluster-randomized controlled trial. Contemporary Clinical Trials. 2020;92:105919.
120. Gussy M, Mnatzaganian G, Dashper S, Carpenter L, Calache H, Mitchell H, et al. Identifying predictors of early childhood caries among Australian children using sequential modelling: Findings from the VicGen birth cohort study. Journal of Dentistry. 2020;93:103276.
121. Higgins JPT, Green S (editors). Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0 [updated March 2011]: The Cochrane Collaboration,; 2011 [Available from: www.cochrane-handbook.org. .
122. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. International Journal of Surgery. 2010;8(5):336-341.
123. Protocol for systematic reviews of measurement properties [press release]. Amsterdam: COSMIN2011.
124. Mokkink L, Terwee C, Patrick D, Alonso J, Stratford P, Knol D, et al. COSMIN checklist manual. Amsterdam; 2012.

125. Terwee CB, Mokkink LB, Knol DL, Ostelo RW, Bouter LM, de Vet HC. Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. Quality of Life Research2012;21(4):651-657.
126. Stamm JW, Disney JA, Graves RC, Bohannan HM, Abernathy JR. The University of North Carolina Caries Risk Assessment Study. I: Rationale and content. Journal Of Public Health Dentistry. 1988;48(4):225-232.
127. Ryan R. Cochrane Consumers and Communication Review Group. 'Cochrane Consumers and Communication Review Group: data synthesis and analysis' 2013 [Available from: http://cccrg.cochrane.org.
128. de Silva-Sanigorski AM, Calache H, Gussy M, Dashper S, Gibson J, Waters E. The VicGeneration study--a birth cohort to examine the environmental, behavioural and biological predictors of early childhood caries: background, aims and methods. BMC Public Health. 2010;10:97.
129. Centrelink Australia. Health Care Card: Australian Government Services Australia; 2020 [Available from: https://www.servicesaustralia.gov.au/individuals/services/centrelink/health-care-card.
130. Hallgren KA. Computing Inter-Rater Reliability for Observational Data: An Overview and Tutorial. Tutorials in Quantitative Methods for Psychology. 2012;8(1):23-34.
131. Ismail AI, Sohn W, Tellez M, Amaya A, Sen A, Hasson H, et al. The International Caries Detection and Assessment System (ICDAS): an integrated system for measuring dental caries. Community Dentistry and Oral Epidemiology. 2007;35:170-178.
132. Landis JR, Koch GG. The measurement of observer agreement for categorical data. Biometrics. 1977;33:159-174.
133. Litaker MS, Gordan VV, Rindal DB, Fellows JL, Gilbert GH. Cluster Effects in a National Dental PBRN restorative study. Journal of Dental Research. 2013;92(9):782-787.

134. Masood M, Masood Y, Newton JT. The Clustering Effects of Surfaces within the Tooth and Teeth within Individuals. Journal of Dental Research. 2015;94(2):281-288.
135. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, et al. The REDCap consortium: Building an international community of software platform partners. Journal of Biomedical Informatics. 2019;95:103208.
136. Christian B, Armstrong R, Calache H, Carpenter L, Gibbs L, Gussy M. A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. International Journal of Paediatric Dentistry. 2019;29(2):106-116.
137. Featherstone JD. The caries balance: the basis for caries management by risk assessment. Oral Health Prevention and Dentistry. 2004;2 Suppl 1:259-264.
138. Tellez M. Caries risk profiles using cariogram are predictive of caries incidence in permanent dentition. Journal of Evidence based Dental Practice. 2013;13(2):58-59.
139. Whiting PF, Rutjes AW, Westwood ME, Mallett S, Deeks JJ, Reitsma JB, et al. QUADAS-2: a revised tool for the quality assessment of diagnostic accuracy studies. Annals of Internal Medicine. 2011;155(8):529-536.
140. Guyatt GH, Oxman AD, Vist GE, Kunz R, Falck-Ytter Y, Alonso-Coello P, et al. GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. British Medical Journal. 2008;336(7650):924-936.
141. Terwee CB, Schellingerhout JM, Verhagen AP, Koes BW, de Vet HC. Methodological quality of studies on the measurement properties of neck pain and disability questionnaires: a systematic review. Journal of Manipulative and Physiological Therapeutics. 2011;34(4):261-272.
142. Beck JD, Weintraub JA, Disney JA, Graves RC, Stamm JW, Kaste LM, et al. University of North Carolina Caries Risk Assessment Study: comparisons of high risk prediction, any risk prediction, and any risk etiologic models. Community Dentistry & Oral Epidemiology. 1992;20(6):313-321.
143. Disney JA, Graves RC, Stamm JW, Bohannan HM, Abernathy JR, Zack DD. The University of North Carolina Caries Risk Assessment study: further developments

in caries risk prediction. Community Dentistry and Oral Epidemiology. 1992;20(2):64-75.
144. Wandera A, Bhakta S, Barker T. Caries prediction and indicators using a pediatric risk assessment teaching tool. Journal of Dentistry Child. 2000;67(6):408-12, 375.
145. Holgerson PL, Twetman S, Stecksen-Blicks C. Validation of an age-modified caries risk assessment program (Cariogram) in preschool children. Acta Odontologica Scandinavica. 2009;67(2):106-112.
146. Gao X, Di Wu I, Lo EC, Chu CH, Hsu CY, Wong MC. Validity of caries risk assessment programmes in preschool children. Journal of Dentistry. 2013;41(9):787-795.
147. Gao XL, Hsu CY, Xu Y, Hwarng HB, Loh T, Koh D. Building caries risk assessment models for children. Journal of Dental Research. 2010;89(6):637-643.
148. MacRitchie HMBL, Christopher; Robertson, Margaret; Nugent, Zoann; Chan, Karen; Radford, John R.; Pitts, Nigel B. Development of the Dundee Caries Risk Assessment Model (DCRAM) - risk model development using a novel application of CHAID analysis. Community Dentistry & Oral Epidemiology. 2012;40(1):37-45.
149. Yoon RK, Smaldone AM, Edelstein BL. Early childhood caries screening tools: a comparison of four approaches. Journal of the American Dental Association 2012;143(7):756-763.
150. Chaffee BW, Featherstone JD, Gansky SA, Cheng J, Zhan L. Caries Risk Assessment Item Importance: Risk Designation and Caries Status in Children under Age 6. JDR Clinical and Translational Research. 2016;1(2):131-142.
151. Custodio-Lumsden CL, Wolf RL, Contento IR, Basch CE, Zybert PA, Koch PA, et al. Validation of an early childhood caries risk assessment tool in a low-income Hispanic population. Journal of Public Health Dentistry 2016;76(2):136-142.
152. Levine J, Wolf RL, Chinn C, Edelstein BL. MySmileBuddy: an iPad-based interactive program to assess dietary risk for early childhood caries. Journal of the Academy of Nutrition and Dietetics. 2012;112(10):1539-1542.
153. Domejean S, Banerjee A, Featherstone JDB. Caries risk/susceptibility assessment: its value in minimum intervention oral healthcare. British Dental Journal. 2017;223(3):191-197.

154. Disney JAG, R. C.; Stamm, J. W.; Bohannan, H. M.; Abernathy, J. R. The University of North Carolina Caries Risk Assessment Study. II. Baseline caries prevalence. Journal Of Public Health Dentistry. 1990;50(3):178-185.
155. Broder HL, Wilson-Genderson M. Reliability and convergent and discriminant validity of the Child Oral Health Impact Profile (COHIP Child's version). Community Dentistry and Oral Epidemiology 2007;35 Suppl 1:20-31.
156. Petersson GH. Assessing caries risk--using the Cariogram model. Swedish Dental Journal. 2003(158):1-65.
157. Petersson GHB, D. Caries risk assessment: a comparison between the computer program 'Cariogram', dental hygienists and dentists. Swedish Dental Journal. 2000;24(4):129-137.
158. Petersson GHE, E.; Isberg, P. E.; Twetman, S. Caries risk assessment in young adults using Public Dental Service guidelines and the Cariogram--a comparative study. Acta Odontologica Scandinavica. 2013;71(3-4):534-540.
159. The COSMIN initiative. About the COnsensus-based Standards for the selection of health Measurement INstruments initiative Amsterdam: Amsterdam Public Health; 2020 [Available from: https://www.cosmin.nl/about/.
160. Walsh CJ, Gansky SA, Vaderhobli RM, Vijan PF, M., Rechmann P, White JM. Usability testing of clinical decision support caries risk assessment tools. International Association for Dental Research; Boston2015.
161. Nelson S, Eggertsson H, Powell B, Mandelaris J, Ntragtakis M, Richardson T, et al. Dental examiners consistency in applying the ICDAS criteria for a caries prevention community trial. Community Dental Health. 2011;28:238-242.
162. Boye U, Walsh T, Pretty IA, Tickle M. Comparison of photographic and visual assessment of occlusal caries with histology as the reference standard. BMC Oral Health. 2012;12:10.
163. Elfrink ME, Veerkamp JS, Aartman IH, Moll HA, Ten Cate JM. Validity of scoring caries and primary molar hypomineralization (DMH) on intraoral photographs. European archives of paediatric dentistry : official journal of the European Academy of Paediatric Dentistry. 2009;10 Suppl 1:5-10.

164. Boye U, Foster GR, Pretty IA, Tickle M. The views of examiners on the use of intra-oral photographs to detect dental caries in epidemiological studies. Community Dental Health. 2013;30(1):34-38.
165. Boye U, Pretty IA, Tickle M, Walsh T. Comparison of caries detection methods using varying numbers of intra-oral digital photographs with visual examination for epidemiology in children. BMC Oral Health. 2013;13:6.
166. Boye U, Willasey A, Walsh T, Tickle M, Pretty IA. Comparison of an intra-oral photographic caries assessment with an established visual caries assessment method for use in dental epidemiological studies of children. Community Dentistry and Oral Epidemiology. 2013;41(6):526-533.
167. Bader JD, Shugars DA, Bonito AJ. A systematic review of the performance of methods for identifying carious lesions. Journal of Public Health Dentistry. 2002;62(4):201-213.
168. Gomez J, Tellez M, Pretty IA, Ellwood RP, Ismail AI. Non-cavitated carious lesions detection methods: a systematic review. Community Dentistry and Oral Epidemiology 2013;41(1):54-66.
169. Topping GVA, Hally J, Bonner B, Pitts NB. International Caries Detection and Assessment System (ICDAS) e-Learning Package. Interactive CD ROM and Web-based software. London: Smile-on; 2008.
170. Altarakemah Y, Al-Sane M, Lim S, Kingman A, Ismail AI. A new approach to reliability assessment of dental caries examinations. Community Dentistry and Oral Epidemiology. 2013;41(4):309-316.
171. Braga MM, Oliveira LB, Bonini GA, Bonecker M, Mendes FM. Feasibility of the International Caries Detection and Assessment System (ICDAS-II) in epidemiological surveys and comparability with standard World Health Organization criteria. Caries Research. 2009;43(4):245-249.
172. Pine CM, Pitts NB, Nugent ZJ. British Association for the Study of Community Dentistry (BASCD) guidance on the statistical aspects of training and calibration of examiners for surveys of child dental health. A BASCD coordinated dental epidemiology programme quality standard. Community Dental Health. 1997;14(Suppl 1.):18-29.

173. Fleiss JL. The equivalence of weighted kappa and the intraclass correlation coffecient as measures of reliability. Educational and Psychological Measurement. 1973;33:613-619.
174. Maclure M, Willett WC. Misinterpretation and misuse of the kappa statistic. American Journal of Epidemiology. 1987;126(2):161-169.
175. Piovesan C, Ardenghi TM, Guedes RS, Ekstrand KR, Braga MM, Mendes FM. Activity assessment has little impact on caries parameters reduction in epidemiological surveys with preschool children. Community Dentistry and Oral Epidemiology. 2013(3):204.
176. Banting DW, Amaechi BT, Bader JD, Blanchard P, Gilbert GH, Gullion CM, et al. Examiner training and reliability in two randomized clinical trials of adult dental caries. Journal of Public Health Dentistry. 2011;71(4):335-344.
177. Chrisopoulos S, Harford J, Ellershaw A. Oral health and dental care in Australia: key facts and figures 2015. Cat. no. DEN 229. Canberra: AIHW; 2016. Contract No.: Cat. no. DEN 229.
178. Christian B, Amezdroz E, Calache H, Gussy M, Sore R, Waters E. Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical. Community Dental Health. 2017;34(4):248-253.
179. Beck JD. Risk revisited. Community Dentistry and Oral Epidemiology 1998;26(4):220-5.
180. Featherstone JD. Caries prevention and reversal based on the caries balance. Pediatric Dentistry. 2006;28(2):128-132.
181. Gibbs L, de Silva AM, Christian B, Gold L, Gussy M, Moore L, et al. Child oral health in migrant families: A cross-sectional study of caries in 1-4 year old children from migrant backgrounds residing in Melbourne, Australia. Community Dental Health. 2016;33(2):100-116.
182. Chaffee BW, Feldens CA, Rodrigues PH, Vitolo MR. Feeding practices in infancy associated with caries incidence in early childhood. Community Dentistry and Oral Epidemiology. 2015;43(4):338-348.
183. Feldens CA, Rodrigues PH, de Anastacio G, Vitolo MR, Chaffee BW. Feeding frequency in infancy and dental caries in childhood: a prospective cohort study. International Dental Journal. 2018;68(2):113-121.

184. Chaffee BW, Featherstone JDB, Zhan L. Pediatric Caries Risk Assessment as a Predictor of Caries Outcomes. Pediatric Dentistry. 2017;39(3):219-232.
185. Jørgensen MR, Twetman S. A systematic review of risk assessment tools for early childhood caries: is there evidence? European Archives of Paediatric Dentistry. 2020;21(2):179-184.
186. International Association of Paediatric Dentistry. Early Childhood Caries: IAPD Bangkok Declaration. International Journal of Paediatric Dentistry. 2019;29(3):384-386.
187. Cagetti MG, Bontà G, Cocco F, Lingstrom P, Strohmenger L, Campus G. Are standardized caries risk assessment models effective in assessing actual caries status and future caries increment? A systematic review. BMC Oral Health. 2018;18(1):123.
188. Lim S, Alqahtani S, Tellez M, Ismail AI. An innovative individual-level caries severity and progression indicator among children. Journal of Public Health Dentistry. 2020;80(2):168-174.
189. Selwitz RH, Ismail AI, Pitts NB. Dental caries. Lancet. 2007;369(9555):51-59.
190. Mokkink LB, Terwee CB, Gibbons E, Stratford PW, Alonso J, Patrick DL, et al. Inter-rater agreement and reliability of the COSMIN (COnsensus-based Standards for the selection of health status Measurement Instruments) checklist. BMC Medical Research Methodology. 2010;10:82.

PhD Appendices

Appendix 1. Background published paper 1

Citation: *Christian B, Hall M, Martin R. A paradigm shift in models of oral health care: An example and a call to action. Family Medicine and Community Health. 2015;3(4):32-7.*

Family Medicine and Community Health
REVIEW

REVIEW

A paradigm shift in models of oral health care: An example and a call to action

Bradley Christian, Martin Hall, Rachel Martin

Abstract

The consequences of oral disease are wide-ranging and can have a major impact on an individual's and that person's family's quality of life. A range of factors interact to determine a person's oral and general health. Such factors can be biological, social, economic, political, cultural, or environmental, in addition to knowledge, attitudes and behaviors. Traditional models of oral health care, however, have generally ignored these factors and instead have focused on the treatment and management of existing pathology (tertiary prevention/downstream approach). This has had no effect on the rate of hospitalization or the inequitable distribution of dental diseases. To reduce the prevalence and severity of oral diseases at the individual and population levels, holistic evidence-informed prevention-based health-promoting models of care that focus on upstream determinants of health are required. The Oral Health Program at North Richmond Community Health in the state of Victoria, Australia, has developed an innovative model of oral health care based on the following principles: health promotion, disease prevention, risk-based access to care, client- and family-centered care, team-based provision of care, multidisciplinary care, and innovation. Evaluation of this approach is currently being conducted to study the sustainability of such a model under the current public dental service funding model.

Keywords: Delivery of health care; health promotion; oral health; dental health services; dentistry

Oral Health, North Richmond Community Health Limited, Richmond, VIC 3121, Australia

CORRESPONDING AUTHOR:
Dr. Bradley Christian, BDS, MDSc (Hons)
Research Fellow, Oral Health, North Richmond Community Health Limited, 23 Lennox Street, Richmond, VIC 3121, Australia
Tel.: +61-9418-9825
Fax: +61-9428-2269
E-mail: bradleyc@nrch.com.au

Received 22 May 2015;
Accepted 31 August 2015

Introduction

Oral health is essential to general health and quality of life. It is a state of being free from mouth and facial pain, oral and throat cancer, oral infection and sores, periodontal (gum) disease, tooth decay, tooth loss, and other diseases and disorders that limit an individual's capacity in biting, chewing, smiling, speaking, and psychosocial well-being [1]. The consequences of oral disease are wide-ranging and can have a major impact on an individual's and that person's family's quality of life. Pain, infection, and tooth loss are the most common consequences of oral disease, which can cause difficulties with chewing and swallowing, as well as speech, sleep, and productivity. There can also be significant impacts on self-esteem, psychological and social well-being, employment, interpersonal relations, and quality of life. Poor oral health has also been linked to a range of conditions including cardiovascular disease, diabetes, stroke, kidney disease, and respiratory infections [2, 3].

Recent Australian data show that nearly half of young children (6-year-old) have caries in their deciduous (baby) teeth, with approximately 62% of this decay being untreated. For older children (12-year-old), 39% have caries, of which approximately 52% is untreated [4]. Recent hospital admission data in Victoria highlight that for 0–14-year-old children, treatment for dental extractions and restorations is second to treatment of asthma as the most common cause of hospital admission for children [5]. Vulnerable populations such as people from low socioeconomic and disadvantaged backgrounds, Aboriginal and Torres Strait Islanders, refugee and asylum seekers, pregnant women, the elderly, and people with complex special needs have higher levels of oral disease and poorer overall health [3, 6, 7]. The Healthy Mouths, Healthy Lives National Oral Health Plan identified the importance of good oral health in all population groups within Australia.

Models of care

In this article 'model of care' is defined as a multifaceted concept, which broadly defines the way health services are delivered. It outlines best-practice care and services for a person, population group, or patient cohort as they progress through the stages of a condition, injury, or event [8]. The particular model of care at a health care service can significantly impact on both the client accessing the services and the service provider. A model of care's outcomes can be related to domains such as client health outcomes, service accessibility, service quality, service economics, organization policy, workforce structure, and sustainability. Given this range of outcomes, a best-practice model of care will need a multipronged approach to positively address the various domains. Models of care which encompass prevention and seek to address the broader determinants of oral health outcomes will have the most potential to reduce the inequitable social gradient in oral health [9]. Such models need to be continually evaluated, evolving, adapting to circumstances and new evidence, and be able to operate in the various domains, stated previously. Evaluations of models of care should be ongoing as there is often very limited evidence on which to make decisions, and as such policy and economics heavily influence the direction of new approaches.

Traditional models of oral health care

A range of factors interact to determine a person's oral and general health. Such factors can be biological, social, economic, political, cultural, or environmental, in addition to knowledge, attitudes, and behaviors [10, 11]. Traditional models of oral health care, however, have generally ignored the above-stated determinants and instead have focused on the treatment and management of existing diseases (tertiary prevention/downstream approach). This has had no effect on the rate of hospitalization or the inequitable distribution of dental diseases [12–15]. An example in the oral health sphere is the conventional practice for caries management, where a reliance on surgical intervention (drilling and filling) is informed more by opinion derived from subjective experience among practicing clinicians than by current evidence on the caries process [16]. An analysis of epidemiologic studies on (1) diagnosis, (2) lesion incidence and progression rates of dental caries, (3) noninvasive primary and secondary preventive strategies, and (4) social determinants of health, and laboratory studies into the (1) biology, (2) chemistry, and (3) physics of lesion progression and regression indicates that timely intervention by nonsurgical treatment modes could avoid the need for most restorative dentistry (fillings) [17, 18]. However, Australian data from 2003–2004 show that restorative treatment continues to be common practice for managing caries, including for noncavitated lesions [19]. Nadanovsky and Sheiham [15] showed that dental services explained only about 3% of the differences in the change in 12-year-old caries levels across several countries. Socioeconomic factors had the largest impact independent of the use of fluoridated toothpaste. Thus, with the traditional model of oral health care, oral diseases and inequalities in health will continue to be prevalent and may even increase.

Paradigm shift in models of oral health care

In contemporary times, changing service needs, workforce deficiencies, and an outcome-based approach have created an urgent need for new approaches to both the delivery and the types of oral health care provided in the community. The need for flexible, evidence-based and client-centered models of care has been identified by some health services as being important to improving client oral health, and the efficiency

and effectiveness of service delivery. In response to a broader understanding of the social determinants of health and well-being, and the critical importance of prevention and oral health promotion to health outcomes, innovative evidence-informed models of care are being developed and tested. These new models will require efficient, effective, and sustainable integration of population-based health promotion and disease prevention strategies along with, but with a reduced emphasis on, treatment-oriented clinical interventions. They will also need to include strategies for early detection and disease identification, and be based on the principles of minimal intervention dentistry (MID). The concept of MID came about in the late 1980s, for several reasons: (1) the dramatic decline in caries rates; (2) the evidence to support fluoridation and widespread use of fluoride toothpaste and other fluoride vehicles; (3) failure of restorations – repeated restoration cycles; and (d) new evidence on the caries process and associated factors – progression, regression, bacterial biofilm (plaque), and saliva. The guiding principles of MID are (1) early caries detection and risk assessment, (2) remineralization of demineralized enamel and dentine; (3) optimal caries preventive measures; (4) minimally invasive operative interventions; and (5) repair rather than replacement of restorations [20]. In its earlier form, MID was focused very much on the clinical aspects of disease management; however, in contemporary times, in recognition of the social, cultural, economic, and environmental influences on oral health, the concept of MID has become enmeshed in health promotion principles [21] – thus the new terminology 'health-promoting models of oral health care.' Risk assessment, a component of MID, is important for several reasons: it prioritizes access to care on the basis of risk; it helps identify particular risk factors for the disease and thus supports targeted, effective, and efficient disease management (including patient recall times); and it helps patients understand their risk of developing disease and associated factors, which empowers them to make informed decisions and take actions to reduce risk. Several caries risk assessment tools are currently available, and the evidence to support the reliability, validity, and implementation of these tools in everyday practice is being generated through research and evaluation studies [22].

