Adolescent and Parent Decision-Making and Preferences in Hypodontia Care Using Discrete Choice Experiment

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The candidate confirms that the work submitted is her own, except where work which has formed part of jointly authored publications has been included. The contribution of the candidate and the other authors to this work has been explicitly indicated (Page IV). The candidate confirms that appropriate credit has been given within the thesis where reference has been made to the work of others.

Publications relating to the thesis

Barber, S.K., Bekker, H. L., Meads, D., Pavitt, S. and Khambay, B.S. 2018. Identification and appraisal of outcome measures used to evaluate hypodontia care: A systematic review. Am J Orthod Dentofacial Orthop. 153(2), 184-194 e118.

Barber, S.K., Pavitt, S., Meads, D., Khambay, B.S. and Bekker, H.L. 2018. Assessment of information resources for people with hypodontia. BDJ Open. 4, 18001.

Barber, S.K., Lam, Y., Pavitt, S. and Hodge, T.M. 2018 Is social media the way to empower people to share their experiences of dental care? JADA. 149(6), 451-459.

Barber, S.K., Pavitt, S., Khambay, B.S., Bekker, H.L. and Meads, D. 2018. Eliciting Preferences in Dentistry with Multiattribute Stated Preference Methods: A Systematic Review. JDR Clinical and Translational Research. 3(4), 326-335.

Barber, S.K, Bekker, H.L, Marti, J., Pavitt, S., Khambay, B.S and Meads, D. 2019. Development of a Discrete Choice Experiment (DCE) to Elicit Adolescent-Parent Preferences for Hypodontia Treatment. The Patient: Patient-Centred Outcomes Research. 12(1), 137-148

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Work attributed to Sophy Barber

- Design, data collection, data analysis and interpretation, draft and revision of manuscript

Contribution of co-authors

- Hilary Bekker, Balvinder Khambay, David Meads, Sue Pavitt (Supervisors): Input into study design and interpretation, revision of manuscripts
- Joachim Marti: Input into DCE study design, analysis and interpretation
- Yung Lam: Data collection, data analysis and interpretation, revision of manuscript
- Trevor Hodge: Input into study design and interpretation, revision of manuscript

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Abstract

Hypodontia is the failure of developmental of one or more teeth, resulting in their permanent absence. Different treatment options exist and decision-making requires collaboration between adolescents, parents and the dental team. Currently there is little evidence about decision-making practice and patient preferences for care.

Aim: To examine adolescent and parent decision-making and preferences in hypodontia

Design: Mixed methods

Population & setting: Adolescents (aged 12-16 years) with hypodontia and their parents in NHS Hospitals in England and Wales

Methods:

Stage One: Examination of hypodontia care using mixed methods (systematic review of hypodontia literature, analysis of patient information resources and social media, observation of clinical consultations, interviews with adolescents and parents) to establish current decision-making practice.

Stage Two: Development of a preference elicitation instrument for adolescents and parents, using Discrete Choice Experiment (DCE) tasks to examine decision-making processes and elicit preferences. Piloting used ‘Think aloud’ methods and verbal probing to test the validity and acceptability of the tool and methods.

Stage Three: Preference elicitation survey with two approaches: 1) Online survey of adolescents and parents 2) Face-to-face survey completion by adolescent-parent dyads under observation to examine joint decision-making.

Results: Adolescent and parent understanding of hypodontia and its treatment is limited. The current pathway does not promote collaborative decision-making and adolescent and parent participation in care decisions is low. Appearance after treatment, severe dental problems, severe discomfort during treatment and a long treatment time are important to decision-making. Individuals show important heterogeneity in preferences. Trade-offs tasks appeared to help some adolescents and parents think about care and understand what is important to them.

Conclusions: Hypodontia care decisions require a collaborative approach, with appropriate information exchange about risks, benefits and outcomes with inclusion of patient preferences. Trade-off tasks might be useful in future decision support tools to help adolescents and parents think about treatment.

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Abbreviations

BWS	Best Worst Scaling
CL	Conditional logit
DCE	Discrete Choice Experiment
GDC	General Dental Council
GDP	General Dental Practitioner
IPDAS	International Patient Decision Aids Standards Collaboration
LDI	Leeds Dental Institute
MLIA	Maxillary lateral incisor agenesis
MSPA	Mandibular second premolar agenesis
NIHR	National Institute of Health Research
NHS	National Health Service
OHRQoL	Oral health-related quality of life
OSC	Orthodontic space closure
OSO	Orthodontic space opening
PDA	Patient Decision Aid
PIC	Participant identification centre
PIR	Patient information resource
PPI	Patient and public involvement
RBB	Resin-bonded bridge
RUM	Random Utility Model
SDM	Shared-decision making
SLH	St Luke’s Hospital, Bradford
SP	Stated preference
VCM	Value clarification method

Chapter One:

Introduction to the Research

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Chapter One provides an introduction to the thesis, which examines adolescent and parent decision-making and preferences for a specific developmental dental condition. The research problem is described to provide background to the thesis and the rationale for the research. The aims and ambition of the research are given to explain how this work will contribute to driving future changes in decision-making in clinical practice. The role of patient and public contributors in the research design and conduct are outlined. The chapter summarises the ethical considerations and sources of funding.

1.1 The research problem

Hypodontia is a developmental dental condition that causes failure of development of one or more teeth, resulting in their permanent absence (Larmour et al., 2005). It is one of the most common developmental dental anomalies, estimated to affect the permanent dentition in approximately 4-6% of people (Khalaf et al., 2014, Polder et al., 2004). Hypodontia is associated with reduced oral health-related quality of life due to concerns about appearance, function and impact on daily activities (Meaney et al., 2012, Akram et al., 2011, Anweigi et al., 2013a, Hvaring et al., 2014a, Hashem et al., 2013, Wong et al., 2006b, Locker et al., 2010, Laing et al., 2010), with dental appearance cited as a key motivator for seeking treatment (Hobkirk et al., 1994). Functional difficulties may present as impacts on eating (avoidance of certain foods, inability to chew, food getting stuck between teeth) and speech (particularly lisping with 's' sounds) (Meaney et al., 2012, Akram et al., 2011, Anweigi et al., 2013a, Hvaring et al., 2014a, Hashem et al., 2013, Wong et al., 2006b, Locker et al., 2010, Laing et al., 2010).

Diagnosis of hypodontia can occur at any age, but tooth absence is commonly identified in adolescence when the tooth or teeth fail to erupt (Gill and Barker, 2015). The life-long and changing nature of the condition means treatment can continue throughout life with significant impact on the person and family (Stevenson et al., 2013). Management of hypodontia in the permanent dentition often involves interdisciplinary treatment from different dental specialities (Gill and Barker, 2015, Lewis et al., 2010). The suitability of different treatment options depends on the individual’s clinical presentation and preferences; often there is no single ‘best’ treatment. Treatment choice requires collaborative decision-making between adolescents, parents and the dental team, reflecting the shared decision-making (SDM) approach advocated in the National Health Service (NHS) (NHS England, 2019). In SDM patients and professionals are equal partners in the decision-making process, recognising that patients and families have expertise in their values and preferences, motivation for treatment, ability to accept treatment and ability to comply with self-care (Coulter and Collins, 2011, Da Silva, 2012).

Despite the complexity of decision-making for adolescents with hypodontia and their families, to date there has been little research to examine this process from a patient and parent perspective. Much of the existing literature discussing decisions about hypodontia treatment has been from dental professionals’ perspective with a focus on clinical factors (Zachrisson et al., 2011, Kokich et al., 2011). Studies capturing patient perspective are limited to two studies examining adolescent ratings of the outcome of simulated treatment for lateral incisor agenesis (Rayner et al., 2015, Barber et al., 2015) and qualitative studies examining young adults experience of hypodontia treatment (Meaney et al., 2012, O'Keeffe et al., 2016). The present study aimed to understand current clinical decision-making processes and information, identify what is important to adolescents and parents using quantitative and qualitative approaches and synthesise this information to provide recommendations for improving patient-centred decision-making in hypodontia. Examining current practice in hypodontia care addresses gaps in knowledge about the approach to decisions by adolescents, parents and the dental team, and any barriers to a collaborative model of decision-making. The adolescent-parent perspective has been somewhat overlooked in dental research despite the clinical situation often requiring adolescents, parents and the dental team to make decisions together.