The following section provides information on an evidence-informed innovative prevention-based health-promoting model of oral health care, which was developed and is being implemented at North Richmond Community Health (NRCH) in Victoria, Australia.

An example: the North Richmond prevention-based model of oral health care

NRCH has been providing quality community health services since 1974. The organization's vision is to be an innovative organization, responsive to and supportive of community needs and aspirations: open to change and challenges. NRCH currently operates more than 15 health service programs, administers an annual budget of $8 million, and employs a committed staff group of more than 100 people [23].

The oral health program (NRCH-OH) employs 40 staff and operates with a clinical team consisting of oral health educators, dental and oral health therapists, hygienists, prosthetists, dentists, and a specialist dentist. NRCH-OH operates over three sites and two mobile outreach programs. The infrastructure at the Richmond site includes a modern seven-chair clinic and two portable chairs used for outreach. The NRCH-OH program aims to improve oral health of clients by moving away from the traditional treatment/disease-focused care, toward a more holistic health promotion–based model of care. This is in recognition of the fact that people's health and health-related behaviors are intricately associated with their social, cultural, economic, demographic, and political environments. The NRCH-OH model of oral health care is strongly aligned with the health promotion priority action areas as determined by the Ottawa Charter [24]. The guiding principles of the NRCH-OH model of oral health care, to achieve positive oral health outcomes for clients, staff, and the organization, are as follows:

A. ***Health promotion based on the themes of the Ottawa Charter.*** The Ottawa Charter health promotion themes ensure a holistic approach to improving the health of individuals, communities, and populations. The themes are as follows:
   - Reorienting health services. The role of the health sector must move increasingly in a health promotion direction, beyond its responsibility for providing clinical and curative services. Health services need to embrace an expanded mandate which is sensitive

to and respects cultural needs. This mandate should support the needs of individuals and communities for a healthier life, and open channels between the health sector and broader social, political, economic, and physical environmental components.

- Developing personal skills. Enabling people to learn throughout life, to prepare themselves for all of its stages, and to cope with chronic illness and injuries is essential. This has to be facilitated in school, home, work, and community settings.
- Strengthening community action. At the heart of this process is the empowerment of communities – their ownership and control of their own endeavors and destinies.
- Create supportive environments. Our societies are complex and interrelated. Health cannot be separated from other goals. The inextricable links between people and their environment constitutes the basis for a socioecological approach to health.
- Build healthy public policy. It is coordinated action that leads to health, income, and social policies that foster greater equity.

B. Prevention. The NRCH-OH model of oral health care incorporates two concepts of disease prevention: (1) the individual-focused traditional concept of primary, secondary, and tertiary prevention of disease; and (2) the concept of prevention which aims to understand the broader environment people live in and how this influences oral health (population health approach).

C. Risk-based access (both disease and social). Classifying and prioritizing clients on the basis of their risk of oral diseases. Risk profiles for individual clients and their families are assigned on the basis of their current disease profile and indicators of their social, economic, cultural, and physical status.

D. Client- and family-centered approach to care. This ensures that clients feel safe, respected, and empowered as partners in decision making regarding their oral health management. Clients are empowered to make informed decisions regarding their health and treatment options in consultation with the oral health provider. The family-centered approach to health care recognizes that health is intricately related to various aspects of an individual's daily life, and that family/community-level intervention is required to create supportive environments to facilitate effective and sustainable changes in healthy behavior and health outcomes.

E. Partnership- and dental team–based care. Efficient use of the whole team, avoiding duplication of roles within the dental team (dentists, therapists, hygienists, and assistants having clearly defined complementary roles) and a greater role of dental assistants in both prevention and the client experience.

F. Multidisciplinary. Recognizing that oral health is but one aspect of general health and that it shares common risk factors for diseases with general health [25], so working in partnership with other health services within and external to NRCH such as nutrition, diabetes, and smoking cessation services.

G. Innovative use of staff and resources. Constant evaluation, reassessment, and preparedness to change and evolve. Identification of individual talents, support for professional development, and creating career pathways for enhancement of skills and job satisfaction.

Prevention requires a long-term view and is fundamentally different from the treatment-based approach that most clients and staff have been accustomed to. There will always be factors beyond the clients' control; however, facilitating increased dental awareness and oral hygiene practice may help improve not only the clients' oral health but also their general health. Empowering and supporting clients to make simple dietary changes or to use fluoride toothpaste twice a day to brush their teeth are effective actions in producing improvements in oral and general health. However, to move a treatment-based program funded to fix teeth (traditional model of care) to a model based on health promotion and disease prevention is a major challenge. Robust and ongoing evaluation of this approach to study its ability to exist and be sustainable under the current funding formulation is required.

Conclusion

Oral health and healthy behavior is determined by the complex action and interaction of social, economic, demographic, political, and environmental factors at various levels (individual,

family, community, etc.). Traditionally, models of oral health care have focused on downstream drivers of health, without much success, and as a result, oral diseases continue to be highly prevalent in the population. In addition, most traditional oral health care practices are determined by expert opinion (lowest form of evidence) rather than through scientific enquiry. To reduce the prevalence and severity of oral diseases at the individual and population levels, holistic evidence-informed prevention-based models of care that focus on upstream determinants of health are required. The challenges associated with changing to an evidence-informed model of oral health care will be significant, particularly since most dental practitioners and clients view mechanical (drilling and filling), technology-based, disease-focused and expert opinion–driven care as being best-practice dentistry. However, with innovative and passionate clinicians, supportive professional and academic leadership, and a commitment to improve oral health, steady progress toward this change can be achieved.

Acknowledgment

The authors acknowledge North Richmond Community Health for administrative and resource support.

Conflict of interest

The authors declare no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

References

1. World Health Organization. Oral Health. Geneva: World Health Organization [cited 2015]. Available from: www.who.int/topics/oral_health/en/.
2. Petersen PE. The World Oral Health Report 2003: continuous improvement of oral health in the 21st century – the approach of the WHO Global Oral Health Programme. Community Dent Oral Epidemiol 2003;31 Suppl 1:3–23.
3. US Department of Health and Human Services. Oral health in America: a report of the Surgeon General Rockville: US Department of Health and Human Services, National Institute of Dental and Craniofacial Research, Health NIo; 2000.
4. Mejia GC, Amarasena N, Ha DH, Roberts-Thomson KF, Ellershaw AC. Child Dental Health Survey Australia 2007: 30-year trends in child oral health. Canberra: AIHW; 2012.
5. Department of Health and Human Service/State Government of Victoria. Victorian Health Information Surveillance System (VHISS) Melbourne: Department of Health and Human Services; [cited 2015 May 16]. Available from: www.health.vic.gov.au/healthstatus/interactive/vhiss.htm.
6. Christian B, Blinkhorn AS. A review of dental caries in Australian Aboriginal children: the health inequalities perspective. Rural Remote Health 2012;12(4):2032.
7. Davidson N, Skull S, Calache H, Murray SS, Chalmers J. Holes a plenty: oral health status a major issue for newly arrived refugees in Australia. Aust Dent J 2006;51(4):306–11.
8. Queensland Health. Changing models of care framework. Brisbane: Queensland Health; 2000.
9. Marmot M. Social determinants of health inequalities. Lancet 2005;365(9464):1099–104.
10. Sanders AE. Social determinants of oral health: conditions linked to socioeconomic inequalities in oral health in the Australian population. Canberra: Australian Institute of Health and Welfare, 2007 Contract No.: cat. no. POH 7.
11. World Health Organization. The determinants of health. Geneva: World Health Organization; [cited 2015]. Available from: www.who.int/hia/evidence/doh/en/#.
12. Kruger E, Tennant M. Ten years of hospitalisation for oral health-related conditions in Western Australia: an unjust dichotomy. Aust J Prim Health 2015. [Epub ahead of print]
13. Madan C, Kruger E, Perera I, Tennant M. Trends in demand for general anaesthetic care for paediatric caries in Western Australia: geographic and socio-economic modelling of service utilisation. Int Dent J 2010;60(3):190–6.
14. Wadhawan S, Kumar JV, Badner VM, Green EL. Early childhood caries-related visits to hospitals for ambulatory surgery in New York State. J Public Health Dent 2003;63(1):47–51.
15. Nadanovsky P, Sheiham A. Relative contribution of dental services to the changes in caries levels of 12-year-old children in 18 industrialized countries in the 1970s and early 1980s. Community Dent Oral Epidemiol 1995;23(6):331–9.
16. Sbaraini A, Carter SM, Evans RW, Blinkhorn A. How do dentists and their teams incorporate evidence about preventive care? An empirical study. Community Dent Oral Epidemiol. 2013;41(5):401–14.
17. Fejerskov O, Kidd E, editors. Dental caries: the disease and its clinical management. Oxford: Blackwell Munksgaard Ltd; 2008.

18. Ricketts D, Lamont T, Innes NP, Kidd E, Clarkson JE. Operative caries management in adults and children. The Cochrane Database of Systematic Reviews 2013;3:CD003808.
19. Brennan DS, Spencer AJ. Service patterns associated with coronal caries in private general dental practice. J Dent 2007;35(7):570–7.
20. Frencken JE, Peters MC, Manton DJ, Leal SC, Gordan VV, Eden E. Minimal intervention dentistry for managing dental caries – a review: report of a FDI task group. Int Dent J 2012;62(5):223–43.
21. Calache H, Hopcraft MS, Martin JM. Minimum intervention dentistry – a new horizon in public oral health care. Aust Dent J 2013;58 Suppl 1:17–25.
22. Tellez M, Gomez J, Pretty I, Ellwood R, Ismail AI. Evidence on existing caries risk assessment systems: are they predictive of future caries? Community Dent Oral Epidemiol 2013;41(1):67–78.
23. North Richmond Community Health. About us Melbourne: North Richmond Community Health; 2015 [cited 2015 May 16]. Available from: www.nrch.com.au/about.
24. World Health Organization. The Ottawa Charter for Health Promotion. Ottawa: World Health Organization; 1986 [cited 2015]. Available from: www.who.int/healthpromotion/conferences/previous/ottawa/en/.
25. Watt RG, Sheiham A. Integrating the common risk factor approach into a social determinants framework. Community Dent Oral Epidemiol 2012;40(4):289–96.

Appendix 2. Background published paper 2

Citation: *Hall M, Christian B. A health-promoting community dental service in Melbourne, Victoria, Australia: protocol for the North Richmond model of oral health care. Australian journal of primary health. 2017;23(5):407-14.*

CSIRO PUBLISHING
Australian Journal of Primary Health
https://doi.org/10.1071/PY17007
Practice & Innovation

A health-promoting community dental service in Melbourne, Victoria, Australia: protocol for the North Richmond model of oral health care

Martin Hall[A,C] *and Bradley Christian*[A,B]

[A]North Richmond Community Health Ltd, 23 Lennox Street, Richmond, Vic. 3121, Australia.
[B]Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University, Edwards Road, Flora Hill, Vic. 3550, Australia.
[C]Corresponding author. Email: martin.hall@dhsv.org.au

Abstract. Despite the best efforts and commitment of oral health programs, there is no evidence that the current surgical output-based model of oral health care is delivering better oral health outcomes to the community. In fact, Australian evidence indicates the oral health of the community could be getting worse. It is now well-understood that this traditional surgical model of oral health care will never successfully manage the disease itself. It is proposed that a health-promoting, minimally invasive oral disease management model of care may lead to a sustainable benefit to the oral health status of the individual and community groups. The aim of this paper is to describe such a model of oral health care (MoC) currently being implemented by the North Richmond Community Health Oral Health (NRCH-OH) program in Melbourne, Victoria, Australia; this model may serve as a template for other services to re-orient their healthcare delivery towards health promotion and prevention. The paper describes the guiding principles and theories for the model and also its operational components, which are: pre-engagement while on the waitlist; client engagement at the reception area; the assessment phase; oral health education (high-risk clients only); disease management; and reviews and recall.

Received 21 January 2017, accepted 14 June 2017, published online 21 August 2017

Introduction

Despite the best efforts and commitment of oral health programs, there is no evidence of a reduction in oral diseases and corresponding dental treatment required by the Australian community. In fact, Australian evidence indicates the oral health of the community could be getting worse (Brennan 2008), particularly among children, with dental caries prevalence increasing by 26% among 6-year-olds and 14% among 12-year-olds (Spencer and Harford 2008). Caries severity (average number of teeth per child that experienced caries) among 6-year-olds increased from 1.45 (1996) to 2.58 (2010). Among 12-year-olds, caries severity increased from 0.83 in 1998 to 1.34 in 2010 (Mejia *et al.* 2012, Chrisopoulos *et al.* 2016). Concurrently, a 60% increase was observed in the number of disease-free children, which indicates that dental caries is increasingly concentrated in certain sections of the community that are considered to be at high risk of dental caries (Spencer and Harford 2008).

The current model of oral health service delivery in Australia needs to change for several reasons: (A) population demographics are rapidly changing, with a projected increase in Australia's population of 56% by 2056 (Australian Bureau of Statistics 2009); increasing overseas migration (Australian Bureau of Statistics 2013); and the doubling of the proportion of people over 65 years by 2056 (Australian Bureau of Statistics 2009). This will result in increased complexity of care as people live longer and retain more teeth, and may see a probable increase in the proportion of eligible patients. There is concern that such changes will put immense and unsustainable pressure on an already overextended healthcare system (Australian Institute of Health and Welfare 2014*b*). A key challenge will be to manage non-communicable diseases, which are more prevalent among the elderly (65+ years), and include conditions such as oral diseases, diabetes, heart, stroke and vascular diseases; arthritis; cancers; and hypertension (Australian Institute of Health and Welfare 2014*a*). In addition, dementia is a significant health issue among older Australians, with the prevalence expected to double by 2050 (Australian Institute of Health and Welfare 2012). Oral health significantly affects overall health and quality of life, and is a key factor for healthy ageing (US Department of Health and Human Services 2000). Evidence shows that people visit the dentist more often as they grow older (Christian *et al.* 2013). It is well documented that newly arrived refugees and migrants are also at a greater risk of oral diseases because of their socio–cultural–economic–environmental backgrounds (Davidson *et al.* 2006). Dental services need to be prepared for these challenges and opportunities that will accompany the projected change in demographics by re-orienting their service delivery

What is known about the topic?

- The current traditional surgical model of oral health care does not treat oral diseases, and as such, will not improve oral health.

What does this paper add?

- This paper provides health service planners, policymakers and practitioners, with a protocol for an innovative health-promoting, minimally invasive model of oral health care, which will enable them to re-orient their service delivery away from the traditional surgical model of oral health care to one that is based on the principles of health promotion and prevention.

models of care (Christian *et al.* 2015), especially if effectiveness and efficiency are to be maintained in a financially constrained public dental service environment (Dental Health Services Victoria 2012). (B) It is now well-understood that the traditional surgical model of oral health care will never successfully manage the disease itself (Christian *et al.* 2015). This is because its focus is on the symptomatic surgical treatment of presenting disease pathology (such as cavities) and does not attempt to manage the causative risk and protective factors for diseases such as caries and periodontitis. As a result, oral diseases and the disparities in disease levels between population subgroups continue to be highly prevalent (Spencer and Harford 2008; Christian *et al.* 2015). A study by Nadanovsky and Sheiham (1995) showed that dental services explained only ~3% of the differences in change in 12-year-old caries levels across several countries. Socioeconomic factors had the largest effect independent of the use of fluoridated toothpaste. Hence, for effective disease management and sustained oral health outcomes, it is necessary to manage underlying risk factors as well as the disease presentation (Christian *et al.* 2015). It is proposed that a risk-based, minimally invasive oral disease management model of care may lead to a sustainable benefit to the oral health status of the individual and community group (Calache *et al.* 2013). (C) The chances of success in reducing the risk of oral disease is greatly increased by considering caries and periodontal disease to be 'behavioural diseases with a bacterial component' (Featherstone 2004) and empowering clients to self-manage and take ownership of their health by educating and involving them as equal partners in the treatment planning processes; thereby, making personal behaviour change key in management strategies.

The purpose of this document is to describe North Richmond Community Health's Oral Health model of care (NRCH-OH MoC) in terms of its intellectual and operational aspects. Although reasonably easy to describe, embedding this shared understanding within the everyday clinical situation is challenging. The process of refining and integrating this model of care into daily practice is ongoing through action research based on staff and client feedback, regular group discussion and integrating an evaluation system to monitor the model's performance.

The North Richmond Community Health oral health model of care (NRCH-OH MoC)

Program philosophy, theory and principles

The development of the NRCH-OH MoC was guided by assumptions conducive to good health. The first assumption was, if clients are empowered to look after their own oral health effectively, then this would translate into better oral health outcomes, more successful treatment outcomes and more efficient management of health services. However, changing unhealthy behaviour can be a challenging, time-consuming process that requires specialist techniques such as motivation interviewing (Harvard Women's Health Watch 2007). The second assumption relates to client need; that is, the service providers should cater to the client's needs rather than what the clinician determines as being important for the client (client-centred care) (Australian Commission on Safety and Quality in Health Care 2016). It is common practice for dental practitioners to come up with a treatment plan based on their assessment and not focus on the client's needs. This plan may not be what the client wants, but, because of the perceived clinician–client power relationship, clients agree to take the practitioner's treatment recommendations. The third assumption, which relates to the second, is that clients should have input into their oral healthcare plan. This makes them equal partners in the decision process, addresses the power dynamic and gives the client a sense of ownership over their health.

The theoretical frameworks that informed this model of care included: (1) the Health Belief Model (HBM); and (2) the Community Organisation and Community Building Theory (COCBT). The widely used HBM theoretical framework guided our approach to client behaviour change and was developed in the 1950s in an attempt to rationalise why people fail to participate in health programs (Champion and Skinner 2008). The theory of the HBM is: 'If individuals regard themselves as susceptible to a condition, believe that condition would have potentially serious consequences, believe that a course of action available to them would be beneficial in reducing either their susceptibility to or severity of the condition, and believe the anticipated benefits of taking action outweigh the barriers to (or costs of) action, they are likely to take action that they believe will reduce their risks' (Champion and Skinner 2008). The COCBT was used to guide the re-orientation of the health service towards a more preventive minimally invasive approach to disease management. The 'community' in this instance reflects the health service and its various operational aspects including, but not limited to: service accessibility, service quality, client health outcomes, service economics, organisation policy, workforce and workforce structure (Glanz *et al.* 2008).

Motivational Interviewing (MI) is a method that works on facilitating and engaging intrinsic motivation within the client in order to change behaviour. This model of care utilises MI in a goal-oriented and client-centred counselling approach to eliciting behaviour change by helping clients to explore and resolve ambivalence. Client are presented with evidence and relevant information incorporating an active listening approach to enable them to develop skills and make the behaviour change required. The measure of acceptance of this model of care is by questionnaire, which the client completes at the end of the

oral health education sessions. This model utilises the clients' feedback to continuously improve the processes and client flow.

Ultimately, it is the client who decides to participate in this model of care. 'Drop in' clinics or *ad hoc* emergency visits are available to those rare clients who reject the preventive approach, with treatment provided using a more traditional approach. This model does not work as well for clients who have limited capacity for a sustainable change to self-management. Clients with significant mental health conditions or chronic substance abuse issues do not automatically enter the pathway requiring behaviour changes. Their inclusion is dependent on further discussions with their established dental practitioner. The principles guiding the NRCH-OH MoC are: a client- and family-centred approach, team-based care, innovative use of staff and resources, health promotion, prevention, risk-based access and periodic program evaluation (Christian *et al.* 2015).

Program operating context

Public dental services

Public dental services in Australia are provided to eligible low-income individuals and pre-determined priority groups by each State through combined State-funding system and Commonwealth partnership agreements (Australian Government Department of Health dental website, see http://health.gov.au/dental, accessed April 2016). However, eligibility criteria as well as administration of services varies between States. Dental Health Services Victoria (DHSV), funded by the State Department of Health and Human Services, administers public dental services in Victoria at the Royal Dental Hospital and also by sub-contracting to community health services such as NRCH (Dental Health Services Victoria website, see https://www.dhsv.org.au/, accessed September 2016). The introduction of National Health Care reforms has resulted in a range of changes to how dental health services are funded within Australia, and is supported through the Council of Australian Governments (COAG). DHSV's discussion paper on Innovative Models of Care was an inspiration to re-orient the NRCH-OH MoC towards prevention and health promotion (Dental Health Services Victoria 2012).

Economics

The funding allocation to the Victorian public dental services is fixed each year by Treasury, with DHSV redirecting an amount to each dental agency based on the number of effective full-time equivalent dental practitioners employed and the predicted achievement of the year's productivity targets. This output-based system uses clinical item codes, which are nominated a proportion of a funding unit known as the Dental Weighted Activity Unit (DWAU). Each DWAU is funded at a given dollar value set by the State Government, and the funds are then distributed to the dental program based on their total monthly clinical output. Benchmarks are set by DHSV, with dentists and therapists expected to achieve a predetermined amount of DWAUs per year. However, the nominated DWAU to item codes is biased towards restorative and prosthetic activities rather than preventive activity and, therefore, the challenge for financial sustainability is significant when operating under a preventive model of care. The funding of the DWAU is, however, the same whoever is legitimately 'charging' through to DHSV, therefore matching the appropriate practitioner to the activity is critical in terms of cost benefit for a particular model of care. For example, when a dental assistant with an extended scope of practice is providing care, these DWAUs are accepted and charged accordingly. The model of care therefore works best using an all-of-team approach, and this is supported by the Australian Health Practitioner Regulation Agency (AHPRA) through a Structured Professional Relationship in which each practitioner works independently within their individual scope of practice and refer within the team when work outside their scope is required (Dental Board of Australia 2014).

Traditional models of oral health care

Traditional models of oral health care have focused more on the surgical management of existing pathology in a tertiary prevention or downstream approach. This approach does less to manage the dental disease itself and, therefore, has had no effect on the rate of hospitalisation, the inequitable distribution of dental diseases and waiting list times for treatment in public dental services (Health Issues Centre 2009, Kruger and Tennant 2016). At present, this model still defines the majority of dentistry performed globally, including that within Australia. For both providers and consumers, there will be considerable challenges to move from this surgical-based program funded to manage disease symptoms by 'fixing teeth' to a health promotion, client-centred, health-coaching and risk-based model of care.

Program policies

Implementation of the NRCH-OH MoC is informed by a range of policies such as:

- Prioritising access for high-risk individuals and groups.
- Providing new clients, who go on the waiting list, with a basic information sheet on tooth brushing.
- Provision of oral health education, including individualised oral hygiene instruction by a specially trained dental assistant, to all high-risk clients.
- Achievement of a minimum standard of home care, as determined by client plaque scores, before proceeding to other phases of care.
- Provision of minimally invasive dentistry, including Caries Management by Risk Assessment (CAMBRA) (Featherstone 2004).
- All child recalls are based on a risk assessment for future disease.
- All adult recalls and reviews are based on a risk assessment for future disease.
- Drop-in clinics are made available to all clients.