1.2 Purpose of the research

Understanding current decision-making practice and preferences for dental care is essential for improving the quality and delivery of care. Incorporating patient preferences into routine care is an NHS priority (NHS, 2010), yet to date there has been little research to understand adolescents’ and parents’ preferences and how these can be effectively integrated into dental care. This research aimed to explore decision-making, particularly preferences, from an adolescent and parent perspective. This perspective was selected to increase the relevance and applicability of findings for improving patient-centred care, by exploring the experiences of adolescents and their families.

The first part of the research examined current hypodontia practice to identify undesirable variation in practice and areas for improvement (NICE, 2017). This is an essential step in the development and implementation of tools and care pathways to improve treatment experience, outcomes and health service delivery (Campbell et al., 1998). Within hypodontia care, information gained about adolescent and parent decision-making and preferences can inform changes to improve the decision-making process, particularly information exchange, promoting greater patient engagement and inclusion of patient and family preferences in decisions. More broadly across dentistry, the methods used in this research can potentially be transferred to other dental conditions and areas of dental practice to similarly examine decision-making practice.

The latter part of the research used a preference elicitation method to measure adolescent and parent preferences for hypodontia care and to explore the decision-making process between adolescents and parents. The preference elicitation method chosen for this study, Discrete Choice Experiment (DCE), is a stated preference approach that requires respondents to choose between hypothetical scenarios by trading off the treatment components (attributes) presented in the scenarios. Analysis of choice behaviour allows estimation of preferences for the treatment attributes. This approach has been widely used in healthcare to measure a range of different stakeholder perspectives (Clark et al., 2014, de Bekker-Grob et al., 2012, Soekhai et al., 2019); however, a review in dentistry undertaken in preparation for this research found only 12 studies employing DCE (Barber et al., 2017). All studies in dentistry involved adult participants and none examined preferences for interdisciplinary conditions such as hypodontia.

This research was innovative in using DCE to elicit preferences from adolescents individually and with a parent to explore adolescent-parent dyads' joint decision-making processes. The approach provided valuable information about individual and dyadic preferences and the underlying mechanics for joint decision-making to inform future development and testing of decision support interventions. The application of DCE methods with dyads under observation provided rich information that, alongside the methods incorporated to evaluate the validity of the DCE, provided methodological information for future preference elicitation research.

The wider ambition of the research is to provide preliminary information to promote changes in clinical practice and future research that improve clinical communication and support a collaborative approach to decision-making. Collaborative decision-making is underpinned by strong ethical and efficacy arguments (Legare and Thompson-Leduc, 2014) and despite the challenges to implement, it is emerging as the zeitgeist of modern healthcare. Research to support integration of collaborative decision-making effectively into routine practice is essential. The priority setting partnership by the James Lind Alliance (2018) placed research to improve communication between dental teams and patients and carers in the top ten oral and dental health priorities.

1.3 Aims and Objectives

The aim of the research was to examine adolescent and parent decision-making and preferences in hypodontia care.

The research objectives were:

- To describe current decision-making practices in hypodontia care including a) the adequacy of the literature for supporting evidence-based information provision; b) quality of patient information resources for patient preparation and involvement in decision-making; c) the characteristics and suitability of the consultation processes; and d) adolescent-parent experience of decision-making
- To develop a preference elicitation instrument for use with adolescents, parents and adolescent-parent dyads using Discrete Choice Experiment (DCE) methods
- To investigate decision-making processes and preferences for adolescents, parents and adolescent-parent dyads for hypodontia care
- To evaluate the suitability of the DCE method for exploring decision-making and eliciting adolescent and joint adolescent-parent preferences
- To synthesise the evidence from the research stages to 1) provide recommendations for improving patient-centred decision-making in hypodontia and more widely across dentistry, and 2) identify future research priorities that will support improvements to communication and decision-making between adolescents, parents and the dental team

1.4 Research perspective

The research explored decision-making, and particularly preferences, for dental care from the perspective of adolescents with hypodontia and their parents. For the purpose of this research, the term *adolescent* was used to refer to adolescents aged 12-16 years and the term *parent* for anyone with parental or carer responsibilities.

The adolescent age group was chosen to reflect the age when most decisions about definitive treatment for hypodontia are made. This is an important but challenging age clinically, complicated by the emotional, psychological and legal transition from child (dependent) to legal adult (independent) (Zaky, 2016). A range of severities and presentations of hypodontia were included to assess the impact of severity of hypodontia on preferences and decision-making. Dental experience has been found to influence dental attendance (Badri et al., 2014) and willingness of patients to engage in treatment decisions and delivery (Sbaraini et al., 2012) so adolescents at different stages of treatment were included.

Best practice promotes the inclusion of parents in healthcare decisions where possible and appropriate, regardless of the child’s age and capacity (National Health Service, 2016). Clinical experience suggests parents have an important facilitation and advocacy role in decision-making; the research aimed to examine this further to provide evidence to support clinical approaches to joint decision-making between adolescents, parents and the dental team.

1.5 Setting

The research was set in the context of dental care delivery in the National Health Service (NHS) in England and Wales. Secondary care settings were selected based on commissioning guidelines, which indicate interdisciplinary conditions are often best treated within secondary care teams involving different dental specialists (NHS England, 2015, NHS England, 2018). It is acknowledged that a proportion of people with hypodontia will be treated in primary care and the experiences and preferences of this group are not represented in this research.

Methods involving adolescent and parent participants recruited from three secondary care sites in Yorkshire, England, which were chosen to provide a diverse population. Leeds Dental Institute (LDI) is run as a partnership between Leeds Teaching Hospitals NHS Trust and the University of Leeds, providing clinical care, undergraduate and postgraduate training and research facilities. LDI provides a full range of specialist dental care to people with complex dental needs based on referral from primary or secondary care dental practitioners in Leeds and the surrounding areas. Leeds is the fourth largest city in England, with an estimated population of 780,000. St. Luke's Hospital (SLH) is a district general hospital in the Bradford Teaching Hospitals Foundation Trust, which serves a population of approximately 500,000. York Hospital is a district general hospital providing care for York and the surrounding North Yorkshire rural area serving a similar sized population but from a larger geographical area.

Population health data indicates Leeds and Bradford both have reduced life expectancy and increased mortality rates compared to the average for England, and score worse on measures of determinants of health, such as low-income families, child educational attainment and violent crime rates. Bradford has a low employment rate (68%) compared to the England average (75.2%) and Leeds (76.8%) (Public Health England, 2017/2018). In contrast, York has higher than average health values and scores favourably in determinants of health.

The latter part of the research included an online survey with patients recruited from NHS hospitals across England and Wales. A wide-reaching invitation was sent to secondary care hospitals units to request participation as a recruitment site. The selection and location of sites is reported in full in Chapter 5. While the inclusion of sites outside Yorkshire increased the representativeness of the sample, it is acknowledged that not all geographical areas were included.

1.6 Overview of research methods

This cross-discipline research involved decision-making psychology, health economics and applied health research. The research was a mixed methods study conducted in distinct stages (Table 1.1). Prior to commencing the investigative aspects of the research, a literature review was performed to provide context to the study (Chapter 2). The clinical aspects of hypodontia were considered in terms of relevance to choosing treatment. Decision-making theory and the application to clinical practice was reviewed to establish current thinking in this field. The final part of the literature review considered methods for eliciting preferences in research, focusing on the method chosen for this research, Discrete Choice Experiment (DCE).

The first investigative stage of the research examined current decision-making practice in hypodontia care (Chapter 3). Complementary mixed methods were used to capture a variety of experiences and perspectives. The research commenced with a literature review to identify and appraise current evidence for hypodontia treatment methods and to map the outcomes used to evaluate care. Following this, patient information resources were examined including content analysis of written and online patient information resources and observation and analysis of clinical consultations. To obtain information about patient experience, interviews were conducted with adolescents with hypodontia and their parents. Finally, to provide an additional source of patient experience, a systematic search and analysis of social media posts was conducted. The findings were synthesised to provide an overall view of current practice and areas where improvements could be made.