Program operational components

Fig. 1 illustrates how a client flows through the NRCH-OH MoC. The management pathway is determined by the client's assessed risk status. Clients assessed as being at 'Extreme risk', such as those with non-modifiable risk factors for oral disease or complex medical conditions, are managed by a senior dentist working with a selected sub-team including, when appropriate, specialists from the dental hospital.

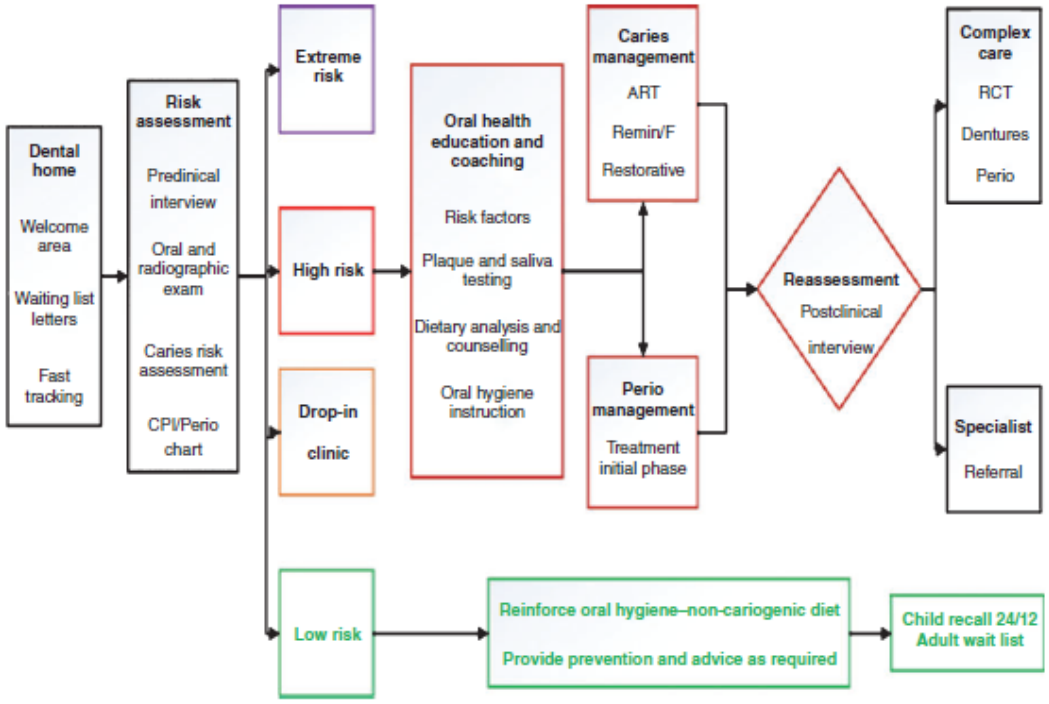


Fig. 1. Flow of clients through North Richmond Community Health's Oral Health model of care (NRCH-OH MoC).

If a client does not wish to participate in the model of care, this will usually become apparent at the assessment and management planning stage or when the client fails to attend the initial education appointment. For these clients, complex care such as root canal therapy, partial dentures or fixed prosthetic work is not offered. It is important that clients understand why certain treatment is not recommended and how it can be inappropriate to proceed with treatment that is more complex while disease remains active and when risk factors have not been addressed.

The following steps describe each stage of the NRCH-OH MoC.

Step 1. Pre-engagement while on the waitlist

It is important that clients are prepared for this health-promoting approach and especially made aware that some treatments may be delayed until they demonstrate a minimum standard of self-care. This message is delivered at least twice, while the client is on the waitlist, by information sheets, which are mailed to all new clients as they go onto the waiting list. Another letter is sent with an appointment date and time when the client reaches the top of the waiting list.

Step 2. Reception

The reception team is the starting point of the client experience and a welcoming, respectful and client-focused approach is expected. A well-presented, tidy and informative welcome area (not waiting area) is available to clients before entering a clinical room. The Reception Team is also trained and expected to reinforce information and recommendations on the appropriate use of home products, such as: fluoride toothpaste, bioavailable calcium and phosphate-releasing paste, fluoride mouth rinses and chlorhexidine rinse.

Step 3. Assessment phase

The assessment phase includes a pre-clinical interview, an oral health examination and the initial management plan. The oral health examination includes a risk assessment for caries and periodontal disease.

The preclinical interview is conducted with all new clients and returning clients on high-risk recall, preferably in a private consulting room where medical, dental, family and social history and dietary information is collected and recorded. Client expectations from the dental visit are also captured at this stage. The dental assistant, from the most appropriate cultural group or who is familiar with specific client health beliefs, will greet the client, conduct this interview in the client's first language if appropriate and continue to support the client throughout their episode of care.

This is followed by a full clinical and radiographic assessment usually conducted by an oral health therapist. A caries risk assessment tool, based on CAMBRA, is used to record information and to assign the client to a caries risk level (Featherstone 2004). Risk for periodontal disease is assessed using the Community Periodontal Index (CPI) (Cutress *et al.* 1987). The client is given a risk rating as either extreme (non-modifiable risk factors) or high or low risk to avoid clients delegated to the 'safe option' of the moderate classification. Assessment of risk offers the dental practitioner information on

the likelihood of a client developing future oral disease, progression of the disease and whether the recall interval should be adjusted to align with risk factors. It also assists clients to recognise the modifiable factors contributing to their disease, which may enable them to take actions to reduce that risk. However, as the evidence to support existing risk assessment tools is not clear (Tellez *et al.* 2013), the tools used in this model are used to support an intellectual process, with the dental practitioner's experience and judgement contributing to the final assessment.

At this point, the information from the assessment phase will be discussed with the client. The dental practitioner will determine whether time is allocated for oral health education and will use information gained from this session to ensure targeted oral health education activities. In private practice, the consumer pays money for the agreed care. In this model of care, outcomes rely to some degree at least on compliance, with money and payments less important. There will be management issues for both providers and receivers of care, and potential for blaming between staff, and between staff and clients where improvements are not achieved or sustained. In all cases, the oral health management plan is negotiated with the client and their input influencing realistic goals. Discussing expectations and understanding the value of compliance are critical for achieving improved oral health outcomes. If agreed outcomes are not achieved or only partially achieved during education sessions, the client is referred to the senior dental practitioner for further management planning. Oral health management plans may change as risk factors reduce, which can be an additional motivating influence for improved oral hygiene and diet modification for the client.

Step 4. Oral health education

High risk clients only. The oral health education session, a unique feature of this model, is conducted by dental assistants who have had special training in health promotion (Certificate IV), which qualifies them as an oral health educator (OHE) and enables funding by DWAUs. All clients assessed as high risk for either caries, periodontal disease or both are referred to the OHE, who reviews the client's pre-clinical interview questionnaire and handover notes from the dental practitioner to determine relevant factors contributing to the client's high-risk status. They then provide tailored education sessions, which include: plaque and saliva testing; instruction on the use of preventive products (high-fluoride tooth paste, bioavailable calcium and phosphate releasing paste); dietary analysis and advice; and review and follow up. Saliva and plaque scores, when indicated or suggested by the dental practitioner, are conducted by the OHE and contribute to the clients' understanding of their risk. These sessions are conducted in a collaborative approach, in which goals for change are agreed upon together rather than instructions directed from 'above'. All high-risk clients must demonstrate a good level of oral hygiene before the next phase of their management plan. The number of review visits required to achieve this is determined by the OHE in negotiation with the client, and when appropriate, the review visits may be combined with other dental visits. Reviews are set by the OHE based on factors such as health literacy, competency, motivation, medical history and family and home factors.

Step 5. Management

Management is based on risk, with higher risk clients having more intense management (Table 1).

Extreme caries (CE) management. Clients who are undergoing head and neck radiotherapy, cancer therapy, or who have major social or mental disability, chronic substance abuse, Sjogren's syndrome, uncontrolled diabetes, etc., require specialised care and a senior dentist must be consulted in the management plan. A more integrated plan and specialised management will usually be required to address these clients' ongoing risk and treatment needs. Recalls and reviews for this category are determined on a case-by-case basis. Short-term expectations and outcomes will vary for these clients depending on the whether any risk factors are modifiable; however, it is unlikely that these clients will move to a lower-risk category given their special circumstances.

Table 1. Management strategies by risk status

ART, Atraumatic Restorative Treatment; MID, Minimal Intervention Dentistry; OH, Oral Health; OPG, Orthopantomogram

Code	Description	Oral health education	Restorative	Periodontal	Prosthetic	Recall
CE	Extreme caries	•Depends on risk factors	•Caries control ART	•Depends on risk factors	•Consider full denture	Ongoing or *ad hoc*
C1 P1	High caries–high periodontal	•OH education plaque scores saliva testing diabetes risk smoking cessation reviews	•Caries control ART (until risk reduced) routine care using MID principles	•Periodontal charting OPG initial therapy specialist referral	•Only when maintaining adequate OH	6–12 months
C1 P2	High caries–low periodontal	•OH education plaque scores saliva testing reviews	•Caries control ART (until risk reduced) routine care using MID principles	•Routine scale and clean	•Only when maintenance of adequate OH	6–12 months
C2 P1	Low caries–high periodontal	•OH education plaque scores diabetes risk smoking cessation reviews	•Routine care using MID principles	•Periodontal charting OPG initial therapy specialist referral	•Only when maintenance of adequate OH	6–12 months
C2 P2	Low caries–low periodontal	•Not applicable	•Routine care using MID principles	•Routine scale and clean	•Yes	Waiting list after 12 months

High caries–high periodontal (C1P1) management. This involves the following:

- Preventive phase: the OHE manages the oral hygiene and dietary risk factors. An oral hygiene plan is developed with the client and includes monitoring, plaque scores, oral hygiene instruction and nutritional and quit smoking advice, with additional dedicated smoking cessation staff available on referral.
- Periodontal management requires full charting and initial supragingival debridement by a hygienist. A specialist periodontist referral is included, as appropriate.
- Caries control: the dental practitioner may choose stabilisation of active carious lesions by Atraumatic Restorative Treatment (ART) and application of silver diamine fluoride (SDF) during this initial preventive phase (Alliance for a Cavity-Free Future 2016).
- The OHE reviews the client for ongoing dietary analysis and nutritional support, and when plaque scores meet minimum standards, refers the client to the dental practitioner.
- Restorative and prosthetic phases: the dental practitioner has the option at any stage to return the client to the OHE for further visits or can opt to continue to finish depending on the client's compliance. Prosthetic treatment commences, if required, on completion of the restorative phase. Further reviews take place with the OHE as required and there is a short recall with the dental practitioner to monitor risk status.

High caries–low periodontal (C1P2) management. This involves the following:

- Preventive phase: the OHE manages the oral hygiene and diet risk factors. An oral hygiene plan is developed with the client.
- Caries control: the dental practitioner may choose stabilisation, ART, SDF during the initial preventive phase.
- The OHE reviews client for ongoing dietary analysis and nutritional support, and when plaque scores meet minimum standards, refers the client back to the dental practitioner.
- Restorative and prosthetic phases. The dental practitioner has the option to return the client to the OHE for further visits or can opt to continue to finish. Further reviews take place with the OHE as required and there is a short recall with the dental practitioner to monitor risk status.

Low caries–high periodontal (C2P1) management. This involves the following:

- Any routine restorative work is performed.
- Preventive phase: managed by the OHE.
- Periodontal management requires full charting and initial supragingival debridement by the hygienist. A specialist periodontist referral is included, as appropriate.
- Further reviews take place with OHE or short recall with the dental practitioner.

Low caries–low periodontal (C2P2) management. This involves the following:

- The dental practitioner develops a management plan with the client.

Step 6. Reassessment, post-clinical interview and recalls

Re-assessment and post-clinical interviews are conducted on completion of an episode of care, to ensure the episode of care was implemented as planned and also for comparison against the pre-clinical interview. Client risk status is also re-assessed at this point, which determines the recall interval.

The frequency and type of oral health care a client needs depends on the likelihood of disease developing. The ability to assign an appropriate recall interval will improve over time as the dental practitioner builds an accurate picture of the client and determines the rate at which disease is developing. As records will not exist for new or recent clients, there may be some uncertainty about what recall interval is appropriate. In these cases, a conservative recall interval (6–12 months) is assigned and progressively altered over time on the basis of the risk assessment performed at each assessment visit. It is advisable to inform a child as well as their parents, guardians or caregivers that the same interval may not be appropriate at every stage of their life – it will vary if their risk and protective factors alter.

Adult recalls can be a contentious issue and Government policy is that adults are not recalled but that they should approach the service in 12 months to go onto the waiting list, which can be at least another 12-month wait. However, in the preventive minimally invasive NRCH-OH MoC, duty of care requires that clients return for regular visits until their risk status is modified to low risk. Therefore, recall periods determined by mutual agreement with high-risk clients and dental practitioners are used. NRCH-OH takes the responsibility to contact clients placed on recall, inviting them to attend.

Discussion and conclusion

There will be many challenges and complexities to overcome when delivering a new model of care. Health services have to balance the costs of running an oral health program while providing effective, evidence-informed and culturally appropriate services. Public oral health programs, in most instances, operate on a limited budget, and programs need to be dynamic and innovative to ensure efficient use of these limited available funds. The NRCH-OH program achieves this by utilising: public health principles, oral health promotion strategies, innovative use of staff, risk-based management and minimally invasive dentistry; yet, the program continues to be challenged with a finite budget. As a result, the program is always exploring ways to deliver the best outcomes for the lowest cost and this is where robust economic evaluation can provide the evidence to inform decision-making.

The current public dental service system in Australia is output driven, and there is no evidence that this system and associated policies are delivering the intended health outcomes. Health services should be held accountable if they do not deliver positive health outcomes. To ensure quality services, new models of oral health care will need to move away from being output driven to one that focuses on value and outcomes (Community Health Services Forum 2014). Achieving positive health outcomes in the most cost-effective approach should be the end goal for any health service provider, yet, outcomes are rarely measured or reported on and existing services tend to be profit driven (Community Health Services Forum 2014). In addition, with costs and expenditures for health care increasing (Christian and Chattopadhyay 2014), it is necessary for services to remain viable and efficient by delivering the best outcomes for the lowest cost.

In these modern times, efficiency and effectiveness in the healthcare system can be improved by the innovative use of information technology (Ortiz and Clancy 2003), with applications such as: electronic record systems, handheld electronic devices, electronics to support clinical decision-making, practice-based data for research to build evidence-base, advanced data analytics to inform decision-making and electronic patient self-monitoring systems (Ortiz and Clancy 2003). The health service delivery system will need to adapt and adopt these new information technologies to be relevant to the times, and to ensure that clients are receiving the best quality care possible.

There will also be considerable challenges and complexities to re-orient the dental professional's mindset away from the traditional surgical model of care to a more preventive-based approach. Some key barriers to this change are: economics, lack of time, education system focused on surgical intervention, healthcare systems oriented towards surgical intervention, and peers' opinion valued higher than research (Sbaraini *et al.* 2013). Research has also shown, that, although dentists are good at attending professional development opportunities, they take a long time to implement innovative evidence-informed practices (Chapko 1991).

It is time for change, and the Oral Health Program at North Richmond Community Health looks forward to addressing some of these challenges through the next phase in the development and refinement of this model of care, through an evaluation of the model's processes. This will be followed by a comprehensive comparative evaluation to test the effectiveness (including cost-effectiveness) of the NRCH-OH model of care.

Conflicts of interest

The authors declare that they have no conflicts of interest.

Acknowledgements

The authors thank Mr Demos Krouskos, CEO NRCH, Ms Emily Chalmers-Robinson and the staff of North Richmond Community Health Ltd. Prof. Hanny Calache, Head, Oral Health Research Stream, Deakin Health Economics, Centre for Population Health Research, Faculty of Health for their cooperation, support and advice.

References

Alliance for a Cavity-Free Future (2016) Silver diamine fluoride. (ACFF: London, UK) Available at http://www.allianceforacavityfreefuture.org/en/us/technologies/silver-diamine [Verified September 2016]

Australian Bureau of Statistics (2009) Future population growth and ageing. Australian social trends. Australian Bureau of Statistics: Canberra, ACT, Australia.

Australian Bureau of Statistics (2013) Population projections, Australia, 2012 (base) to 2101. (Australian Bureau of Statistics: Canberra, ACT, Australia) Available at http://www.abs.gov.au/ausstats/abs@.nsf/Lookup/3222.0main+features42012%20(base)%20to%202101 [Verified April 2016]

Australian Commission on Safety and Quality in Health Care (2016) 'Patient and consumer centred care.' (ACSQHC: Sydney, NSW, Australia).

Australian Institute of Health and Welfare (2012) 'Dementia in Australia.' (Australian Institute of Health and Welfare: Canberra, ACT, Australia).

Australian Institute of Health and Welfare (2014*a*) Australia's health 2014. The 14th biennial health report of the Australian Institute of Health and Welfare. Australian Institute of Health and Welfare, Canberra, ACT, Australia.

Australian Institute of Health and Welfare (2014*b*) Australia's health 2014. Ageing and the health system: challenges, opportunities and adaptations. Australia's health series number 14. Australian Institute of Health and Welfare, Canberra, ACT, Australia.

Brennan DS (2008) Oral health of adults in the public dental sector. Dental statistics and research series number 47. Australian Institute of Health and Welfare, Canberra, ACT, Australia.

Calache H, Hopcraft MS, Martin JM (2013) Minimum intervention dentistry–a new horizon in public oral health care. *Australian Dental Journal* **58**, 17–25. doi:10.1111/adj.12046

Champion VL, Skinner CS (2008) The health belief model. In 'Health Behavior and Health Education: Theory, Research and Practice'. (Eds K Glanz, BK Rimer, K Viswanath) pp. 45–62. (Jossey-Bass: San Francisco, CA, USA).

Chapko MK (1991) Time to adoption of an innovation by dentists in private practice: sealant utilization. *Journal of Public Health Dentistry* **51**, 144–151. doi:10.1111/j.1752-7325.1991.tb02205.x

Chrisopoulos S, Harford J, Ellershaw A (2016) Oral health and dental care in Australia: key facts and figures 2015. Catalogue number DEN 229. Australian Institute of Health and Welfare, Canberra, ACT, Australia.

Christian B, Chattopadhyay A (2014) Determinants and trends in dental expenditures in the adult US population: Medical Expenditure Panel Survey 1996–2006. *Community Dental Health* **31**, 99–104.

Christian B, Chattopadhyay A, Kingman A, Boroumand S, Adams A, Garcia I (2013) Oral health care services utilisation in the adult US population: Medical Expenditure Panel Survey 2006. *Community Dental Health* **30**, 161–167.

Christian B, Hall M, Martin R (2015) A paradigm shift in models of oral health care: an example and a call to action. *Family Medicine and Community Health* **3**, 32–37. doi:10.15212/FMCH.2015.0131

Community Health Services Forum (2014) 'Briefing: Beginning with the End in Mind. How Outcomes-Based Commissioning can help Unlock the Potential of Community Services.' (NHS Confederation: London, UK).

Cutress TW, Ainamo J, Sardo-Infirri J (1987) The community periodontal index of treatment needs (CPITN) procedure for population groups and individuals. *International Dental Journal* **37**, 222–233.

Davidson N, Skull S, Calache H, Murray SS, Chalmers J (2006) Holes a plenty: oral health status a major issue for newly arrived refugees in Australia. *Australian Dental Journal* **51**, 306–311. doi:10.1111/j.1834-7819.2006.tb00448.x

Dental Board of Australia (2014) Guidelines for scope of practice. (Australian Health Practitioner Regulation Agency, AHPRA: Melbourne, Vic., Australia) Available at http://www.dentalboard.gov.au/Codes-Guidelines/Policies-Codes-Guidelines/Guidelines-Scope-of-practice.aspx [Verified 14 July 2017]

Dental Health Services Victoria (2012) Establishing innovative models of care: a discussion paper. Dental Health Services Victoria, Melbourne, Vic., Australia.

Featherstone JD (2004) The caries balance: the basis for caries management by risk assessment. *Oral Health & Preventive Dentistry* **2**(Suppl 1), 259–264.

Glanz K, Rimer BK, Viswanath K (Eds) (2008) 'Health Behavior and Health Education: Theory, Research and Practice.' (Jossey-Bass: San Francisco, CA, USA)

Harvard Women's Health Watch (2007) Why it's hard to change unhealthy behaviour – and why you should keep trying. Researchers say successful change comes only in stages. How long it takes is an individual matter. *Harvard Women's Health Watch* **14**, 4–5.

Health Issues Centre (2009) 'Was I still on the waiting list?' A study about people waiting for public dental care. Health Issues Centre, Melbourne, Vic., Australia.

Kruger E, Tennant M (2016) Ten years of hospitalisation for oral health-related conditions in Western Australia: an unjust dichotomy. *Australian Journal of Primary Health* **22**(2), 153–158. doi:10.1071/PY14087

Mejia GC, Amarasena N, Ha DH, Roberts-Thomson KF, Ellershaw AC (2012) Child Dental Health Survey Australia 2007: 30-year trends in child oral health. Dental Statistics and Research Series Number 60. Catalogue Number DEN 217, Australian Institute of Health and Welfare, Canberra, ACT, Australia.

Nadanovsky P, Sheiham A (1995) Relative contribution of dental services to the changes in caries levels of 12-year-old children in 18 industrialized countries in the 1970s and early 1980s. *Community Dentistry and Oral Epidemiology* **23**, 331–339. doi:10.1111/j.1600-0528.1995.tb00258.x

Ortiz E, Clancy CM (2003) Use of information technology to improve the quality of health care in the United States. *Health Services Research* **38**, xi–xxii. doi:10.1111/1475-6773.00127

Sbaraini A, Carter SM, Evans RW, Blinkhorn A (2013) How do dentists and their teams incorporate evidence about preventive care? An empirical study. *Community Dentistry and Oral Epidemiology* **41**, 401–414.

Spencer AJ, Harford J (2008) 'Improving Oral Health and Dental Care for Australians.' (Australian Research Centre for Population Oral Health: Adelaide, SA, Australia)

Tellez M, Gomez J, Pretty I, Ellwood R, Ismail AI (2013) Evidence on existing caries risk assessment systems: are they predictive of future caries? *Community Dentistry and Oral Epidemiology* **41**, 67–78. doi:10.1111/cdoe.12003

US Department of Health and Human Services (2000) Oral health in America: a report of the Surgeon General. US Department of Health and Human Services, Rockville, MD, USA.