The second investigative stage involved development of a preference elicitation instrument, which included questions to understand the decision-making context and Discrete Choice Experiment (DCE) choice tasks to elicit preferences (Chapter 4). DCEs allow examination of preferences by observing choices between alternatives presented in hypothetical scenarios (choice tasks). Preferences can be estimated by examining the trade-offs underpinning the selection of an alternative in the choice task. Best practice guidance and existing evidence was used to develop the instrument and particular effort was made to include the target population to increase the validity and relevance of the tool. The instrument and planned data collection methods were tested in a pilot study using cognitive interviewing (think aloud methods with some verbal probing) alongside preference data analysis. The pilot informed changes to improve the validity and acceptability of the preference elicitation instrument and method.

The third investigative stage in the research applied the preference elicitation instrument in a cross-sectional survey using two different data collection methods (Chapter 5). The first approach used an online survey to elicit preferences from a larger number of adolescents, parents and adolescent-parent dyads. In the second method, adolescent-parent dyads were invited to complete the preference survey separately and together under observation to allow a more detailed examination of decision-making behaviour and preferences. A range of validity measures were included to evaluate the preference elicitation method and its suitability for adolescents and dyads.

The final part of the research involved synthesis of the findings from the three data collection stages to identify the key research findings (Chapter 6). This information was used to develop key recommendations for improving decision-making in practice and guiding future research priorities.

Table 1.1: Research data collection stages and methods

Stage	Aim	Methods
Overview of research context	To establish current knowledge and evidence about hypodontia, decision-making and preferences	• Literature review
Examination of hypodontia care	To establish current decision-making practices in hypodontia care	• Identification and synthesis of existing data • Semi-structured interviews • Naturalistic observation
Preference elicitation instrument development & testing	To develop a preference elicitation instrument for adolescents and adolescent-parent dyads To test the suitability of the instrument and data collection methods	• Quantitative survey methods • Stakeholder consultation • Use of best practice guidelines and research evidence • Cognitive interviewing – Think aloud with some verbal probing • Analysis of preference data
Cross-sectional preference elicitation survey	To measure adolescent and parent preferences for hypodontia care To explore adolescent and parent decision-making processes	• UK-wide online survey • Survey in interview setting with observation • Assessment of reliability and validity of DCE
Synthesis and recommendations	To synthesise information from the investigative stages to provide recommendations	• Synthesis and critique of research findings

1.7 Key Stakeholder Involvement

Patient and public involvement (PPI) in research is the conduct of research ‘with’ rather than ‘about’ the public, including patients, carers and other contributors (Hayes et al., 2012). PPI promotes inclusion of patients and lay members as experts with lived experience, to contribute to design, conduct and dissemination of research as advisors and partners. Roles can include being a member of a steering committee or stakeholder group, acting as a research advisor or co-production of information resources and being involved in consultation, collaboration and user-led approaches.

In this research PPI contributors were identified to provide the important lived experience of the patient and family, alongside a public perspective to the research. This included representation of the target group (adolescents with hypodontia and their parents) as well as adolescents without hypodontia, who provided a lay adolescent perspective.

Currently there is no official patient support group or charitable body for people with hypodontia and apart from clinical care, the avenues for identifying people with hypodontia were limited. As a result, adolescents with hypodontia and their parents were largely identified through routine clinical activity. The potential power imbalance between dentist and patient was acknowledged and managed with the advice of a supervisor with extensive experience in PPI (Professor Pavitt). Professor Pavitt provided training and support throughout the research and acted as a point of contact for all adolescent and parent contributors to discuss any queries or concerns regarding the research.