Appendix 3. Research Strand 1, published paper: Systematic Review

Citation: *Christian B, Armstrong R, Calache H, Carpenter L, Gibbs L, Gussy M. A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. International Journal of Paediatric Dentistry. 2019;29(2):106-16.*

Received: 16 July 2018 | Revised: 9 October 2018 | Accepted: 5 November 2018
DOI: 10.1111/ipd.12446

REVIEW ARTICLE

WILEY INTERNATIONAL JOURNAL OF PAEDIATRIC DENTISTRY

A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children

Bradley Christian[1,2] | **Rebecca Armstrong**[1] | **Hanny Calache**[3] | **Lauren Carpenter**[1] | **Lisa Gibbs**[1] | **Mark Gussy**[2]

[1]Jack Brockhoff Child Health and Wellbeing Program, Centre for Health Equity, Melbourne School of Population and Global Health, The University of Melbourne, Melbourne, Victoria, Australia
[2]Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University, Bendigo, Victoria, Australia
[3]Deakin Health Economics, Centre for Population Health Research, Deakin University, Burwood, Victoria, Australia

Correspondence
Bradley Christian, Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University, Bendigo, Vic., Australia.
Email: b.christian@latrobe.edu.au

Funding information
Centre for Health Equity, Melbourne School for Population and Global Health, The University of Melbourne and La Trobe Rural Health School, La Trobe University for open access

Background: At present, there are numerous caries risk assessment tools (CRATs) being promoted for disease management. The evidence to inform CRAT selection however is unclear.
Aim: This review aimed to assess the strength of evidence to inform the selection of CRATs for children ages 6 years and less.
Design: MEDLINE was the principal search database for this review. Other key databases, the reference lists of included articles, known cariology literature and experts were also consulted. Peer-reviewed papers describing CRATs and their development methodology were included. The Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist guided the quality assessment. The reporting of the key measurement properties (reliability, validity, and responsiveness) informed the quality assessment.
Results: The search resulted in 10 papers, reporting on eight different CRATs. The identified CRATs were as follows: Caries Management By Risk Assessment (CAMBRA), Cariogram, National University of Singapore CRAT (NUS-CRAT), MySmileBuddy, Dundee Caries Risk Assessment Model, University of North Carolina Risk Assessment Models, University of Michigan paediatric dental clinic caries risk assessment sheet, and American Academy of paediatric Dentistry (AAPD) CRAT. Common across all CRATs was the lack of information to determine the levels of evidence for the measurement properties of reliability and construct validity. Studies on tools that were assessed as having strong evidence for content validity identified the relevant risk factors for caries in the population being studied, before developing and testing their respective CRATs.
Conclusions: The evidence to inform the selection of current CRATs for children is mostly yet to be established. Overall, the NUS-CRAT studies reported the most information to inform the assessment of its measurement properties, and as a result, this tool attained a higher quality rating than other CRATs studied.

1 | INTRODUCTION

Dental caries is a complex multi-factorial disease. The Fisher-Owens conceptual model illustrates this complexity by showing the various levels and factors that influence child oral health.[1] The distinction between caries risk assessment methods (CRAMs) and caries risk assessment tools (CRATs) is important for this topic. A CRAM is any

This is an open access article under the terms of the Creative Commons Attribution License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited.
© 2018 The Authors. *International Journal of Paediatric Dentistry* published by BSPD, IAPD and John Wiley & Sons Ltd.

method that uses any individual risk or protective factor to predict caries development or progression. Two recent systematic reviews on CRAMs report on the accuracy of these various methods in caries prediction.[2,3] To further improve caries prediction, various combinations of CRAMs have been developed into so-called caries risk assessment tools (CRATs) or systems (the term "tools" will be used in this paper). At present, there are a variety of CRATs used in dentistry. Popular CRATs include the following: Caries Management By Risk Assessment (CAMBRA), American Association of Paediatric Dentistry Caries Assessment Tool (AAPD CAT), and Cariogram.[4-6] Conceptually, risk assessment seeks to determine an accurate and precise risk level categorisation for the purposes of identifying those at highest risk of disease, to guide treatment decisions, to determine appropriate recalls and health economics.[7]

For any health measurement instrument, be it an index test, clinical rating scale or a self/patient-reported health outcome measure, the key measurement properties that ensure the results of the tool are interpreted properly are validity (accuracy), reliability (precision), and responsiveness. Reliability is the property of the tool to produce similar results under different conditions, whereas validity is the property of the tool to measure what it claims to measure. Responsiveness is the ability of a tool to validly detect change in a measured construct over time. Reliability includes the domains of internal consistency and measurement error. Validity includes domains such as content, construct, and criterion validity.[8]

Most of the research on CRATs and CRAMs has focused only on validity (mostly limited to criterion validity (reported as sensitivity and specificity)) in predicting a caries outcome.[2,3,9] Without evidence for the quality of all relevant measurement properties for a CRAT, making an informed decision on tool selection is impossible. The authors of this review experienced this issue first hand, in an attempt to select a CRAT for use in a risk-based caries management model of care at a public dental service in Melbourne, Australia. Contemporary caries management is reliant on valid and reliable risk assignment, making the necessary first step to: use evidence to inform CRAT selection; and where required, generate the knowledge to inform the selection of CRATs. Without this essential body of work, risk-based models of caries management run the risk of not delivering the intended outcomes.

This is not a review of caries risk assessment methods (CRAMs) and is not limited to criterion validity. Rather, this review will focus exclusively on CRATs and the collation and assessment of evidence on the important measurement properties of CRATs for children under 6 years of age. This review offers a critical assessment of the methodological quality of included studies and the measurement properties reported in these studies to inform the selection of CRATs for young children.

Why this paper is important to paediatric dentists

- This paper identified current caries risk assessment tools for children 6 years and less, for which there is peer-reviewed published evidence.
- Paediatric dentists should be informed by evidence when selecting a caries risk assessment tool for use in risk-based caries management.
- This study also provides paediatric dentists with an understanding of the important measurement properties to consider when selecting a health measurement instrument.

The research question guiding this review was: What is the strength of evidence to inform the selection of CRATs for children ages 6 years and less? The objectives were as follows:

1. To identify existing CRATs for use with young children.
2. To assess the methodological quality of studies that assess the measurement properties of existing CRATs, including quality of the reported measurement property.
3. To assess the strength of evidence (overall quality) for a CRAT.
4. To identify gaps in the research on CRATs.

2 | MATERIALS AND METHODS

2.1 | Identification of studies

MEDLINE was the principal search database for this review. Other databases searched included CINAHL, PubMed, and Scopus. Key search terms were selected in consultation with experts in the field and a University librarian. These keywords were used to generate a Boolean search string, modified slightly for each database. The base Boolean string was—(caries or dental caries) AND (risk assessment or risk measur*) AND (measur* or tool*). In addition, we also conducted a phrase search for known caries risk assessment tools. The Boolean string for this search was—("caries management by risk assessment" OR CAMBRA OR cariogram OR "caries management system"). A University of Melbourne librarian familiar with the terminology in this field helped design and implement the search strategy.

The reference lists of articles selected for inclusion in the review and known cariology literature (including textbooks) were also screened to identify studies that may have been missed in

the electronic database search. Finally, experts (individuals and organisations) in the field were contacted to determine whether they had knowledge of publications on caries risk assessment tools that were not picked up by the other search methods.

2.2 | Study selection

Decisions on the inclusion and exclusion criteria (Table 1), to determine study selection, were guided by the review question and objectives. These criteria were finalised through periodic meetings of the review team, an iterative process that developed with literature familiarity. For example, the inclusion criterion "children ≤6 years" was introduced only at the full-text screening stage as the literature showed that this was a sufficiently different age cohort, in terms of having different risk/protective factors to older people,[10] to warrant a separate analysis. Several systematic reviews were identified but not included after application of inclusion/exclusion criterion. These excluded systematic reviews were screened for primary research papers that might not have been captured by the search strategy.

A two-step screening process was employed—title/abstract and full text. Two members (BC and LC) of the review team independently screened selected articles against the inclusion and exclusion criteria. Differences were resolved via discussion. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram was used to illustrate the flow of information through the study selection process and is shown in Figure 1.[11]

2.3 | Quality assessment

Several tools were considered for the quality assessment component of this review. The QUADAS (Quality Assessment for Diagnostic Accuracy Studies) tool has been used in previous systematic reviews on caries risk assessment methods.[12] As the name indicates, however, QUADAS focuses only on the single measurement property, validity, with the domains mainly related to criterion validity. For CRATs, content validity and construct validity are equally important. The QUADAS tool also has relatively more subjective questions than the COSMIN checklist and needs to be paired with another relatively subjective method like Grading of Recommendations Assessment, Development and Evaluation (GRADE) to determine the overall strength of evidence.[13] This review required a more objective method of quality assessment which focused on all the important measurement properties for CRATs. Therefore, the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist was used.[14]

COSMIN provides a standard set of items to inform the selection of health measurement instruments. The COSMIN checklist consists of nine boxes, to reflect important measurement properties (Appendix 1), with 5-18 items describing methodological standards for how each measurement property should be assessed.[14] For this review, the measurement properties of internal consistency, structural validity, and cross-cultural validity were not assessed as they were considered not applicable to CRATs. Internal consistency and structural validity are based on reflective models where all items that make up an instrument are a manifestation of the same underlying construct and, hence, are highly correlated,[8] whereas for CRATs the items together form the construct "risk" status and are not necessarily correlated. Cross-cultural validity was not assessed as it relates to the validity of translation of CRATs into other languages and this was not the study focus.

TABLE 1 Inclusion and exclusion criteria

Criterion	Inclusion	Exclusion
Population	Children ≤6 y of age (only applied at the full-text screening stage)	Special populations: People with a disability and chronic conditions, who are already at high risk and orthodontic patients
Index tests	Papers that describe CRATs or systems and their development processes—which can include evidence/theory behind the CRAT and the methodological development including the validity and reliability of the tool	Original research that only reports on caries risk-based management and does not discuss the application of the CRAT to assign risk and its development
Target conditions	CRAT to determine an individual's risk—any instrument to determine an individual's risk of developing future dental caries Coronal caries	No reference to a CRAT Population-level CRAT Tooth-level caries risk Fluorosis Periodontal disease Root caries
Study and information types	Peer-reviewed publications of original research that report on the application of CRATs to determine caries risk Non-scientific reports on CRATs Peer-reviewed thesis	Conference presentations and/or abstracts Commentaries Literature and systematic reviews Non-English publications

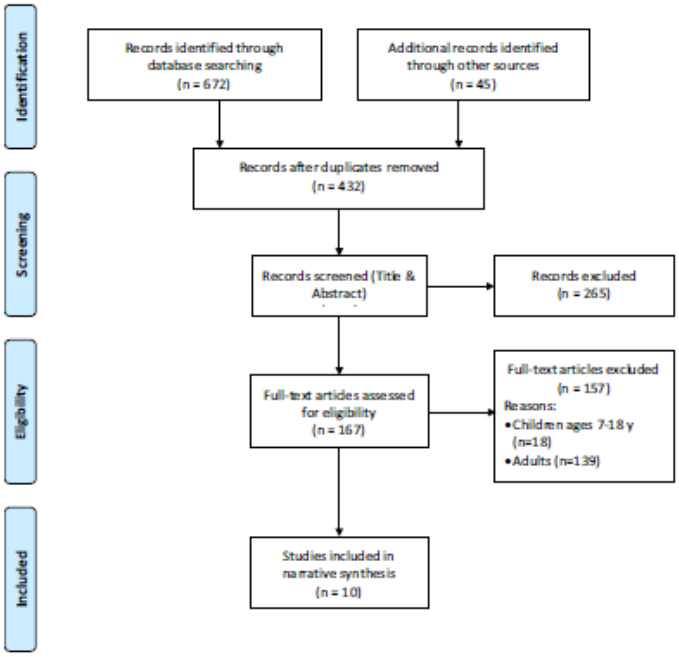


FIGURE 1 PRISMA flow diagram for study selection. Adapted from: Moher et al[30]

The quality assessment in this review was conducted in three steps. For these three steps, the quality assessment was performed by one assessor (BC) and 20% independently assessed by a second assessor (LC). Discrepancies were resolved by discussion.

2.4 | Step A. Assessing the methodological quality (risk of bias) of included studies

Firstly, the methodological quality of a study was evaluated using the COSMIN checklist with 4-point rating score (excellent, good, fair, and poor) and the overall score for a particular study was determined by the lowest rating of any item in the checklist for the particular measurement property.[14]

2.5 | Step B. Assessment of quality of measurement properties in each study by CRAT

The quality of the measurement properties in each study was assessed by CRAT using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (Appendix 2).[15] Minor revisions to the criteria were made to add clarity to the rating methodology and to also reflect criteria used to determine study quality rating in the COSMIN checklist. For example, for criterion validity "Sens+Spec ≥160" was added to the criteria for a "+" rating. This criterion of Sens+Spec ≥160 is generally accepted in the literature as being appropriate for a tool to predict caries.[16]

2.6 | Step C. Assessment of the overall strength of evidence, by measurement property, for a CRAT

The overall strength of evidence to support the selection of a CRAT was assessed using criteria as described in COSMIN's protocol for systematic reviews of measurement properties (Appendix 3).[15] This rating was informed by the two previous quality assessment steps. For example, an "Excellent" study rating for criterion validity in Step A and a "–" rating in Step B, for quality of the measurement property criterion validity, achieved an overall rating of "−−−" in Step C (this step) of the quality assessment. This was interpreted as strong evidence of a negative rating for criterion validity in one study of excellent methodological quality.

2.7 | Data synthesis and analysis

The analysis assessed and compared the methodological quality of included studies on CRATs, followed by a comparison of the overall strength of evidence for each identified CRAT. The in-text results were described by CRAT and summarised the key results from each quality assessment step. The narrative synthesis of the results was guided by the

Cochrane Consumers and Communication Review Group's document on data synthesis and analysis[17] and was aligned with the research question and focused on the overall strength of evidence for each CRAT.

3 | RESULTS

The search strategy resulted in the identification of 432 unique papers, of which 167 were selected for full-text review based on their title and abstract. On completion of the full-text screening, 10 (of the 167) papers, assessing eight different CRATs, were included in this review (Figure 1). The general characteristics of included studies and their respective CRATs are presented in Table 2.

To align with the research question, the main results are reported below by CRAT. Table 3 presents the methodological quality of each study by measurement property and CRAT. Table 4 presents the quality of each measurement property by CRAT. Table 5 presents the overall strength of evidence for each CRAT by measurement property.

The University of North Carolina Caries Risk Assessment studies (UNCCRA) were conducted to improve methods to identify children prospectively at high risk to dental caries.[16,18,19] These CRA models have around fifteen items and the assessor determined the caries risk level.

The methodological quality of these studies was rated "Poor" for reliability and measurement error (Table 3). For quality of measurement properties, only content validity achieved a positive rating (Table 4). There was strong positive evidence for content validity and strong negative evidence for criterion validity and responsiveness in the overall strength of evidence assessment (Table 5).

University of Michigan paediatric dental clinic CRA sheet (UMCRA) was developed for initial and recall patient encounters in the Young Patient and Prevention Clinic (YPPC) at the University of Michigan.[20] The CRA sheet consists of 14 items, which are used by the assessor to determine the caries risk level.

There were no methodologically sound studies found for any of the measurement properties (Table 3). No studies achieved a positive rating for quality of measurement properties (Table 4). For the overall strength of evidence by measurement property, the evidence was mostly unknown due to poor methodological quality (Table 5).

Cariogram is a computer program which graphically represents a caries risk profile for an individual and is expressed as "chance (%) to avoid caries".[5] This tool consists of nine items, and the risk level is program generated based on a predetermined algorithm.[21]

The methodological quality of studies was rated "good" for criterion validity and responsiveness (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). For the overall strength of evidence for the CRAT, by measurement property, there was strong negative evidence for Cariogram's criterion validity and responsiveness (Table 5).

Similar to the Cariogram, the National University of Singapore caries risk assessment tool (NUS-CRAT) classifies children's caries risk into five risk levels and is expressed as "chance (%) to avoid caries." This tool has eleven items (6 items in the community-screening version), and the risk level is program-generated based on a pre-determined algorithm.[22,23]

The methodological quality of studies was rated as "Fair" or higher for all measurement properties with content validity achieving an "Excellent" rating (Table 3). For quality of measurement properties, positive ratings were achieved by responsiveness and content and criterion validity (Table 4). There was strong positive evidence for NUS-CRAT's content, criterion validity, and responsiveness in the overall strength of evidence assessment (Table 5).

The Dundee Caries Risk Assessment Model (DCRAM) is structured in a decision tree format and includes 2-5 items (depending on the particular DCRAM to be used) to identify children at high risk for dental caries.[24]

The methodological quality of studies was mostly rated as "Fair" or higher, except for reliability and measurement error which were rated as "Poor" (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). There was strong positive evidence for DCRAM's content in the overall strength of evidence assessment (Table 5).

The American Academy of paediatric Dentistry Caries Assessment Tool (AAPD CAT) designed for use in 0- to 5-year-olds includes 14 items to classify children as either high, medium, or low risk for dental caries.[4,22,25]

The methodological quality of studies was rated as "Poor" for content validity (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). There was strong negative evidence for AAPD CAT's criterion validity and responsiveness in the overall strength of evidence assessment (Table 5).

The Caries Management by Risk Assessment (CAMBRA) is an approach that couples risk assessment with tailored preventive care and risk monitoring. The CRAT form for 0- to 5-year-olds includes 20 items to classify children as either high, moderate, or low risk for dental caries.[22,26,27] The assessor determines the caries risk level using these twenty items.

The methodological quality of studies was rated "Poor" for reliability and measurement error (Table 3). For quality of measurement properties, a positive rating was achieved for content validity (Table 4). There was strong positive evidence for CAMBRA's content validity in the overall strength of evidence assessment (Table 5).

TABLE 2 Characteristics of included studies

Author, publication year	Study year(s)	Study design	Population and setting	Age[a]	N	CP (%)	Predicted Caries definition	Country	CRAT(s)
Disney et al, 1992[19]	1986-1989	Longitudinal	Grade 1 in the geographic areas surrounding 1. Aiken, SC and 2. Portland, Maine	6	4000	72[b] 40[c]	dmfs—caries into dentine	USA	University of North Carolina Caries Risk Assessment studies (UNCCRA)
Beck et al, 1992[18]	1986-1989	Longitudinal	Grade 1 children in the geographic areas surrounding 1. Aiken, SC and 2. Portland, Maine	6	4000	72[b] 40[c]	dmfs—caries into dentine	USA	University of North Carolina Caries Risk Assessment studies (UNCCRA)
Wandera et al, 2000[20]	1994-1997	Longitudinal	Children attending the Young Patient and Prevention paediatric dental Clinic at the University of Michigan	4	140	41	Not reported Most likely into dentine	USA	University of Michigan paediatric dental clinic tool (UMCRA)
Holgerson et al, 2009[21]	2000-2007	Longitudinal	Children born between January 2000 and March 2001, attending the Public Dental Clinic in Lycksele, a small municipality in northern Sweden	2	103	71	Enamel and dentine lesions	Sweden	Cariogram
Gao et al, 2010[23]	NR	Longitudinal	Children in government kindergartens in Singapore	3-6	1782	40	dmft—caries into dentine	Singapore	National University of Singapore caries risk assessment tool (NUS-CRA)
MacRitchie et al, 2012[24]	1993-1997	Longitudinal	The cohort comprised all children born and resident in Dundee between 1 April 1993 and 31 March 1994	0-4	1890	49	Enamel and dentine lesions	Scotland	Dundee Caries Risk Assessment Model (DCRAM)
Yoon et al, 2012[25]	2006-2009	Longitudinal	Residents served by the University of paediatric dental clinic, with fluoridated water in the northern part of the New York City borough Manhattan, which has predominantly low-income and Hispanic populations	≤3	471	49	Enamel and dentine lesions	USA	American Academy of paediatric Dentistry caries assessment tool (AAPD CAT)
Gao et al, 2013[22]	NR	Longitudinal	Children at kindergartens in Hong Kong	3	544	35	dmft—caries into dentine	Hong Kong	NUS-CRA Cariogram CAMBRA AAPD CAT
Chaffee et al, 2016[26]	2009-2015	Observational retrospective, longitudinal analysis	Patients at the University of San Francisco paediatric Dentistry clinic	0-6	1315	67	Enamel and dentine lesions	USA	Caries Management by Risk Assessment (CAMBRA)

(Continues)

TABLE 2 (Continued)

Author, publication year	Study year(s)	Study design	Population and setting	Age[a]	N	CP (%)	Predicted Caries definition	Country	CRAT(s)
Custodio-Lumsden et al, 2016[28]	2012/2013	Cross-sectional	Caregiver/child dyads presenting to the Columbia University College of Dental Medicine paediatric Dental Clinic	2-6	108	35	Caries into dentine—frank cavities	USA	MySmileBuddy (MSB)

CP, caries prevalence among population in which tool tested.
[a]Age in years.
[b]Caries prevalence for Aiken, South Carolina.
[c]Caries prevalence for the Portland, Maine.

MySmileBuddy (MSB) is a risk assessment, caregiver education, and ECC management tool. MSB consists of a series of five assessment modules containing questions and brief educational units, one each on, feeding practices, attitudes and beliefs, fluoride, and family history. Risk levels are program-generated and classify children as low (score: 1-3), medium (score: 4-6), and high (score: 7-10) for caries.[28,29]

The methodological quality of studies was rated as "Fair" or lower for measurement properties (Table 3). For quality of measurement properties, none achieved a positive rating (Table 4). The overall strength of evidence for the CRAT, by measurement property, was unknown due to the tool being early in its development stages (Table 5).

3.1 | Narrative synthesis

Common across all tools was a lack of information to determine the levels of evidence for the key measurement properties of reliability (including measurement error) and construct validity. In terms of content validity, UMCRA, Cariogram, AAPD CAT, and MSB achieved "unknown" ratings. The main reason for this rating was that the risk assessment items in those CRATs were not demonstrated to be relevant to the target population in which the tools were used. Studies on tools that were assessed as having strong evidence for content validity identified the relevant risk factors for caries in the population being studied, before developing and testing their respective CRATs. For criterion validity and responsiveness, only the NUS-CRAT achieved a strong level of evidence. This was driven mainly by the fact that only the NUS-CRAT achieved a combined sensitivity and specificity score of over 160 and individual sensitivity and specificity scores of at least 75 and 85, respectively (Appendix 4). These are the recommended minimum scores for a tool to predict future caries.[16] Overall, based on reported information, the NUS-CRAT attained a higher quality rating than other CRATs studied by achieving strong evidence ratings in three out of the six measurement properties studied.

4 | DISCUSSION

The review identified the CRATs with published peer-reviewed evidence and the important measurement properties to consider when selecting a health measurement instrument. The review findings showed however that the information reported in the literature, for each CRAT, was insufficient to make a full quality assessment. This finding was mostly due to poor methodological quality and reporting of studies. For example, whereas criterion validity (sensitivity and specificity) is reported in most studies, properties such as reliability in tool application,

TABLE 3 Methodological quality of each study by measurement property and caries risk assessment tool

CRAT and study(ies)	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsiveness
UNCCRA						
Disney et al, 1992[19]	Poor	Poor	Excellent	Fair	Excellent	Excellent
Beck et al, 1992[18]	Poor	Poor	Excellent	Fair	Excellent	Excellent
UMCRA						
Wandera et al, 2000[20]	Poor	Poor	Poor	Fair	Poor	Poor
Cariogram						
Holgerson et al, 2009[21]	na	na	Poor	Fair	Good	Good
Gao et al, 2010[23]	na	na	Poor	Fair	Good	Good
Gao et al, 2013[22]	na	na	Poor	Fair	Good	Good
DCRAM						
MacRitchie et al, 2012[24]	Poor	Poor	Excellent	Fair	Good	Good
AAPD CAT						
Yoon et al, 2012[25]	Poor	Poor	Poor	Fair	Good	Good
Gao et al, 2013[22]	Poor	Poor	Poor	Fair	Good	Good
NUS-CRA						
Gao et al, 2010[23]	na	na	Excellent	Fair	Good	Good
Gao et al, 2013[22]	na	na	Excellent	Fair	Good	Good
CAMBRA						
Gao et al, 2013[22]	Poor	Poor	Poor	Fair	Good	Good
Chaffee et al, 2016[26]	Poor	Poor	Excellent	Fair	Excellent	Excellent
MSB						
Custodio-Lumsden et al, 2016[28]	na	na	Poor	Fair	Poor	na

na, not applicable.

measurement error, construct validity (including discriminatory validity), and content validity have been, in most cases, poorly studied. This review is consistent with and augments the findings of a recent review on the evidence for current caries risk assessment tools/systems.[9] Whereas the review by Tellez et al[9] focused only on criterion validity, this review included other measurement properties relevant to CRATs and in addition focused on a specific age, children ages 6 years and less.

The rationale to focus on very young children was in recognition that factors contributing to caries risk in the early years are unique to this phase of life and disappear as the child gets older and transitions to school. For example, important influences on caries in young children include the following: parenting behaviours, knowledge, beliefs and attitudes; infant feeding practices; maternal circumstances and oral health; and infant-related oral health behaviours.[10]

The authors of this review identified several key issues with studies on current CRATs for young children. First, reliability (which includes measurement error), a key measurement property particularly for reasoning-based user-determined caries risk levels is not reported in any study included in this review. For CRATs where the risk levels are algorithm-based program-generated, such as Cariogram and NUS-CRAT, it is assumed that the same risk level will be generated when the same information is entered for each item by different users. However, there could be inconsistencies in data input for these algorithm-based CRATs that could affect risk level assignment. Hence, until reliability estimates are provided, the evidence on reliability remains unknown.

The second issue identified was the "unknown" rating for evidence on content validity for several tools, mainly due to the tool items not being checked for relevance among the population in which the tool was tested. The UNCCRA studies highlighted this issue where caries prevalence rates,

TABLE 4 Quality of the measurement properties by caries risk assessment tool and study

Author, year	CRAT(s)	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsiveness
Disney et al, 1992[19]	UNCCRA	?	?	+	?	–	–
Beck et al, 1992[18]	UNCCRA	?	?	+	?	–	–
Wandera et al, 2000[20]	UMCRA	?	?	–	?	–	–
Holgerson et al, 2009[21]	Cariogram	?	?	–	?	–	–
Gao et al, 2010[23]	NUS-CRA^S	?	?	+	?	+	+
	NUS-CRA^C	?	?	+	?	+	+
	Cariogram	?	?	–	?	–	–
MacRitchie et al, 2012[24]	DCRAM1	?	?	+	?	–	–
	DCRAM2	?	?	+	?	–	–
	DCRAM3	?	?	+	?	–	–
	DCRAM4	?	?	+	?	–	–
Yoon et al, 2012[a 25]	AAPD CAT	?	?	–	?	–	–
Gao et al, 2013[22]	NUS-CRA^S	?	?	+	?	–	–
	NUS-CRA^C	?	?	+	?	+	+
	Cariogram^S	?	?	–	?	–	–
	Cariogram^C	?	?	–	?	–	–
	CAMBRA^S	?	?	–	?	–	–
	CAMBRA^C	?	?	–	?	–	–
	AAPD CAT^S	?	?	–	?	–	–
	AAPD CAT^C	?	?	–	?	–	–
Chaffee et al, 2016[26]	CAMBRA	?	?	+	?	–	–
Custodio-Lumsden et al, 2016[28]	MSB	?	?	–	?	?	?

DCRAM1, predicting d1mft >0 (All caries lesions and predicting any caries); DCRAM2, predicting d3mft >0 (Dentine caries lesions and predicting any caries); DCRAM3, predicting d1mft ≥3 (All caries lesions and predicting more than two caries lesions); DCRAM4, predicting d3mft ≥3 (Dentine caries lesions and predicting more than two caries lesions); NUS-CRA C, comprehensive, includes all risk factors; NUS-CRA S, screening tool, reduced # risk factors assessed—excludes microbiology and saliva analysis; + = positive rating, ? = indeterminate (Unknown) rating, – = negative rating.

[a]Only overall AAPD CAT used

geographic location, and age of the population determined the factors to include in the risk prediction models and eventually the items in the CRAT. This is a very important consideration for clinical practice as it raises the issue of adopting tools without testing them for relevance to the population being served. This could also be a major issue for caries risk assessments in young children as the caries-related factors can differ from those among older children.[10]

The third issue identified was the lack of reporting on discriminatory validity for all CRAT tools examined, resulting in an "unknown" rating for this property. Discriminatory validity, a component of construct validity, is the relative ability of a CRAT to differentiate among risk levels. Without reporting on discriminatory validity, the risk level categorisation may be arbitrary and based more on a theoretical intellectual process rather than evidence.

The evidence to inform the selection of the Cariogram mostly achieved an "unknown" rating and was due to it mostly being used as a comparator for other tools in the included studies, rather than specifically assessing its usefulness in the testing population. CAMBRA is a well-used tool because it also provides clear management guidelines for each risk category.[27] CAMBRA is mostly a clinician reasoning-based CRAT for which reliability in the application of the tool is important to ensure consistency across clinicians in risk assignment as well as the associated management strategies.

TABLE 5 Overall strength of evidence for each caries risk assessment tool, by measurement property

CRAT	Reliability	Measurement error	Content validity	Construct validity	Criterion validity	Responsiveness
UNCCRA	?	?	+++	?	−−−	−−−
UMCRA	?	?	?	?	?	?
Cariogram	?	?	?	?	−−−	−−−
NUS-CRA	?	?	+++	?	+++	+++
DCRAM	?	?	+++	?	−−	−−
CAMBRA	?	?	+++	?	−−−	−−−
AAPD CAT	?	?	?	?	−−−	−−−
MSB	?	?	?	?	?	?

+++ or −−− strong evidence positive/negative result, ++ or −− moderate evidence positive/negative result, + or − limited evidence positive/negative result, ± conflicting evidence, ? unknown, due to poor methodological quality.

The evidence for this measurement property of CAMBRA is at present unknown. For criterion validity, CAMBRA was assessed as having strong evidence for a negative result which was mainly due to it not achieving the recommended combined sensitivity and specificity score of 160. Studies on CAMBRA however consistently reported high sensitivity scores of 0.84 and above.[22,26] The AAPD CAT developed by the American Association for paediatric Dentistry,[4] as with other CRATs, has an unknown level of evidence for its content which could be due to the limited number of publications that describe the process of identifying and including the items that constitute this tool. DCRAM is a CRAT with the least number of items (2-5 items depending on model) that has strong evidence to support its content and uses a decision tree format to arrive at a risk level.[24] The tool is relatively new compared with other tools and has had limited testing beyond the author group that developed the tool. The NUS-CRAT,[22,23] was recently developed for young children, had the strongest evidence to support its selection. The authors of studies on NUS-CRAT attribute its positive performance to two main factors. First, being algorithm driven, CRAT allows for a mathematical synthesis of risk factors and indicators and defines their relative contribution to risk using pre-determined weights. Second, could be the inclusion of age-specific risk factors such as infant feeding practices in the assessment process (Appendix 5). MySmileBuddy is the most recently developed CRAT that also provides oral health education information and caries management protocols.[28] The tool is in its very early stages of development and hence received an unknown rating.

Some limitations of the COSMIN checklist should be noted. Although it is comprehensive, it is complex and requires the user to be very familiar with the measurement properties and rating criteria. Certain measurement properties required more specific author generated definitions for clarity and standardisation in the rating process. For example, a question on content validity is: "Was there an assessment of whether all items refer to relevant aspects of the construct to be measured?" In this case, a clear definition of the construct and the appropriate assessment was required to ensure consistency in application of the rating criteria. In relation to construct validity, the quality criteria requirement is for studies to state a priori hypothesis. None of the studies however in this review stated a hypothesis and as such received a lower quality rating. As with most quality assessment tools, the rating criteria definitions may need to be revised to better fit an area of research.

5 | CONCLUSION

This systematic review showed that the evidence to inform the selection of current CRATs for children is yet to be established. Overall, the NUS-CRAT studies reported the most information to inform the assessment of its measurement properties, and as a result, this tool attained a higher quality rating than other CRATs studied. The use of CRATs in caries management is very important and highly recommended. Methodologically sound studies are urgently needed to provide the evidence for the measurement properties of CRATs for young children.

ACKNOWLEDGMENTS

The authors acknowledge the Centre for Health Equity, Melbourne School for Population and Global Health, The University of Melbourne and La Trobe Rural Health School, La Trobe University for open access funding support.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

BC conceived the idea; BC and RA designed the methods; BC involved in literature search; BC, RA, and LC involved in paper screening, data extraction, and quality assessments; BC, RA, HC, LC, and LG and MG wrote the paper.

ORCID

Bradley Christian https://orcid.org/0000-0001-5072-3512

REFERENCES

1. Fisher-Owens SA, Gansky SA, Platt LJ, et al. Influences on children's oral health: a conceptual model. *Pediatrics*. 2007;120:e510-e520.
2. Mejare I, Axelsson S, Dahlen G, et al. Caries risk assessment. A systematic review. *Acta Odontol Scand*. 2014;72:81-91.
3. Senneby A, Mejare I, Sahlin NE, Svensater G, Rohlin M. Diagnostic accuracy of different caries risk assessment methods. A systematic review. *J Dent*. 2015;43:1385-1393.
4. AAPD. Guideline on caries-risk assessment and management for infants, children, and adolescents. *Pediatr Dent*. 2014;35:157-164.
5. Bratthall D, Petersson GH. Cariogram – a multifactorial risk assessment model for a multifactorial disease. *Community Dent Oral Epidemiol*. 2005;33:256-264.
6. Featherstone JD, Adair SM, Anderson MH, et al. Caries management by risk assessment: consensus statement, April 2002. *J Calif Dent Assoc*. 2003;31:257-269.
7. Twetman S, Fontana M. Patient caries risk assessment. *Monogr Oral Sci*. 2009;21:91-101.
8. Mokkink L, Terwee C, Patrick D, et al. *Cosmin Checklist Manual*. Amsterdam: EMGO Institute for Health and Care Research, VU University Medical Centre; 2012.
9. Tellez M, Gomez J, Pretty I, Ellwood R, Ismail AI. Evidence on existing caries risk assessment systems: are they predictive of future caries? *Community Dent Oral Epidemiol*. 2013;41:67-78.
10. Leong PM, Gussy MG, Barrow SY, de Silva-Sanigorski A, Waters E. A systematic review of risk factors during first year of life for early childhood caries. *Int J Paediatr Dent*. 2013;23:235-250.
11. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Int J Surg*. 2010;8:336-341.
12. Whiting PF, Rutjes AW, Westwood ME, et al. Quadas-2: a revised tool for the quality assessment of diagnostic accuracy studies. *Ann Intern Med*. 2011;155:529-536.
13. Guyatt GH, Oxman AD, Vist GE, et al. Grade: an emerging consensus on rating quality of evidence and strength of recommendations. *BMJ*. 2008;336:924-926.
14. Terwee CB, Mokkink LB, Knol DL, Ostelo RW, Bouter LM, de Vet HC. Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the cosmin checklist. *Qual Life Res*. 2012;21:651-657.
15. Terwee C, de Vet HC, Prinsen CA, Mokkink L. *Protocol for Systematic Reviews of Measurement Properties*. Amsterdam: COSMIN; 2011:1-15.
16. Stamm JW, Disney JA, Graves RC, Bohannan HM, Abernathy JR. The university of north carolina caries risk assessment study. I: Rationale and content. *J Public Health Dent*. 1988;48:225-232.
17. Ryan R. Cochrane Consumers and Communication Review Group. *Cochrane Consumers and Communication Review Group: data synthesis and analysis*. http://cccrg.cochrane.org. Accessed October 17, 2017.
18. Beck JD, Weintraub JA, Disney JA, et al. University of North Carolina caries risk assessment study: comparisons of high risk prediction, any risk prediction, and any risk etiologic models. *Community Dent Oral Epidemiol*. 1992;20:313-321.
19. Disney JA, Graves RC, Stamm JW, Bohannan HM, Abernathy JR, Zack DD. The University of North Carolina caries risk assessment study: further developments in caries risk prediction. *Community Dent Oral Epidemiol*. 1992;20:64-75.
20. Wandera A, Bhakta S, Barker T. Caries prediction and indicators using a pediatric risk assessment teaching tool. *J Dent Child*. 2000;67:408-412.
21. Holgerson PL, Twetman S, Stecksen-Blicks C. Validation of an age-modified caries risk assessment program (cariogram) in preschool children. *Acta Odontol Scand*. 2009;67:106-112.
22. Gao X, Di Wu I, Lo EC, Chu CH, Hsu CY, Wong MC. Validity of caries risk assessment programmes in preschool children. *J Dent*. 2013;41:787-795.
23. Gao XL, Hsu CY, Xu Y, Hwarng HB, Loh T, Koh D. Building caries risk assessment models for children. *J Dent Res*. 2010;89:637-643.
24. MacRitchie H, Longbottom C, Robertson M, et al. Development of the Dundee Caries Risk Assessment Model (DCRAM) – risk model development using a novel application of chaid analysis. *Community Dent Oral Epidemiol*. 2012;40:37-45.
25. Yoon RK, Smaldone AM, Edelstein BL. Early childhood caries screening tools: a comparison of four approaches. *J Am Dent Assoc*. 2012;143:756-763.
26. Chaffee BW, Featherstone JD, Gansky SA, Cheng J, Zhan L. Caries risk assessment item importance: risk designation and caries status in children under age 6. *JDR Clin Trans Res*. 2016;1:131-142.
27. Ramos-Gomez FJ, Crall J, Gansky SA, Slayton RL, Featherstone JD. Caries risk assessment appropriate for the age 1 visit (infants and toddlers). *J Calif Dent Assoc*. 2007;35:687-702.
28. Custodio-Lumsden CL, Wolf RL, Contento IR, et al. Validation of an early childhood caries risk assessment tool in a low-income hispanic population. *J Public Health Dent*. 2016;76:136-142.
29. Levine J, Wolf RL, Chinn C, Edelstein BL. My smile buddy: an iPad-based interactive program to assess dietary risk for early childhood caries. *J Acad Nutr Diet*. 2012;112:1539-1542.
30. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med* 2009;6(6):e1000097.
31. Terwee CB, Bot SD, de BM, et al. Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007;60:34-42.
32. Domejean S, Banerjee A, Featherstone JB. Caries risk/susceptibility assessment: its value in minimum intervention oral healthcare. *Br Dent J* 2017;223(3):191-197.

SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

How to cite this article: Christian B, Armstrong R, Calache H, Carpenter L, Gibbs L, Gussy M. A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children. *Int J Paediatr Dent*. 2018;00:1–11. https://doi.org/10.1111/ipd.12446

Appendix 4. Research Strand 2, published paper: Calibration Study

Citation: *Christian B, Amezdroz E, Calache H, Gussy M, Sore R, Waters E. Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical. Community dental health. 2017;34(4):248-53.*

Community Dental Health (2017) **34**, 248–253
Received 2 December 2016; Accepted 25 April 2017

© BASCD 2017
doi:10.1922/CDH_4102Christian06

Examiner calibration in caries detection for populations and settings where *in vivo* calibration is not practical

Bradley Christian[1,3], Emily Amezdroz[1], Hanny Calache[2], Mark Gussy[3], Rachel Sore[4] and Elizabeth Waters[1]

[1]*Evidence and Child Health, The University of Melbourne;* [2]*Centre for Population Health Research, Deakin University;* [3]*Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University;* [4]*Statistical Consulting Centre, The University of Melbourne*

Aim: To study examiner calibration methods for populations and settings where clinical (*in vivo*) calibration is not practical. ***Methods:*** Study design was cross-sectional and fully-crossed. The units of analysis were 880 tooth surfaces, from ten children ages 3 to 4 years. The study had three data components: (1) Examiner training and calibration using the *International Caries Detection and Assessment System* (ICDAS) e-Learning programme (2) *In vivo* community-based visual examination and (3) Intra-oral digital photographs of the same tooth surfaces from the *in vivo* visual examination. Kappa and weighted kappa scores were used to study reliability estimates. Systematic differences in caries assessments were determined using the Stuart Maxwell test. Data were analysed using STATA 13.1 and SAS 9.2. ***Results:*** Weighted kappa scores for the *in vivo* component ranged from 0.50 to 0.66 and from 0.64-0.74, for inter- and intra-examiner reliability, respectively. Caries lesions detected *in vivo* were also detected on photographs, albeit with increased severity when using photographs. For example, of 46 tooth surfaces assessed as being sound in the *in vivo* examination, 22 (48%) of these were assessed as having caries when photographs were used as the diagnostic method. ***Conclusions:*** From this research it appears that good quality photographs alone may be used for training and calibration among challenging populations or settings without adversely affecting data quality.

Key words: *dental caries, calibration, photographs, epidemiology, caries detection, oral diagnosis, caries assessment*

Introduction

Data quality in oral health research ensures valid estimates of disease are obtained (Nelson *et al.*, 2011). Validity is the ability of a test to distinguish between those with and without disease, and is commonly measured using sensitivity, specificity and positive predictive and negative predictive values. Reliability is the ability of the test to produce the same results when repeated (Chattopadhyay, 2010). Standardization in caries assessments can be achieved by training and calibrating examiners. Training involves examiners learning examination protocols and calibration is a process that allows measurements to be compared to reference standards (validity), and repeated scoring by the same and different examiners (intra-and inter-examiner reliability).

It is generally recommended that *in vivo* training and calibration exercises be conducted following photograph slide-based training, to give trainee examiners the opportunity to apply their training to the *in vivo* setting, where the clarity of a visual examination may not be as good as a photograph of a caries lesion. This step also allows examiners to study, discuss and evaluate their scoring of the *in vivo* presentation of caries lesions before data collection. *In vivo* calibration entails several examinations on the same person by different examiners, making it a rigorous process requiring maximum co-operation from study participants. Hence, while *in vivo* training and calibration is good practice, it may not always be possible, for reasons such as timing of data collection, lack of resources, specific population limitations (very young children, people with a disability and the elderly). For example, calibration exercises among pre-school children can be demanding on the child, parents/care-givers and examiners. Children, particularly those under five years, are more temperamental, unpredictable and reactive to their environment, thus making examinations more difficult (Aminabdi *et al.*, 2011). *In-vitro* methods for training and calibration could therefore be useful, but require assessment before they are used.

Several studies have investigated the use of intra-oral photographs as a caries diagnostic tool, but fewer have assessed their validity (Boye *et al.*, 2012, Elfrink *et al.*, 2009). A series of studies comparing intra-oral photographs with visual *in vivo* clinical assessments for caries detection demonstrated good comparability between the two methods (Boye *et al.*, 2013a, Boye *et al.*, 2013b, Boye *et al.*, 2013c). However, the unit of analysis in these studies was overall caries experience score (dmft) per study participant, which describes gross error rather than caries diagnosis at the tooth or surface level. In addition, most research has focused on caries diagnosis at the level of cavitation (dentine lesions) which does not recognize early non-cavitated stages of the disease process. More precise and sensitive diagnosis is particularly important for prevention and health promotion (Bader *et al.*, 2002). There has also been limited research in pre-school children, and to our knowledge no comparative studies in a community-based setting (Bader *et al.*, 2002, Gomez *et al.*, 2013).

Correspondence to: Bradley Christian, Evidence and Child Health, Melbourne School of Population and Global Health, The University of Melbourne, Level 5, 207 Bouverie Street, Victoria 3010, AustraliaTel: +61 3 90359926, Email: B.Christian@latrobe.edu.au

The purpose of this research was to study examiner calibration methods for populations and settings where clinical (*in vivo*) calibration is not practical. The objectives were to:

1. Determine whether photographs alone can be used in the training and calibration of examiners without adversely affecting data quality? This was achieved by assessing inter-and intra-examiner reliability in diagnosing the severity of caries lesions at the surface level, using: (a) Photographs of caries lesions sourced from the ICDAS e-Learning training programme and (b) *In vivo* community-based visual examination of children's teeth.
2. Assess systematic bias in *in vivo* caries diagnosis.
3. Compare caries diagnosis using intra-oral photographs of selected tooth surfaces with their corresponding *in vivo* community examination scores; to assess systematic differences in caries diagnosis.

Materials and Methods

Ethics approval for this study was provided by the Human Research Ethics Committee at the University of Melbourne (Ethics ID 1442648).

Study design and setting

This cross-sectional study used a fully-crossed design with the same examiners examining all study participants. This approach allowed systematic bias to be assessed and controlled for in reliability estimates (Hallgren, 2012).

Study participants were a sub-sample of 3- to 4-year-olds from the VicGeneration (VicGen) cohort of children residing in Melbourne, Australia. This age range was chosen for the full complement of primary teeth. The VicGen oral health birth cohort study began in 2008, to gain an understanding of the multifactorial nature of early childhood caries development (de Silva-Sanigorski *et al.*, 2010).