During routine appointments and where it was judged to be appropriate, the research was discussed. Adolescents and parents who expressed interest were informed about the opportunity to be involved as a contributor, the purpose of PPI and brief written information about their potential role was provided for consideration. Contact details for the researcher were provided and the potential collaborator was invited to contact if they wished to be involved. Contributors were invited to take part by email, telephone or individual or group meetings depending on preference. The approach and timescale for feedback, methods for communication, arrangements for meetings and reimbursement for costs was agreed with individuals following best practice guidance (Hayes et al., 2012, Mathie, 2016). Travel and parking costs were covered for all meetings and healthy refreshments were provided. For adolescents, vouchers were provided to recognise their time contribution. No monetary payments were made to PPI contributors.

Due to the length of the research and the likely change in adolescent's circumstance as they progressed through the education system, it was expected that many would not be able to contribute to the full research project. Contributors were informed that there was no obligation to commit indefinitely and a contingency plan was developed for further recruitment if needed. Over the research period five adolescent-parent dyads contributed to various stages of the research (Table 1.2).

Table 1.2: Patient and public involvement in different stages of the research

Research component	Patient and Public involvement
Research development	Discussion of relevance of research question Discussion about most important population and consideration for inclusion of age range Proposed approach for recruitment Suitability of methods, particularly interviews and survey methods
Research conduct	Systematic review of hypodontia care: Defining concept of 'effectiveness', categorisation of outcomes measures Co-development of materials – Participant information sheets, consent and assent forms Co-design of questionnaires Stakeholder consultation for attribute selection
Analysis	Review of study findings and interpretation Perception of importance of findings
Research dissemination	Co-development of lay summary Input into public engagement strategy for sharing results

To gain an alternative perspective, seven adolescents without hypodontia and with little experience of dental care were recruited from the University of Leeds School of Dentistry the SMILE AIDER PPIE Forum (Stakeholder Meaningful Involvement and Engagement Aiding Dental Research). The forum was established in 2015 and now has more than 20 core contributors and additional specialist sub-groups representing patients and carers across the dental specialties and members of the public. The adolescents invited to collaborate in this research provided 'seldom heard' voices of minority ethnic groups with high dental need and low literacy (Pavitt et al., 2018). The SMILE AIDER contributors participated in a group session during the planning stages to discuss the acceptability of the proposed research and again during preference elicitation instrument development. A further session is planned to inform the dissemination strategy. To recognise their contribution, group meetings were incorporated into a half day visit to the dental school where the contributors were able to participate in widening participation activities to learn more about careers in ‘STEM’ (science, technology, engineering and mathematics) and dentistry.

In addition to PPI contributors, dental specialists with experience in treating hypodontia and managing care pathways were invited to contribute. Clinicians involved in the delivery of hypodontia care at Leeds Dental Institute and St Luke’s Hospital provided valuable feedback on the research design, methods and interpretation of findings.

1.8 Ethical considerations

The study was adopted by the NIHR Clinical Research Network and conducted in accordance with the ethical principles outlined in the 1964 Declaration of Helsinki (World Medical Association, 1964) and its later amendments or comparable ethical standards. The Good Clinical Practice (GCP) training was completed prior to commencement of the study and an GCP e-learning refresher course and the e-learning module covering Informed Consent in Paediatric Research course (National Insititute of Health Research, 2017) was completed during the research period.

1.8.1 Adolescents as research participants

Adolescents, defined as young people aged 12-16 years, were the focus of the research so specific considerations were given to their ethical involvement. Recruitment was designed to ensure there were opportunities for adolescents to decide whether they wanted to participate away from the clinical environment to reduce the risk of coercion. Participant information sheets for each study component were co-designed with patient contributors to make the language age-appropriate and jargon-free (National Insititute of Health Research, 2017). Procedures, risk and benefits of participation, confidentiality, anonymity and the opportunity to withdraw were described in child-friendly terms. Copies of information sheets are available on request.

For the purpose of research, anyone under the age of 16 years is considered to be a minor (HM Government UK, 2004) and consent is required from someone with parental responsibility (HM Government UK, 2018). In this study parental consent and participant assent were required prior to enrolment for adolescents aged 12-16 years. For those aged 16 years, consent was required from the participant, and parental consent was also sought to reduce the risk of any conflict in wishes. Looked after children were invited to participate in the research in the same way as children living with parents and best practice guidance was followed for obtaining consent (National Insititute of Health Research, 2017).