Sample size and participants

Based on estimates from the VicGen cohort, and with the intention to set the minimum acceptable weighted kappa level as 0.61, an estimated sample size of 50 teeth (250 surfaces) was required to detect a weighted kappa score of 0.87 (95% CI: 0.64, 1.00). The sample size calculation was performed by manipulating the estimated distribution of ratings until a sufficiently narrow 95% confidence interval was obtained. For this study, 880 tooth surfaces from 10 children were analysed. Ten children were purposively selected (based on caries activity) and invited to participate. This selection process ensured presence of caries lesions to reflect a range of ICDAS caries codes.

Data collection

Data collection involved three components: (1) Examiner training and calibration using the ICDAS e-Learning programme (2) *In vivo* community-based visual examination and (3) Intra-oral digital photographs of the same tooth surfaces from the *in vivo* visual examination.

Components (1) and (2) relate to objective one, which was to determine whether sufficient reliability was achieved in the *in vivo* assessments, when training and calibration was conducted using photographs from the ICDAS e-Learning programme (Topping *et al.*, 2008). Components (2) and (3), relate to objective three (Tables 5 & 6), where the *in vivo* assessments for selected surfaces were compared with their corresponding intra-oral photograph scores to determine whether caries lesions detected *in vivo* would also be detected on photographs to determine their validity.

Three dental examiners were used. Caries was diagnosed and scored using the International Caries Detection and Assessment System (ICDAS) (Ismail, 2007). ICDAS codes (Table 1) were used due to distinguish differing severity of lesions as compared with the WHO and BASCD methods, which only record dentinal lesions (cavities) (Altarakemah *et al.*, 2013, Braga *et al.*, 2009, Pine *et al.*, 1997). Teeth were not air-dried as this was considered impractical in the community setting. This lack of air drying prevented the use of ICDAS caries code 1 on smooth surfaces. The *in vivo* dental examinations were exclusively visual without the use of a dental probe.

Table 1. ICDAS Codes

Treatment Codes	Caries Codes
0 Sound	0 Sound surface
1 Sealant, partial	1 First visual change in enamel
2 Sealant, full	2 Distinct visual change in enamel
3 Tooth coloured restoration	3 Enamel breakdown: no dentine visible
4 Amalgam restoration	4 Dentinal shadow (not cavitated into dentine)
5 Stainless steel crown	5 Distinct cavity with visible dentine
6 Porcelain or gold or PFM crown or veneer	6 Extensive distinct cavity with visible dentine
7 Lost or broken restoration	
8 Temporary restoration	

ICDAS e-Learning programme training and calibration

The 90 Minute e-Learning programme by the ICDAS Foundation was used to train examiners in the application of ICDAS (Topping *et al.*, 2008). An experienced clinician and researcher (HC), facilitated the session, followed by a discussion of the scoring criteria, until agreement on the reference standard ICDAS code was achieved. For the calibration, examiners scored four sets of eight photographs each (32 photographs in total) of the various ICDAS codes, sourced from the e-Learning programme. Examiners were blinded to each other's scoring. Scoring was repeated to measure intra-examiner reliability.

In vivo community-based visual dental assessments

Each participant was visited at home twice, one week apart. At each visit the participant was examined by each of the three examiners who were blinded to each other's assessment. Five minute breaks were provided between examinations for each child. In total each child had six assessments.

Children were examined lying down on a flat surface with the examiner situated behind them. The examination involved standardised light sources (Henry Schein MirrorLites) and infection control protocol. A mouth mirror was used to view the teeth and gauze used to remove debris where required. Each quadrant was examined in a standardised manner. A trained research assistant recorded the caries scores onto paper forms which were then double-entered into an electronic database and checked for data entry consistency.

Intra-oral photographs

Intra-oral digital photographs of all tooth surfaces of participating children were taken to compare with the corresponding *in vivo* examinations. These photographs were taken at the first or second visit depending on the participant's convenience. All the photographs were taken by the same examiner, experienced in the use of the intra-oral camera and trained in the examination protocol. The Sopro Acteon 717 intra-oral camera had its own light source and the following settings: intra-oral mode, colour images and output format as JPG. The protocol for taking intra-oral photographs was similar to the *in vivo* examination. The images were captured digitally and stored directly to an electronic folder. The data were backed up onto an external hard drive at the end of each day.

For the analysis, 71 photographs were chosen based on clarity and to reflect a range of ICDAS codes. The photographs were scored by the same three examiners who conducted the *in vivo* examinations. A standardised environment was used to score the photographs, with the photographs projected onto a screen approximately 3 meters from the examiners. The room was dimmed during the session and each photograph was viewed for approximately 20 seconds. One examiner selected and collated the photographs. To avoid bias this examiner's scores were not used.

Data analysis

The unit of analysis was the tooth surface. A child with a complete primary dentition of 20 teeth will have 88 tooth surfaces in total. Five tooth surfaces for each molar and four for incisors and canines. Though both ICDAS codes were recorded, the focus for this analysis were the ICDAS caries codes.

Kappa and weighted kappa scores were used to determine inter- and intra-examiner reliability for tooth surface level scoring of caries lesions. Weighted kappa is recommended for determining examiner reliability for ordinal data such as ICDAS caries codes (0 (sound surface) – 6 (extensive decay)). This statistic incorporates the factor of agreement by chance alone and also weights proportional agreement (Fleiss, 1973, Machure and Willett, 1987)1987. The 'kapci' command in STATA was used to generate kappa scores and 95% confidence intervals (95% CI), with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95% CI.

Clustering effects (surfaces within teeth and teeth within individuals) were explored and found to be minimal and hence, estimates did not require adjustment for clustering (Litaker *et al.*, 2013, Masood *et al.*, 2015).

To assess systematic bias in scoring caries severity, the Stuart-Maxwell test for marginal homogeneity was conducted. This tests for the marginal homogeneity across all ICDAS categories simultaneously and determined if examiners were systematically scoring differently from other examiners or within themselves.

Data were analysed using STATA 13.1 and SAS 9.2.

Results

Reliability estimates for the two caries diagnostic methods are provided in Table 2. Using the e-Learning photographs, Kappa scores for inter-examiner reliability ranged from 0.56 to 0.70 (moderate to good). In the *in vivo* examination, using ICDAS caries codes only, weighted Kappa scores for inter-examiner reliability ranged from 0.50 to 0.64 (moderate to good).

Systematic differences in scoring caries lesions *in vivo* are summarised in Tables 3 and 4. Testing for marginal homogeneity between the three examiners showed that in some instances significant ($p<0.05$) differences were observed, both between and within examiners. From Table 3, it appears that examiner 2 was scoring differently to examiners 1 and 3, as determined by the non-significant p-value of 0.330 for examiner 1 against 3. This difference is shown in the marginal distribution of ICDAS scores in Table 4.

Tables 5 and 6 compare caries diagnoses of the same tooth surfaces using the *in vivo* examination and intra-oral photographs of the same teeth. To assess systematic differences, the scores for the two methods from one examiner were cross tabulated (Table 5). More caries lesions were diagnosed from photographs (Stuart-Maxwell p=0.003). For example, of the 46 tooth surfaces diagnosed as being sound (ICDAS code 00) *in vivo*, only 24 (52%) were diagnosed as sound using photographs. The remaining 22 experienced caries, off which 16 were non-cavitated caries lesions (ICDAS code 02). Seventeen of these 22 diagnostic differences occurred on posterior teeth.

Table 2. Estimates (95% CI) for inter- (at time 1) and intra-examiner reliability

Examiners	1	2	3
1	0.96 (0.88-1.00)[1] 0.60 (0.54-0.66)[2] 0.74 (0.65-0.81)[3]	0.60 (0.41-0.80)[1] 0.55 (0.48-0.61)[2] 0.52 0.40-0.63)[3]	0.70 (0.54-0.89)[1] 0.54 (0.47-0.60)[2] 0.64 (0.51-0.74)[3]
2		1.00[1] 0.65 (0.58-0.71)[2] 0.64 (0.51-0.74)[3]	0.56 (0.35-0.76)[1] 0.54 (0.48-0.61)[2] 0.50 (0.37-0.63)[3]
3			0.96 (0.88-1.00)[1] 0.56 (0.49-0.63)[2] 0.66 0.52-0.77)[3]

[1]Kappa scores from the ICDAS e-Learning training photographs (n=32 photographs)
[2]Kappa scores (all ICDAS codes) *in vivo* community dental examinations (n=880 tooth surfaces)
[3]Weighted Kappa scores (ICDAS caries codes only) *in vivo* community dental examinations (n=880 tooth surfaces)

Table 3. Stuart-Maxwell test for marginal homogeneity to assess systematic differences in the *in vivo* assessment of caries lesions, both between and within examiners

Examiners	1	2	3
1	P<0.001	*P*=0.030	*P*=0.330
2		*P*=0.093	*P*=0.020
3			*P*=0.444

Discussion

This methodological paper explored issues with examiner calibration in settings where *in vivo* calibration is not practical. The findings present an interesting scenario. On the one hand, moderate to good reliability, in *in vivo* caries diagnosis, was achieved when training and calibration was conducted used photographs. On the other, more caries lesions were detected from intra-oral photographs than *in vivo* on the same tooth surfaces. Two important lines of discussion arise from these observations: (1) the training & calibration of dental examiners and (2) caries detection, both of which are key to collecting quality epidemiological data.

Table 4. Marginal distribution of ICDAS caries codes by examiner from the *in vivo* community-based dental examination

ICDAS Caries Codes (n=880)	Examiners		
	1 (%)	2 (%)	3 (%)
00	89.5	89.2	91.0
01	0.5	0.1	0.1
02	6.4	8.0	6.2
03	1.1	0.9	0.5
04	0.9	1.3	0.6
05	0.8	0.2	0.8
06	0.5	0.1	0.5
Column totals	100	100	100

As stated earlier, calibration using participants in epidemiological studies is a rigorous process and may not always be possible because of the timing of data collection, lack of resources and/or specific population limitations. In such cases, alternative methods such as photographs and extracted teeth have been used, without an *in vivo* component, which may reduce data quality. *In vivo* data quality, which is affected by factors such as: magnification, saliva and debris on the tooth surfaces, lack of appropriate equipment and participants' behavior is rarely assessed following the use of in-vitro calibration. Similarly, the quality of intra-oral photographs is affected by factors such as the inability to move the camera head to a different angle to confirm diagnosis, image size, screen resolution, distance of camera head from tooth, number of teeth per photograph and difficulty in accessing particular tooth surfaces.

This study found that moderate to good reliability can be achieved in *in vivo* caries detection when training and calibration used photographs only. However, when the *in vivo* scores were assessed for systematic bias, one examiner scored differently to the others. This may have been the result of the presentation of caries lesions in vivo. Though the difference was statistically significant, whether it was clinically significant is questionable. This is because the difference was mainly observed in the scoring of non-cavitated lesions and sound surfaces, ICDAS caries codes 0-2.

Table 5. Cross tabulation (marginal frequencies) of caries diagnosis (ICDAS codes) using *in vivo* visual examinations and intra-oral photographs of the same tooth surfaces for one examiner

In vivo ICDAS Scores	Intra-oral Photograph ICDAS Scores												
	00	01	02	03	04	05	06	30	32	34	36	84	Total
00	24	0	16	0	3	2	0	1	0	0	0	0	46
01	0	0	2	0	0	0	0	0	0	0	0	0	2
02	1	0	5	1	0	0	0	0	1	0	0	1	9
03	0	0	3	1	0	0	1	0	1	0	0	0	6
04	0	0	0	0	0	0	0	0	0	0	0	0	0
05	0	0	0	0	0	2	0	0	0	0	0	0	2
06	1	0	0	0	0	1	0	0	0	0	0	0	2
30	0	0	1	0	0	0	0	0	0	0	0	0	1
32	0	0	0	0	0	0	0	0	0	2	0	0	2
34	0	0	0	0	0	0	0	0	0	0	0	0	0
36	0	0	0	0	0	0	0	0	0	0	1	0	1
84	0	0	0	0	0	0	0	0	0	0	0	0	0
Total	26	0	27	2	3	5	1	1	2	2	1	1	71

Stuart Maxwell test for marginal homogeneity: p=0.003

Table 6. Distribution of 22 (of the 46) tooth surfaces that were diagnosed as sound (ICDAS code 00) in the *in vivo* examination but carious when using photographs in Table 5, by tooth location and surface.

Tooth location	Tooth surfaces					Total
	Distal	Occlusall	Mesial	Buccal	Lingual	
Anterior	0	n/a	1	2	2	5
Posterior	2	6	0	5	4	17
Total	2	6	1	7	6	22

[1] Anterior teeth did not have an option to score occlusal surfaces and are hence not applicable (n/a)

Though there is no standard classification for the Kappa score, it is generally accepted that scores of over 0.5 indicate moderate to good reliability (Landis and Koch, 1977). The kappa estimates in this study were similar to those reported in the paper on the development of ICDAS (Ismail, 2007), in spite of being conducted in community conditions and in a challenging population.

Reliability estimates are influenced by the diagnostic method used, as well as by disease prevalence. Greater precision risks greater disagreement. A system like ICDAS with 7 levels of disease severity is more prone to disagreement than the WHO dmf/DMF index, which is dichotomous at the tooth level. Hence, the reason for WHO dmf/DMF achieving Kappa scores of 0.8. This paper used ICDAS rather than the WHO DMFT, because (1) is characterised by several distinct stages of progression not captured by the WHO dmf/DMF assessment of caries lesions, (2) non-cavitated lesions are reversible and (3) disease management should be based on the stage of disease (Fejerskov and Kidd, 2008).

Comparing the *in vivo* and intra-oral photographs, this study showed that lesions detected *in vivo* were also detected using photographs, by the same examiners, with a trend towards greater sensitivity when using photographs. From a calibration perspective, it means that photographs are not missing out those lesions detected *in vivo*, which shows that while *in vivo* is the ideal calibration method, photographs may also be used without adversely affecting data quality. The photographs used in this analysis were selected for their clarity ensuring the absence of introduced artefacts such as light being reflected off the tooth surface. In the absence of artefacts and assuming that the photographs were valid, the better examination conditions and magnification when using photographs could be the reason for more caries being detected using this method. This finding raises the issue of validity of caries detection methods, particularly clinical (visual) versus photographs.

The visual and visual-tactile methods continue to be the standard for caries detection in the clinic. These have also been used as the gold standard in validation studies (Bader *et al.*, 2002, Gomez *et al.*, 2013). However, the strength of evidence to support the validity of this method is poor, calling into question its use as the gold standard (Bader *et al.*, 2002). When studying the validity of caries detection methods, histological sectioning of a tooth for visual evidence of caries activity is considered the most appropriate gold standard, even though it has limitations (Bader *et al.*, 2002). A recent study, comparing the validity of occlusal caries assessments using *in vivo* visual and intra-oral photographs, with histology as the reference standard, demonstrated that the use of intra-oral photographs was the more sensitive method (Boye *et al.*, 2012). Median sensitivity was 81.3% for the photographs compared with 65.6% for the *in vivo* method. Specificity for both methods was similar at 81.3% and 82.4%, respectively. However, these values may be overestimated since the visual assessment and photographs were performed on extracted teeth without the challenges of the *in vivo* oral examination. In addition, caries was assessed at the dentine level, which excluded early non-cavitated caries lesions, which may have also contributed to the high specificity estimates (Boye *et al.*, 2012). Nonetheless, the evidence shows that good quality photographs are more valid for caries detection. Further research is required to improve the strength of this evidence, particularly in relation to non-cavitated lesions.

For epidemiological caries studies and oral health programs focused on prevention, it is important to detect lesions before they become cavitated (Gussy *et al.*, 2015). In such cases, the use of intra-oral photographs to detect caries offers several advantages over the commonly used visual-tactile method: (1) greater sensitivity, (2) lower cost if a trained research assistant takes the photographs, (3) multiple examinations can be performed on the photographs without burdening the participants, (4) reduced participant anxiety as a result of the absence of the power dynamic a dental practitioner brings to the examination and, (5) a database of intra-oral photographs can be created to verify any discrepancies in the assessment, which can also be used for training and calibration of new examiners. These advantages will have to be balanced against disadvantages such as: the cost of the cameras, training and standardisation of personnel, participant compliance, accessibility to the tooth surfaces, photographs providing only a 2-dimensional picture and electronic data management.

A limitation of this study was the number of examiners. More than three examiners are generally recommended to achieve the highest score on a quality rating scale (Bader *et al.*, 2002). It should be noted that the use of the modified ICDAS method (no air drying) eliminates ICDAS caries code 1 on the smooth surfaces and lesions in the pits and fissures that would have only been visible after drying. This would cause an underestimation of the true population caries estimate. However, for this study on reliability estimates and systematic differences, the results would remain unchanged as the caries assessment protocol used was similar across detection methods.

Conclusions

It appears that good quality photographs alone may be used for training and calibration, without adversely affecting data quality. Intra-oral photography could be a more sensitive method for caries detection over *in vivo* visual examination, with promising application in epidemiology. More research is required to determine the most effective and efficient examiner calibration methods in oral health, including larger studies testing different combinations of calibration techniques.

References

Altarakemah, Y., Al-Sane, M., Lim, S., Kingman, A. and Ismail, A. I. (2013): A new approach to reliability assessment of dental caries examinations. *Community Dentistry and Oral Epidemiology* **41**, 309-16.

Aminabdi, N. A., Puralibaba, F., Erfanparast, L., Najafpour, E., Jamali, Z. and Adhami, S. E. (2011): Impact of Temperament on Child Behavior in the Dental Setting. *Journal of Dental Research* **5**, 119-22.

Bader, J. D., Shugars, D. A. and Bonito, A. J. (2002): A systematic review of the performance of methods for identifying carious lesions. *Journal of Public Health Dentistry* **62**, 201-13.

Boye, U., Foster, G. R., Pretty, I. A. and Tickle, M. (2013a): The views of examiners on the use of intra-oral photographs to detect dental caries in epidemiological studies. *Community Dental Health* **30,** 34-8.

Boye, U., Pretty, I. A., Tickle, M. and Walsh, T. (2013b): Comparison of caries detection methods using varying numbers of intra-oral digital photographs with visual examination for epidemiology in children. *BMC Oral Health* **13,** 6.

Boye, U., Walsh, T., Pretty, I. A. and Tickle, M. (2012): Comparison of photographic and visual assessment of occlusal caries with histology as the reference standard. *BMC Oral Health* **12,** 10.

Boye, U., Willasey, A., Walsh, T., Tickle, M. and Pretty, I. A. (2013c): Comparison of an intra-oral photographic caries assessment with an established visual caries assessment method for use in dental epidemiological studies of children. *Community Dentistry and Oral Epidemiology* **41,** 526-33.

Braga, M. M., Oliveira, L. B., Bonini, G. A., Bonecker, M. and Mendes, F. M. (2009): Feasibility of the International Caries Detection and Assessment System (ICDAS-II) in epidemiological surveys and comparability with standard World Health Organization criteria. *Caries Research* **43,** 245-9.

Chattopadhyay, A. (2010): *Oral Health Epidemiology Principles and Practice,* Sudbury, Massachusetts, Jones and Bartlett.

De Silva-Sanigorski, A. M., Calache, H., Gussy, M., Dashper, S., Gibson, J. and Waters, E. (2010): The VicGeneration study--a birth cohort to examine the environmental, behavioural and biological predictors of early childhood caries: background, aims and methods. *BMC Public Health* **10,** 97.

Elfrink, M. E., Veerkamp, J. S., Aartman, I. H., Moll, H. A. and Ten Cate, J. M. (2009): Validity of scoring caries and primary molar hypomineralization (DMH) on intraoral photographs. *European Archives of Paediatric Dentistry* **10 Suppl 1,** 5-10.

Fejerskov, O. and Kidd, E. (eds.) (2008): *Dental Caries: The Disease and Its Clinical Management,* Oxford: Blackwell Munksgaard Ltd.

Fleiss, J. L. (1973): The equivalence of weighted kappa and the intraclass correlation coffecient as measures of reliability. *Educational and Psychological Measurement* **33,** 613-619.

Gomez, J., Tellez, M., Pretty, I. A., Ellwood, R. P. and Ismail, A. I. (2013): Non-cavitated carious lesions detection methods: a systematic review. *Community Dentistry and Oral Epidemiology* **41,** 54-66.

Gussy, M., Ashbolt, R., Carpenter, L., Virgo-Milton, M., Calache, H., Dashper, S., Leong, P., De Silva, A., De Livera, A., Simpson, J. and Waters, E. (2015): Natural history of dental caries in very young Australian children. *International Journal of Paediatric Dentistry*.

Hallgren, K. A. (2012): Computing Inter-Rater Reliability for Observational Data: An Overview and Tutorial. *Tutor Quant Methods Psychol* **8,** 23-34.

Ismail Ai, S. W., Tellez M, Amaya a, Sen a, Hasson H, Pitts Nb (2007): The International Caries Detection and Assessment System (ICDAS): an integrated system for measuring dental caries. *Community Dentistry and Oral Epidemiology* **35,** 170-178.

Landis, J. R. and Koch, G. G. (1977): The measurement of observer agreement for categorical data. *Biometrics* **33,** 159-174.

Litaker, M. S., Gordan, V. V., Rindal, D. B., Fellows, J. L. and Gilbert, G. H. (2013): Cluster Effects in a National Dental PBRN restorative study. *Journal of Dental Research* **92,** 782-7.

Maclure, M. and Willett, W. C. (1987): Misinterpretation and misuse of the kappa statistic. *American Journal of Epidemiology* **126,** 161-9.

Masood, M., Masood, Y. and Newton, J. T. (2015): The Clustering Effects of Surfaces within the Tooth and Teeth within Individuals. *Journal of Dental Research* **94,** 281-8.

Nelson, S., Eggertsson, H., Powell, B., Mandelaris, J., Ntragtakis, M., Richardson, T. and Ferretti, G. (2011): Dental examiners consistency in applying the ICDAS criteria for a caries prevention community trial. *Community Dental Health* **28,** 238-242.

Pine, C. M., Pitts, N. B. and Nugent, Z. J. (1997): British Association for the Study of Community Dentistry (BASCD) guidance on the statistical aspects of training and calibration of examiners for surveys of child dental health. A BASCD coordinated dental epidemiology programme quality standard. *Community Dental Health* **14,** 18-29.

Topping, G. V. A., Hally, J., Bonner, B. and Pitts, N. B. (2008): International Caries Detection and Assessment System (ICDAS) e-Learning Package. Interactive CD ROM and Web-based software. London: Smile-on.

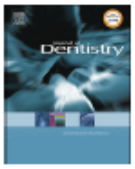
Appendix 5. Research Strand 3, published paper: CRAT Testing

Citation: *Christian B, Calache H, Adams G, Hall M, Dashper S, Gibbs L, et al. A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children. Journal of dentistry. 2020;95:103324.*

Contents lists available at ScienceDirect

Journal of Dentistry

journal homepage: www.elsevier.com/locate/jdent



A methodological study to assess the measurement properties (reliability and validity) of a caries risk assessment tool for young children

Bradley Christian[a,e,*], Hanny Calache[b], Geoff Adams[c], Martin Hall[a,d], Stuart Dashper[c], Lisa Gibbs[e], Mark Gussy[f]

[a] Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University, Bendigo, Victoria, Australia
[b] Centre for Population Health Research, Deakin University, Burwood, Victoria, Australia
[c] Melbourne Dental School, The University of Melbourne, Melbourne, Australia
[d] Dental Health Services Victoria, Melbourne, Australia
[e] Jack Brockhoff Child Health and Wellbeing Program, Centre for Health Equity, Melbourne School of Population and Global Health, The University of Melbourne, Melbourne, Australia
[f] Lincoln International Institute for Rural Health, College of Social Science, University of Lincoln, Lincoln, United Kingdom

ARTICLE INFO

Keywords:
Dental caries
Risk factors
Children
Birth cohort
Risk assessment

ABSTRACT

Objectives: To assess the CAMBRA caries risk assessment tool (CRAT) in terms of its measurement properties: reliability, validity and responsiveness to change in caries estimates over time.

Methods: Secondary data from the VicGeneration (VicGen) birth cohort study were used. Caries risk status at child age18 months was compared against caries development at 36 and 48 months. Questionnaire data for the 18-month-old children, were used to generate the CRAT item responses. Five examiners assessed the caries risk profile for each 18-month-old child and assigned a risk status. The analytic method was informed by the COnsensus-based Standards for the selection of health Measurement INstruments Checklist.

Results: Each examiner applied the CAMBRA CRAT to 214 cases from the VicGen study. At 18 months of age, the proportion of low-, moderate- and high-risk children were 17 %, 17 % and 66 %, respectively. Caries prevalence at 36 and 48 months of age was 18 % and 35 %, respectively. Almost perfect reliability (both inter- and intra-reliability) was achieved in caries risk categorisation. For criterion validity and responsiveness, the area under the curve was approximately 0.50. For the dichotomised risk, the sensitivity of the CAMBRA CRAT to predict caries at 36 and 48 months, was 74 % and 70 %, respectively. The corresponding specificity scores were 35 % and 36 %, respectively.

Conclusions: The results from this study show that the CAMBRA CRAT, in its current form, is not a good predictor of future caries among young Victorian children and in fact overestimates the proportion of children at high-risk of future disease. As such, it may not be ideal for use among young Victorian children and it is highly likely it's use could be driving over treatment.

Clinical Significance: : This study provides evidence for the applicability of the CAMBRA CRAT with young Victorian children to inform decision making on caries risk-based management. It also highlights the important measurement properties and related analytics (applicable to any CRAT) that should be considered when selecting a CRAT for use in clinical practice.

1. Introduction

Dental caries in children is common. In the deciduous dentition the disease can manifest as early as the first year of life and continue until exfoliation [1]. The factors contributing to caries risk in the early years (0–5 years) have been shown to differ from those in older children [2]. This is understandable given the rapid changes in diet, tooth development and oral hygiene practices as a child progresses through developmental stages from infant to preschool age. The well substantiated influences on caries in young children are: parenting behaviours, knowledge, beliefs and attitudes; infant feeding practices; maternal circumstances and oral health; and infant-related oral health behaviours [2,3]. Understanding the age specific influences on child oral health and using this knowledge to inform the design and

* Corresponding author at: Room 2.31, HHS 1, Dentistry and Oral Health, La Trobe Rural Health School, La Trobe University, Bendigo, Victoria 3550, Australia.
E-mail address: b.christian@latrobe.edu.au (B. Christian).

https://doi.org/10.1016/j.jdent.2020.103324
Received 7 September 2019; Received in revised form 14 March 2020; Accepted 17 March 2020

0300-5712/ © 2020 Elsevier Ltd. All rights reserved.

implementation of interventions (including service delivery), has greater potential to translate to effective action, compared with blanket (broad spectrum) interventions to improve oral health or service delivery across all ages.

A recent study showed that caries prevalence, among children in Victoria, Australia, increased from 8 % to 23 % between 18 and 36 months of age, with the development of new lesions on previously sound tooth surfaces [4]. It is evident from the study by Gussy et al. [4], that the time period between 18 and 36 months is a window of opportunity for early intervention and disease prevention. However, to act effectively during this window of opportunity, at-risk children need to be accurately and precisely identified. Valid and reliable tools that discriminate children truly at risk of dental caries from those not at risk are required.

Risk-based management using caries risk assessment tools (CRATs) is promoted as a way of: identifying those at highest risk of disease, guiding treatment decisions, guiding discussions with patients (including parents/caregivers) regarding caries risk/protective factors, determining appropriate recalls and to inform the economics of producing, delivering and using health care [5]. However, whether risk-based management is achieving these outcomes for either the patient or the health service organization is unclear. From the information reported in two recent systematic reviews of the measurement properties (reliability and validity) of CRATs [6,7], it can be extrapolated that current CRATs used in risk-based management may in fact result in overtreatment and as such are wasteful of resources. This has major implications for public/community dental services with limited resources and where risk-based management is used as a mechanism to ensure appropriate resource allocation, so that people with the most need get the required care. Hence, research is urgently required to provide the evidence to demonstrate that risk-based management and CRATs are an efficient and effective method to improve oral health and health service outcomes.

Numerous caries risk assessment tools (CRATs) are currently being used in clinical dental practice with the popular ones being: Caries Management by Risk Assessment (CAMBRA), American Academy of Pediatric Dentistry CRAT and Cariogram [8–10]. These CRATs are a collation of various caries risk assessment methods (CRAMs), that have been compiled into prediction models, with the assumption that together the CRAMs will be able to predict future disease better than any single method alone. As stated previously, the evidence that CRATs accurately predict future disease is unclear and in fact may be resulting in overtreatment and wastage of resources. Commonly used CRAMs include socio-economic status, dental service use, diet and oral health behaviours.

The ability of a CRAT to perform well by correctly and consistently identifying those individuals at risk of future disease is affected by the quality of two core measurement properties – reliability and validity. Reliability is the property of the CRAT to produce similar results under different conditions, whereas validity relates to the degree to which the CRAT actually measures what it was designed to measure. Reliability includes the domains of internal consistency and measurement error. Validity includes the domains - content, construct, and criterion validity. Responsiveness, a relatively new addition to these classic measurement properties, is the ability of an instrument to detect change in the construct over time [11].

Evidence for the quality of measurement properties of CRATs is required to inform tool selection for use in practice, however, current research on the topic is very limited [7,12]. To investigate this topic in-depth, we conducted a comprehensive systematic review on the quality of measurement properties for current CRATs for young children [6]. This review identified a major knowledge gap by demonstrating the lack of methodological studies to provide the evidence to assess the quality of the measurement properties for the identified CRATs.

The authors of this study aimed to address the knowledge gap by conducting a methodological study to assess the measurement properties for the CAMBRA CRAT. Specifically, the objectives were to assess the CAMBRA CRAT in terms of its: reliability in application, validity in predicting future caries in young children, and responsiveness to change in caries estimates over time. The CAMBRA CRAT for children under 6 years of age was selected for use in this study, as it showed strong evidence for content validity [6]; and although not the best performing CRAT in our recent review, is currently (in its original or modified versions) the most commonly used CRAT across many public dental services in Victoria, Australia. The CAMBRA CRAT was used as a case study and the method demonstrated in this study could be used as a road map for testing other CRATs.

2. Materials and methods

The University of Melbourne Human Research Ethics Committee approved this study Ethics ID 1851421.

The CAMBRA CRAT was assessed using a secondary analysis of data from the VicGeneration (VicGen) study, an oral health birth cohort study to understand the multifactorial nature of early childhood caries development [13]. Seven waves of data collection occurred in VicGen, at child ages (months) 1, 6, 12, 18, 36, 48 and 60; and each wave included a clinical dental examination, saliva collection and a self-administered questionnaire [13]. Dental caries was assessed visually (no probe) and recorded using the International Caries Detection and Assessment System (ICDAS-II) [14]. The ICDAS-II protocol was modified to suit the field setting by omitting air-drying of teeth and as such the ICDAS-II caries code 1 on smooth surfaces of examined teeth could not be detected. All clinical examiners were trained and calibrated with good examiner reliability in caries diagnosis [15]. Structured self-administered questionnaires were used to collect data (caregiver self-reported) on: family socio-demographic-economics; child feeding habits; child and caregivers' oral hygiene-related behaviour; self-reported oral/general health status for child and care-giver; care-giver's knowledge and attitudes about oral health. A more comprehensive description of the VicGen study methods has been published previously [13].

Based on the data collected in the VicGen study, the premise of this study was to assign a caries risk level to the 18-month-old children prior to the onset of the caries disease process (clinically apparent demineralisation of the dental hard tissue) and validate this risk assignment against future caries lesion development in these children. Questionnaire data for 18-month-old children were used to generate the CAMBRA CRAT item responses. This process gave each 18-month-old child a completed CAMBRA caries risk profile where each CRAT item had a response. Five examiners with a background in clinical dentistry assessed the caries risk profile for each 18-month-old child and assigned a risk status (high-, moderate-, or low-risk). For the reliability analysis, risk assessment data from all the clinical examiners were used. The risk assessments were a desktop exercise. Caries risk status at child age at 18 months of age was compared against actual caries development at child 36 and 48 months of age, as reported in the VicGen data.

The analysis method for this study was informed by the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist [11]. Though the COSMIN checklist was developed to evaluate the methodological quality of studies on measurement properties of Health-Related Patient-Reported Outcomes (HR-PROs), it can be applied to other health-related measurement instruments, such as CRATs, as the design requirements and measurement properties are similar [11].

3. Sample size considerations

To achieve an excellent rating for the quality of a measurement property, the COSMIN checklist recommends a sample size of at least 100 cases. This study included 214 cases which had complete data across the three time points – 18, 36 and 48 months of age.

Table 1
CRAT caries indicators/factors and the corresponding VicGen questions.

CAMBRA CRAT Items	VicGen variables at child 18 months of age	CRAT item derivation and assumptions
Risk indicators (parent interview)		
Mother/caregiver: active caries in the past 12 months	Mother/caregiver caries estimates at child 6 months of age.	Caries lesion activity status was not recorded in VicGen. *Assumption:* A parent was assumed to have active caries in the past 12 months, if they were in the top one third (66th percentile) of parents with the highest DMFT scores ('D' defined as caries into dentine). For this sample of parents, the DMFT score at the 66th percentile was 9.
Child: recent restorations	Not applicable as no restorations at this age.	This CRAT item was excluded from the caries risk assessment.
Parent/caregiver: low socioeconomic status and/or low health literacy	What is the highest post school education that you have completed? Do you or your partner have a health care card?	For this CRAT item, a composite variable was created from the two VicGen variables. *Assumption:* Any respondent with less than an advanced diploma qualification was considered as having low health literacy. *Composite variable:* Any respondent with less than an advanced diploma qualification or who had a health care card.
Child: developmental problems	Has the doctor ever told you that your child has developmental problems?	*Derived variable:* Same as the VicGen variable.
Child: no dental home/episodic dental care	How often does your child have a dental check-up?	*Derived variable:* Same as the VicGen variable. *Assumption:* A response of 'never' or 'less than 1/year' was assumed to be evidence of 'no dental home/episodic dental care'.
Risk factors (biological) (parent interview)		
Child: frequent (> three times daily) between-meal snacks of sugars/cooked starch*/sugared beverages	Does this child drink any of the following? In a usual week how often does the child have the following foods?	For this CRAT item, a composite variable was created from the two VicGen questions. *Composite variable:* Child reported to consume > 3 cariogenic food or drink per day. Decisions on which food and drinks to include were informed by the published VicGen cariogenic scale paper [31]. Only food and drink classified as moderately cariogenic and higher were included for this composite variable.
Child: saliva-reducing factors (medications and medical)	Medically compromised children were not included in VicGen.	This CRAT item was excluded from the caries risk assessment.
Child: continually uses bottle – contains fluids other than water	Does the child sip from bottle on and off during the day? What is usually in the bottle?	For this CRAT item, a composite variable was created from the two relevant VicGen questions. *Composite variable:* Child sips from bottle on and off during the day, and it usually contains fluids other than water.
Child: sleeps with a bottle or nurses on demand	Does this child usually take a bottle to bed?	*Derived variable:* Same as the VicGen variable.
Non-biological protective factors (parent interview)		
Mother/caregiver: decay-free last three years	Mother/caregiver caries estimates at child 6 months of age.	*Assumption:* The mother/caregiver was assumed to be decay free the last three years if they had no decayed teeth ('D' component of the DMFT score) at child age 6 months. The 'D' was defined as an ICDAS caries score of 4-6. The evidence shows that it takes approximately 77 months for enamel lesions to progress to dentine [32]. Hence, those parents with large cavities were most likely not decay free in the past 3 years.
Child: dental home and regular dental care	How often does your child have a dental check-up?	*Derived variable:* Same as the VicGen variable. *Assumption:* A response of '1/year or more' was assumed to be evidence of 'dental home and regular dental care'.
Biological protective factors (parent interview)		
Child: lives in a fluoridated community or takes fluoride supplements by slowly dissolving or as chewable tablets	Water fluoridation status at the postcode where the child lived.	*Derived variable:* Water fluoridation status for the postcode of each case, approximated to the time of data collection when the child was 18 months of age.
Child: fluoridated toothpaste (pea-size) daily.	What type of toothpaste do you use for child? How much toothpaste do you use for child?	For this CRAT item, a composite variable was created from the two relevant VicGen questions. *Composite variable:* Child reported to use fluoridated toothpaste at least 1/day and a pea size amount of toothpaste.
Mother/caregiver: xylitol chewing-gum/lozenges 2–4 × daily	In the last week, how many times did you chew sugar free gum for at least 10 minutes continuously?	*Derived variable:* Same as the VicGen variable.
Risk indicators/factors (child clinical examination)		
Obvious white spots, decalcifications, or obvious decay	Child ICDAS caries codes at 18 months of age.	This CRAT item was excluded from the caries risk assessment as all children were caries lesion free at 18-months of age.
Restorations in the last 2 years	Not applicable as no restorations at this age.	This CRAT item was excluded from the caries risk assessment as all children were caries lesion free at 18-months of age.
Obvious plaque on teeth and/or gums bleed easily	Child clinical examination at 18 months of age.	*Derived variable:* Visible plaque on anterior teeth. *Assumption:* If there is evidence of plaque on anterior teeth it was assumed that there was plaque on the posterior teeth too.
Visually inadequate saliva flow (dry mouth)	Not assessed in VicGen.	This CRAT item was excluded from the caries risk assessment.

4. CAMBRA CRAT items and VicGen variables

As the CAMBRA CRAT items did not have specific definitions, the author team defined, jointly discussed and agreed on the VicGen variables that best represented the CAMBRA CRAT items. In some cases, two or more VicGen variables were aligned with the CRAT item and in such cases composite variables of the two (or more) variables were generated. For CRAT items where there was no clearly aligned VicGen variable, logic and evidence-informed assumptions were used to derive the appropriate variables. The CAMBRA CRAT items, the corresponding VicGen variable(s) at 18-months of age and the assumptions used to derive the CAMBRA CRAT item variables are shown in Table 1.

5. Data cleaning and management

STATA was used to clean and manage the VicGen data, and in creating the analysis datasets. Four separate datasets were used to create the master dataset – the 18-month questionnaire data and the clinical data for 18, 36 and 48 months of age. The master dataset was then used to derive the variables for the CAMBRA CRAT items based on the 18-month questionnaire data. Once the CAMBRA variables were generated, another dataset with only the child unique identification numbers and the CAMBRA CRAT items was created. This CAMBRA dataset was used for the next stage of the research – developing the online caries risk assessment forms and the risk assessment process.

REDCap (Research Electronic Data Capture Version 9.1.21), hosted at the author's institution, a secure web platform for building and managing online databases and surveys, was used to create the web-based caries risk assessment forms and manage the risk assessment process and data [16]. VicGen data were imported into RedCap and a CAMBRA CRA form was created for each child, which had responses for each CRAT item. The examiners then completed the risk assessments in REDCap, based on the CAMBRA item responses, and categorised each case as high-, moderate- or low-risk for future dental caries.

6. Caries risk assessments

Five examiners (four examinations as BC + HC jointly examined to establish the reference standard) with a background in clinical dentistry were trained to apply the CAMBRA CRAT using the online CAMBRA Basic training course available at https://course.mycambra.com/courses/cambra-basic/. The caries risk assessments were conducted in REDCap, where the examiner was required to determine whether a child was high-, moderate- or low-risk based on yes/no responses to the CAMBRA CRAT items (which were pre-populated into the CAMBRA CRAT REDCap form). Caries risk was summated based on the balance of risk vs protective factors – high, if more risk than protective factors; moderate, if equal numbers of both risk and protective; and low, if more protective than risk factors.

Two examiners (HC and BC) established the reference standard caries risk status at 18 months of age for each child, by jointly assessing 25 cases to achieve consensus on the scoring method, which was followed by an independent assessment of a further 50 cases where any discrepancies in the assessments were resolved via discussion. Once consensus was achieved, BC assessed the rest of the cases to establish the reference standard risk categorization for the 18-month-old children. This reference standard was used in the validation analysis. To reflect real world conditions in current practice, examiners were not calibrated in using the CAMBRA CRAT.

7. Data analysis

On completion of the caries risk assessment stage of the research, the data were exported from REDCap into STATA (version 15.1, StataCorp, College Station, TX, USA) for analyses. Eighteen-month-old children who had a clinical examination at 36 and 48 months of age were included in this analysis (n = 214). All children were caries free at 18-months of age. In this analysis, caries/caries lesion refers to any clinically apparent sign of tooth demineralisation [non-cavitated and cavitated lesions (ICDAS-II scores 1–6)]. A sub-analysis was also conducted to determine which CAMBRA CRAT items were responsible for the increased high-risk classification.

7.1. Reliability and measurement error

A fully-crossed design was used, where the same examiners were used for all study participants, therefore allowing systematic bias to be assessed and controlled for in the reliability estimate [17]. Both inter- and intra-examiner reliability estimates were calculated. For inter-examiner reliability, three examiners and the reference standard examination (BC + HC joint examination) assessed all 214 cases (blinded to each other's assessment) and assigned each case to a risk category (high-, moderate- or low-risk) based on responses to the CAMBRA CRAT items. For intra-examiner reliability, the first 100 cases were re-assessed by all examiners a few days after the first round of assessments. Reliability estimates were reported as weighted kappa scores and 95 % confidence intervals (95 %CI) and computed using the 'kapci' command in STATA with the 'rep' option set at 999. This option uses a bootstrap method to generate estimates of the 95 % CI. To assess systematic bias i.e. measurement error in the caries risk assessments, the Stuart-Maxwell test for marginal homogeneity was conducted to determine if examiners were systematically scoring differently from other examiners or within themselves.

7.2. Content validity

The content validity of the CAMBRA CRAT was assessed in our recently published systematic review on the quality of measurement properties for current CRATs for young children [6].

7.3. Construct (discriminatory) validity

Correlation estimates were computed between caries risk status at 18 months of age and clinical evidence of caries at both 36 and 48 months of age. It was hypothesised that being categorised as high-risk at 18 months of age would be correlated with the greater likelihood of developing caries lesions at the two future time points, compared with being low- or moderate-risk. As the variables used to assess this measurement property were ordinal, Spearman's rank coefficients were used to estimate correlation between variables. The distribution of children with and without caries by risk status was also explored as part of the assessment for this measurement property.

7.4. Criterion validity

The reference standard for this measurement property was caries development at 36 and 48 months of child age. For the classic CAMBRA CRAT categorisation of low-, moderate- and high-risk (no extreme risk), the area under the curve (AUC) was estimated. In addition, for consistency and comparison with other research on this topic, the risk categories were dichotomised into high-risk versus low-risk (low and moderate risk categories were combined) and the following measures reported - sensitivity, specificity, positive predictive values and negative predictive values.

7.5. Responsiveness

The only difference between criterion (concurrent) validity and responsiveness is that criterion validity refers to the validity of a single score, and responsiveness refers to the validity of a change score. The change score for this analysis was the percentage of disease-free children at 36 months who developed caries lesions at 48 months of age. The analysis conducted were the same as for criterion validity.

8. Results

Five examiners applied the CAMBRA CRAT to 214 cases from the VicGen birth cohort study. At 18 months of age, the proportion of children categorised as low-, moderate- and high-risk were 17 %, 17 % and 66 %, respectively. The prevalence of caries at 36 and 48 months of age was 18 % and 35 %, respectively. Results of the sub-analysis showed that the CRAT items responsible for the higher proportion of high-risk categorisations were related to the child's dental service utilisation - 'Child: no dental home/episodic dental care' and 'Child: dental home and regular dental care'. The following results are presented by

Table 2
Weighted Kappa scores (95 % Confidence Intervals) for reliability and the Stuart-Maxwell test (*P*=) for marginal homogeneity in risk categorisation using CAMBRA.

Examiners	1	2	3	4
1	0.98 (0.93–1.00) *P* = 0.61	0.97 (0.94-0.99) *P* = 0.37	0.96 (0.89-0.99) *P* = 0.61	0.98 (0.97–1.00) *P* = 1.00
2		1.00 *P* = 1.00	0.96 (0.88-0.99) *P* = 0.22	0.98 (0.96–1.00) *P* = 0.37
3			1.00 *P* = 1.00	0.97 (0.91–1.00) *P* = 0.61
4				0.98 (0.92–1.00) *P* = 0.61

measurement property.

8.1. Reliability and measurement error

The trained examiners achieved almost perfect reliability (both inter- and intra-reliability) in caries risk categorisation using the CAMBRA CRAT (Table 2). There was no evidence of a measurement error either between or within examiners.

8.2. Discriminatory validity

Fig. 1 shows the proportions of children with caries by risk status at 36 and 48 months, respectively. There was some evidence of a correlation between the caries risk assessment at 18 months and actual caries development at 36 months of age (Spearman correlation coefficient = 0.13, *P* = 0.06), but not at 48 months of age (Spearman correlation coefficient = 0.08, *P* = 0.27).