Incentives were offered to participants for some components of the research to encourage participation and recognise participant's contribution and time commitment. A flat-rate reimbursement was offered to those attending for additional appointments to compensate for travel and parking costs (Health Research Authority, 2014). There is no consensus on appropriate levels for compensation and incentives so advice from patient and parent contributors was sought. The selected value of incentives aimed to reflect the level of burden and encourage participation while being non-coercive.

The main potential harms to adolescents and parents participating in the study were distress relating to hypodontia or dental treatment, distress caused by the research itself, and identification of poor practice, personal or social issues. To manage participant distress, sources of support were given in patient information sheets including contact details for the researcher and reassurance that the researcher could provide advice and further support. At the end of face-to-face data collection, participants were offered an opportunity to ask questions and raise any concerns. A framework for escalating concerns was agreed for any situations where the researcher was unable to provide adequate support. For any concerns relating to the conduct of the research an independent point of contact was given.

For possible situations involving evidence of poor practice or personal or social issues, a senior clinical colleague (Nadine Houghton, Consultant Orthodontist) was available to provide advice. Trust policies for whistle-blowing and safeguarding were referenced in the site file.

1.8.2 Data management

Data was collected and processed in accordance with the University of Leeds Data Protection Code of Practice (University of Leeds, 2018) and General Data Protection Regulations (GDPR) (General Data Protection Regulation, 2016). All research data was stored anonymously and securely on a University of Leeds hard drive. Personal data required to track participants was separated from the research data immediately and only matched to the research data via a unique participant number available. Consent forms with participant name and signature were stored in a locked filing cabinet in the researchers' office in the University of Leeds School of Dentistry with restricted access. Data was only stored for the duration of the research and processed for the purpose of the research for which explicit consent had been obtained. Online survey data was collected and managed by UK-based companies who comply with GDPR. To access data, a password known only by the researcher was required.

Transcription of audio-recordings was undertaken by the researcher or a University of Leeds School of Dentistry secretary bound by the University of Leeds code of conduct. Audio-recordings and transcriptions were labelled only with the study identifier, stored on the University of Leeds secure drive and were only accessible to the researcher and secretary.

1.8.3 Ethical approval

NHS ethical approval was obtained for all study components involving NHS patients (Table 1.3). An overarching NHS approval was granted for the study in October 2015, then this was amended in 2018 based on the findings from the pilot. Additional ethical approval was sought from the University of Leeds Dental Research Ethics Council for two surveys that were planned during the research period which did not involve NHS patients (the survey to dentists to identify patient information resources and the attribute rating/ranking survey). Copies of the full ethical applications are available on request.

Table 1.3: Research Ethic Committee approvals

Study component	Ethics body	Approval
▪ Adolescent-parent interviews ▪ Clinical observations ▪ Pilot study ▪ Preference elicitation survey	North West Lancaster Ethic Research Committee (REC) (IRAS 174789)	15th October 2015 (Reference 15/NW/0804)
▪ Preference elicitation instrument ▪ Preference elicitation survey	HRA / HRCW North West Lancaster REC	11th May 2018 12th April 2018
Questionnaire to NHS dentists regarding patient information resources	University of Leeds Dental Research Ethics Committee	12th April 2016 (Reference 050216/SB/188)
Recruitment of participants from a Facebook support group for people with hypodontia to complete an online survey	University of Leeds Dental Research Ethics Committee	9th Nov. 2017 (Reference 040917/SB/235)

1.9 Funding

The research was funded through an NIHR Doctoral Research Fellowship (2015-08-052) awarded after a competitive peer-reviewed application process. Funding supported research and training costs but the research is independent. NIHR had no involvement in the design and conduct of the research and the interpretation of research data. The thesis and associated publications represent the view of the authors and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care.