8.3. Criterion validity

For the CAMBRA three-level risk categorisation (low, moderate and high), at 36 and 48 months of age, respectively, the AUC was close to 0.50, which indicates a low ability of the CRAT to predict future caries development. Table 3 shows the measures of criterion validity for the dichotomised risk categorisation (high-risk vs low-risk) and caries development at 36 and 48 months. The sensitivity (95 % CI) of the CAMBRA CRAT to predict caries at 36 and 48 months, was 74 % (58 %, 87 %) and 70 % (58 %, 80 %), respectively. The corresponding specificity scores were 35 % (28 %, 43 %) and 36 % (28 %, 44 %), respectively.

8.4. Responsiveness

The measures to determine the responsiveness of the CAMBRA CRAT to the change in caries development between 36 and 48 months (Table 3), followed a similar pattern to those for criterion validity with sensitivity and negative predictive values achieving the highest scores, 64.8 % and 73.6 %, respectively. The AUC was 0.48.

9. Discussion

The results of this study show that the CAMBRA CRAT in its current form may not be useful in this population. An obvious observation from the analysis was the higher proportion of children (66 %) designated as high-risk at baseline (18-months of age) compared with the proportion that developed caries lesions at 36 (18 %) and 48 (35 %) months of age. Gao et al. [18], made a similar observation in their validation study and reported that the majority of children were considered high-risk when the CAMBRA CRAT was used. The sub-analysis on this issue revealed that the CRAT items related to child's dental service utilisation were driving the higher proportion of high-risk categorisations - 'Child: no dental home/episodic dental care' and 'Child: dental home and regular dental care'. To investigate whether the omission of these two items improved validity estimates, the full analyses were replicated on a dataset where these two items were removed (not reported in this paper because the modification did not improve criterion validity and our study was to test the CAMBRA CRAT in its original form). Though this modification to the CAMBRA CRAT reduced the proportion of 18-month-old children assessed as being at high-risk (dropped from 66 % to 51 %), it did not improve estimates of validity. In very young children the utility of 'child dental service use' items as a risk indicator should be further investigated. Systematic reviews of risk factors/indicators in the early years do not show child dental service use as being

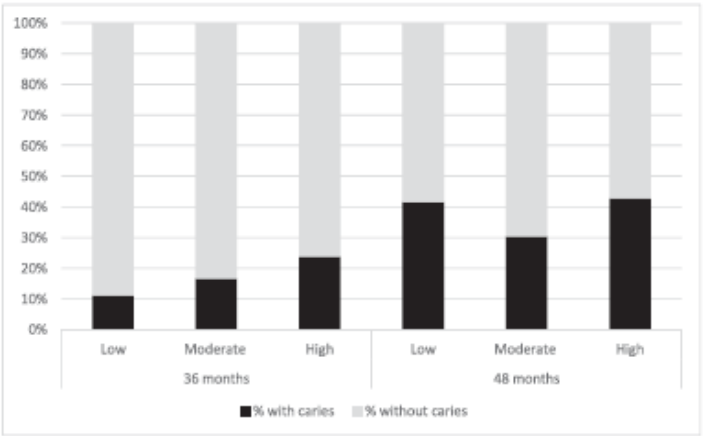


Fig. 1. The proportion of children with and without caries lesions at 36 and 48 months of age, by risk status (determined at 18 months of age).

Table 3
Criterion validity of the dichotomised (high-risk vs no-risk) caries risk assessment at 18 months of age, in predicting caries development at 36 and 48 months of age; Responsiveness of the CRAT in identifying children with caries at 48 months who were disease free at 36 months.

	Disease prevalence (%)	Sensitivity (%)	Specificity (%)	PPV (%)	NPV (%)
Criterion validity 36 months	18.2(13.2,24.0)	74.3(57.8,86.9)	35.4(28.3,43.0)	20.4(14.1,28.0)	86.1(75.9,93.1)
Criterion validity 48 months	34.5(28.2,41.3)	70.3(58.5,80.3)	35.7(27.8,44.2)	36.6(28.7,45.1)	69.4(57.4,79.7)
Responsiveness	**% of new cases at 48 months**	**Sensitivity (%)**	**Specificity (%)**	**PPV (%)**	**NPV (%)**
	25.2(19.5,31.6)	64.8(50.6,77.3)	33.1(25.9,40.9)	24.6(17.8,32.5)	73.6(61.9,83.3)

PPV-positive predictive value; NPV-negative predictive value.

associated with early childhood caries [2,19]. The recent paper by Chaffee et al. [20], validating the CAMBRA 0–5 CRAT and its individual items did not include the CRAT items for dental service use (dental home) and as such the use of this indicator as a CRAT item for very young children is not clear.

Evidence for the reliability and measurement error in the application of CRATs is limited in the literature [12]. The examiners in this study demonstrated excellent reliability in the application of the CAMBRA CRAT to assign caries risk status to individual cases. During the training the examiners were instructed to be very objective in the application of the CRAT, i.e. rather than making subjective decisions based on particular risk/protective factors/indicators, the examiners were instructed to assign risk based on the balance of 'yes' and 'no' responses to the CRAT items, as recommended in the CAMBRA training course. For example, three 'yes' responses to risk factors/indicators versus two 'yes' to protective factors/indicators tipped the balance towards being high-risk for future caries. Such objectivity in the risk assessments contributed to the excellent reliability scores. Reporting on the examiner's reliability in the application of CRATs has generally been ignored in the literature on CRAT development, and also in studies where CRATs were used in practice. The reliability in risk assessments affects the reliability in allocating patients to care pathways and disease management, which in turn has implications for resources and cost. In public and community dental services, which operate on a tight budget, risk-based management is used as a method to control demand and waiting lists (using risk-based recall protocols) and to ensure the most urgent cases are prioritised for care. A lack of consistency among clinicians in applying the risk-based disease management protocols, particularly low-risk patients being incorrectly recalled too soon, will have significant implications on the services' ability to deliver positive outcomes and to ensure efficient use of its resources. Hence, one of the first steps in a risk-based model of service delivery is to ensure consistency in the risk-assessments and associated risk-based management pathways.

Studying the correlation of risk categories to future lesion development, a positive (non-significant) correlation was observed between the three risk categories at 18-months and caries lesion development at 36 months, but not at 48 months of age. Chafee et al. [20], also reported a positive correlation for the CAMBRA CRAT, where increasing risk was strongly associated with more caries lesion development at follow-up. When reporting on the distribution of caries lesion development by risk categories, it is also important to simultaneously report on the distribution of healthy/disease free individuals by risk categories. It may be the case that while the distribution of those people with caries increases with increasing risk, the distribution of healthy people does not decrease with increasing risk, as was observed in this study. The observed trend of more children with caries at 36 months of age in the high-risk category could be an artefact of the over categorisation as high-risk at 18-months-of age. That is, this finding could be mirroring the distribution of the high-risk categorisation rather than the CRAT being truly discriminative in identifying children with caries. The discriminant property of a CRAT is dependent on testing and reporting of such estimates, however, the literature on this measurement property for CRATs is relatively non-existent.

Criterion validity is the most studied and commonly reported measurement property for CRATs [6,7,12]. A possible reason for the focus on criterion validity (while ignoring other measurement properties) is the view that CRATs are similar to medical diagnostic tests for which the validation method is well established, where a new test is validated against an established reference/gold standard; and tools developed to assess the quality of diagnostic tests (such as the Quality Assessment for Diagnostic Accuracy Studies – QUADAS) have been adopted in CRAT research [21,22]. However, CRATs are made up of multiple caries risk/protective factors/indicators which have been complied together, with the rationale being that together these factors/indicators will improve outcome prediction compared with only using a single factor/indicator. As such, in the case of CRATs all the domains of validity (content, construct and criterion) require careful investigation. However, the methods to adequately test content and construct validity are relatively new and yet to be widely adopted in CRAT research.

In this study, for the CAMBRA CRAT risk levels (low-, moderate- and high-risk) the criterion validity analysis showed that the ability of the CAMBRA CRAT to predict caries, at 36 and 48 months among disease free 18-month-old children, was very low (the AUC was close to 0.50 for both ages). When the risk categorisation was dichotomised to high- and low-risk (moderate and low were combined), sensitivity of the CAMBRA CRAT was between 70 %–74 % while specificity was low at approximately 35 %. Similar findings of acceptable sensitivity but low specificity scores for the CAMBRA CRAT have been reported previously [18,20]. Several issues need to be explored to help explain the low validity for the CAMBRA CRAT among these children. First, was the CAMBRA0–5 CRAT developed with the intention that it would be predictive of caries lesion development in young children who currently had no sign of the disease process (i.e. sound tooth surface)? The answer to this specific question is probably 'yes' as the main paper on the CAMBRA CRAT for young children [23], states the following "To achieve the best management and outcomes for good dental health, an appropriate caries risk assessment screening must be executed as early as possible and preferably prior to the onset of the disease process." [p. 688]. The original consensus statement on CAMBRA in 2002 [10], clearly states that CAMBRA was developed out of a clear need to manage the underlying causes (risk factors) for dental caries "rather than just the results of the disease" [p. 258], in an attempt to maintain the balance between the protective and pathologic factors [24]. Second, the definitions for each of the CRAT items are not explicitly stated in the CAMBRA background papers. For example, how was the CAMBRA item 'Parent/caregiver: low socioeconomic status and/or low health literacy' to be determined? The reason could be that the CAMBRA developers wanted it to be adaptable to any situation or population. However, in the absence of definitions, the authors of this study used their own interpretation and evidence to derive the CRAT items from the VicGen data, as shown in Table 1. It is possible that the authors' item definitions were different to those used in the development of CAMBRA and as such could be a reason for the low validity estimates in this study. Third, the use of the three risk-levels (no extreme risk as special needs and medically compromised children were excluded from the VicGen study) in very young children who have no signs of the disease process, may be diluting the predictive capacity of the CRAT. A better option, as shown in this study, could be to have only two risk-levels – high/at-risk and low-risk. Interestingly, the two other studies that report on

criterion validity for the CAMBRA CRAT both report a similar dichotomised risk [18,20], with the emphasis being on predicting caries among high-risk children. Another reason for dichotomising risk in this study was our observation of clinical practice data showing that clinicians tend to categorise most people as moderate risk (the safe option), which in terms of caries management triggers shorter recall periods and more intense preventive services being provided, when they were not required i.e. no effect on oral health outcomes. This again goes back to the crux of the problem, that current CRATs result in over treatment and wastage of resources. By dichotomising the risk assessment scores we are likely to reduce the possibility of over treatment and wastage of resources Fourth, the predictors of dental caries among young children in Victoria, Australia may not be the same for children in San Francisco, USA, where CAMBRA was developed and tested. In fact, a recent cross-sectional study among migrant pre-school aged children in metropolitan Melbourne, Australia, reported length of residence in Australia as an independent predictor of caries [25]. Results from our recently published longitudinal analysis of predictors for early childhood caries among Victorian children, also using data from the VicGen birth cohort, shows a relatively different mix of independent risk and protective factors, compared with those currently in the CAMBRA CRAT [26]. The University of North Carolina Caries Risk Assessment studies (UNCCRA) also clearly demonstrated the variation in independent predictors of dental caries, by population [27,28]. Understanding the drivers of disease in a specific population is very important, as it highlights the potential risk of adopting CRATs without testing them for relevance to the population being served. Finally, the time between the risk assessment and first signs of the disease process may be long enough to allow for the risk/protective factors from the original assessment to have changed. For example, it is well-known that feeding practices are quite dynamic in the early years [29,30], so a risk assessment at 18-months of age may only be valid up to that age where the risk/protective factors remain the same. Future research should study the time-based nature of caries risk profiles. It should also be noted that the reliability in diagnosing the clinical outcome can be a potential source of variability in the validation of risk categorisation and as such should be reported in studies on caries risk assessments. The clinical examiners in VicGen were shown to have moderate to good reliability in diagnosing dental caries using ICDAS and as such we do not believe this was a major source of variability in the CRAT assessment outcomes for this study.

An obvious limitation of our analysis was the use of data that were not collected specifically for the CAMBRA CRAT items. As a result, the created variables may not be the same as those used in the development of CAMBRA. However, given that the assumptions were extensively discussed with experts in the field and where possible validated using evidence, the authors believe that the generated variables were the best representation of CAMBRA items for the Victorian child population. The ideal methodological study would have collected data specific to the CRAT of interest and then followed the cohort of children prospectively to measure caries lesion development. This method, however, is resource and time intensive and in most cases may not be feasible. The use of secondary data is the more viable option, and, in such cases, variable derivation should be guided by discussion and evidence-informed assumptions.

10. Conclusions

This study provides the evidence to show that the CAMBRA CRAT, in its current form, may not be ideal for use in risk-based disease management among young Victorian children. Due to its low specificity scores it is highly likely that the use of this CRAT in Victoria could be driving over treatment, which is a major issue particularly for public dental services where risk-based service delivery is used as a resource allocation and waitlist management method, to prioritise care for those most at risk of future disease. An important next step on this topic of research is to investigate, at the practice/service delivery level, whether caries risk assessments are aligned with the appropriate management strategies.

Future research on the topic of CRATs should follow a similar methodological approach and endeavour to assess all relevant measurement properties; which will assist clinicians and decision makers in CRAT selection for use in practice.

CRediT authorship contribution statement

Bradley Christian: Conceptualization, Methodology, Software, Data curation, Writing - original draft, Writing - review & editing, Project administration, Formal analysis. **Hanny Calache:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Supervision. **Geoff Adams:** Conceptualization, Methodology, Data curation, Writing - original draft, Writing - review & editing, Supervision, Formal analysis. **Martin Hall:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Supervision. **Stuart Dashper:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Supervision. **Lisa Gibbs:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Supervision. **Mark Gussy:** Conceptualization, Methodology, Writing - original draft, Writing - review & editing, Supervision.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgements

Dr. Ghulam Murtaza, Digital Research Analyst, La Trobe University, for support with REDCap. Ms. Lauren Carpenter for advice on the VicGen data. The clinical examiners for providing their valuable time for this study.

References

[1] S. Chrisopoulos, J. Harford, A. Ellershaw, Oral Health and Dental Care in Australia: Key Facts and Figures 2015. Cat. No. DEN 229, AIHW, Canberra, 2016.
[2] P.M. Leong, M.G. Gussy, S.Y. Barrow, A. de Silva-Sanigorski, E. Waters, A systematic review of risk factors during first year of life for early childhood caries, Int. J. Paediatr. Dent. 23 (4) (2013) 235–250.
[3] M. Hooley, H. Skouteris, L. Millar, The relationship between childhood weight, dental caries and eating practices in children aged 4-8 years in Australia, 2004-2008, Pediatr. Obes. 7 (6) (2012) 461–470.
[4] M. Gussy, R. Ashbolt, L. Carpenter, M. Virgo-Milton, H. Calache, S. Dashper, P. Leong, A. de Silva, A. de Livera, J. Simpson, E. Waters, Natural history of dental caries in very young Australian children, Int. J. Paediatr. Dent. 26 (3) (2016) 173–183.
[5] S. Twetman, M. Fontana, J.D. Featherstone, Risk assessment - can we achieve consensus? Community Dent, Oral Epidemiol. 41 (1) (2013) e64–70.
[6] B. Christian, R. Armstrong, H. Calache, L. Carpenter, L. Gibbs, M. Gussy, A systematic review to assess the methodological quality of studies on measurement properties for caries risk assessment tools for young children, Int. J. Paediatr. Dent. 29 (2) (2019) 106–116.
[7] M. Tellez, J. Gomez, I. Pretty, R. Ellwood, A.I. Ismail, Evidence on existing caries risk assessment systems: are they predictive of future caries? Community Dent, Oral Epidemiol. 41 (1) (2013) 67–78.
[8] AAPD, Guideline on caries-risk assessment and management for infants, children, and adolescents, Pediatr. Dent. 35 (5) (2014) E157–164.
[9] D. Bratthall, G.H. Petersson, Cariogram–a multifactorial risk assessment model for a multifactorial disease, Community Dent, Oral Epidemiol. 33 (4) (2005) 256–264.
[10] J.D. Featherstone, S.M. Adair, M.H. Anderson, R.J. Berkowitz, W.F. Bird, J.J. Crall, P.K. Den Besten, K.J. Donly, P. Glassman, P. Milgrom, J.R. Roth, R. Snow, R.E. Stewart, Caries management by risk assessment: consensus statement, April 2002, J. Calif. Dent. Assoc. 31 (3) (2003) 257–269.
[11] L. Mokkink, C. Terwee, D. Patrick, J. Alonso, P. Stratford, D. Knol, L. Bouter, H. de Vet, COSMIN Checklist Manual, Amsterdam (2012).
[12] N. Tinanoff, R.J. Baez, C. Diaz Guillory, K.J. Donly, C.A. Feldens, C. McGrath, P. Phantumvanit, N.B. Pitts, W.K. Seow, N. Sharkov, Y. Songpaisan, S. Twetman,

Early childhood caries epidemiology, aetiology, risk assessment, societal burden, management, education, and policy: global perspective, Int. J. Paediatr. Dent. 29 (3) (2019) 238–248.
[13] S. Johnson, L. Carpenter, E. Ameerdroz, S. Dashper, M. Gussy, H. Calache, A.M. de Silva, E. Waters, Cohort Profile: The VicGeneration (VicGen) study: An Australian oral health birth cohort, Int. J. Epidemiol. 46 (1) (2017) 29–30.
[14] A.I. Ismail, W. Sohn, M. Tellez, A. Amaya, A. Sen, H. Hasson, N.B. Pitts, The International Caries Detection and Assessment System (ICDAS): an integrated system for measuring dental caries, Community Dent. Oral Epidemiol. 35 (2007) 170–178.
[15] B. Christian, E. Ameerdroz, H. Calache, M. Gussy, R. Sore, E. Waters, Examiner calibration in caries detection for populations and settings where in vivo calibration is not practical, Community Dent. Health 34 (4) (2017) 248–253.
[16] P.A. Harris, R. Taylor, B.L. Minor, V. Elliott, M. Fernandez, L. O'Neal, L. McLeod, G. Delacqua, F. Delacqua, J. Kirby, S.N. Duda, The REDCap consortium: building an international community of software platform partners, J. Biomed. Inform. 95 (2019) 103208.
[17] K.A. Hallgren, Computing inter-rater reliability for observational data: an overview and tutorial, Quant. Methods Psychol. 8 (1) (2012) 23–34.
[18] X. Gao, I. Di Wu, E.C. Lo, C.H. Chu, C.Y. Hsu, M.C. Wong, Validity of caries risk assessment programmes in preschool children, J. Dent. 41 (9) (2013) 787–795.
[19] R. Harris, A.D. Nicoll, P.M. Adair, C.M. Pine, Risk factors for dental caries in young children: a systematic review of the literature, Community Dent. Health 21 (1 Suppl) (2004) 71–85.
[20] B.W. Chaffee, J.D. Featherstone, S.A. Gansky, J. Cheng, L. Zhan, Caries risk assessment item importance: risk designation and caries status in children under age 6, JDR Clin. Trans. Res. 1 (2) (2016) 131–142.
[21] A. Senneby, I. Mejare, N.E. Sahlin, G. Svensater, M. Rohlin, Diagnostic accuracy of different caries risk assessment methods. A systematic review, J. Dent. 43 (12) (2015) 1385–1393.
[22] P.F. Whiting, A.W. Rutjes, M.E. Westwood, S. Mallett, J.J. Deeks, J.B. Reitsma, M.M. Leeflang, J.A. Sterne, P.M. Bossuyt, QUADAS-2: a revised tool for the quality assessment of diagnostic accuracy studies, Ann. Intern. 155 (8) (2011) 529–536.
[23] F.J. Ramos-Gomez, J. Crall, S.A. Gansky, R.L. Slayton, J.D. Featherstone, Caries risk assessment appropriate for the age 1 visit (infants and toddlers), J. Calif. Dent. Assoc. 35 (10) (2007) 687–702.
[24] J.D. Featherstone, The caries balance: the basis for caries management by risk assessment, Oral Health Prev. Dent. 2 (Suppl 1) (2004) 259–264.
[25] L. Gibbs, A.M. de Silva, B. Christian, L. Gold, M. Gussy, L. Moore, H. Calache, D. Young, E. Riggs, M. Tadic, R. Watt, I. Gondal, E. Waters, Child oral health in migrant families: a cross-sectional study of caries in 1-4 year old children from migrant backgrounds residing in Melbourne, Australia, Community Dent. Health 33 (2) (2016) 100–106.
[26] M. Gussy, G. Mnatzaganian, S. Dashper, L. Carpenter, H. Calache, H. Mitchell, E. Reynolds, L. Gibbs, S. Hegde, G. Adams, S. Johnson, E. Ameerdroz, B. Christian, Identifying predictors of early childhood caries among Australian children using sequential modelling: findings from the VicGen birth cohort study, J. Dent. 93 (2020) 103276.
[27] J.D. Beck, J.A. Weintraub, J.A. Disney, R.C. Graves, J.W. Stamm, L.M. Kaste, H.M. Bohannan, University of North Carolina Caries Risk Assessment Study: comparisons of high risk prediction, any risk prediction, and any risk etiologic models, Community Dent. Oral Epidemiol. 20 (6) (1992) 313–321.
[28] J.A. Disney, R.C. Graves, J.W. Stamm, H.M. Bohannan, J.R. Abernathy, D.D. Zack, The University of North Carolina Caries Risk Assessment study: further developments in caries risk prediction, Community Dent. Oral Epidemiol. 20 (2) (1992) 64–75.
[29] B.W. Chaffee, C.A. Feldens, P.H. Rodrigues, M.R. Vitolo, Feeding practices in infancy associated with caries incidence in early childhood, Community Dent. Oral Epidemiol. 43 (4) (2015) 338–348.
[30] C.A. Feldens, P.H. Rodrigues, G. de Anastacio, M.R. Vitolo, B.W. Chaffee, Feeding frequency in infancy and dental caries in childhood: a prospective cohort study, Int. Dent. J. 68 (2) (2018) 113–121.
[31] E. Ameerdroz, L. Carpenter, S. Johnson, V. Flood, S.G. Dashper, H. Calache, M. Gussy, E. Waters, Feasibility and development of a cariogenic diet scale for epidemiological research, Int. J. Paediatr. Dent. 29 (3) (2019) 310–324.
[32] P. Arrow, Incidence and progression of approximal carious lesions among school children in Western Australia, Aust. Dent. J. 52 (3) (2007) 216–226.