Chapter Two:

Overview of hypodontia care, decision-making and preference elicitation research

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Chapter Two provides an overview of the clinical aspects of hypodontia, the current management options and the impact of both the untreated condition and receiving treatment from a patient and parent perspective. This provides context for the research and identifies important aspects of hypodontia care for decision-making. The second part of the literature review considers decision-making theory and practice relevant to the clinical setting. The current care pathway for hypodontia and key decision points and decision-makers are discussed. To provide context for the research and inform the examination of decision-making practice in hypodontia care, existing theories supporting different approaches to individual and collaborative decision-making in clinical practice were reviewed. The final section of the literature review outlines some of the methods used in research to elicit preferences. The justification for using one specific type of stated preference elicitation method, Discrete Choice Experiment (DCE), is provided and the methodology outlined.

2.1 Hypodontia

Hypodontia, or tooth agenesis, is a life-long dental condition characterised by the failure of development of one or more primary or secondary teeth excluding the third molars resulting in permanent absence of the tooth or teeth (Larmour et al., 2005). Hypodontia is distinct from tooth absence for other reasons, such as loss due to trauma, pathology such as periodontal disease and caries, elective extraction to facilitate orthodontic treatment and iatrogenic loss.

2.1.1 Characteristics of hypodontia

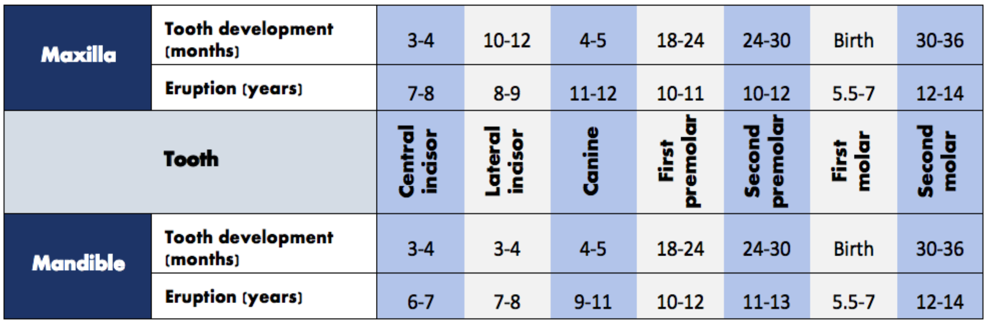
An arbitrary classification based on phenotypic presentation is widely used: severe hypodontia (oligodontia) for >6 missing teeth, moderate for 3-5 missing teeth and mild hypodontia for 1-2 missing teeth (Hobkirk et al., 2011). More than 80% of cases of hypodontia are mild, 15% are moderate and less that 5% are severe (Khalaf et al., 2014).

Hypodontia is one of the most common developmental dental anomalies, although reported prevalence varies within and between populations. A study of British school children found the prevalence of hypodontia to be 0.1-0.9% in the primary dentition and 3.5-6.5% in permanent dentition (Brook, 1974). Absence of primary teeth is associated with increased risk of tooth absence in the permanent dentition (Arte, 2004). A recent meta-analysis of epidemiological studies from 2002 onwards reported a mean prevalence of 6% with variation in distribution related to tooth type, ethnicity and gender, with a female tendency of 3:2 (Khalaf et al., 2014, Polder et al., 2004). The most commonly missing teeth are mandibular second premolars, where unilateral absence predominates, and maxillary lateral incisors, where bilateral absence is more common (Al-Ani et al., 2017).

Hypodontia has a polygenic multifactorial aetiology, influenced by both genetic and environmental factors, and can occur as an isolated trait or as part of a syndrome. Developmental anomalies, systemic disturbances such as infection, irradiation and medical treatment, and local factors such as pathology, physical and chemical trauma, have all been cited as aetiological factors in non-syndromic hypodontia (Brook, 1984, Arte, 2004). The genetic component has been linked to a number of candidate genes and can result in spontaneous or familial hypodontia (Cobourne, 2007). Syndromic hypodontia and oligodontia are associated with over 113 and 73 syndromes, respectively, according to the Online Mendelian Inheritance in Man catalogue of genetic disorders (OMIM, 2019). The associated features of these syndromes are highly variable and for many craniofacial syndromes treatment starts at birth and continues throughout life, with only a small part devoted to managing hypodontia. For this reason, people with syndromic hypodontia associated with marked systemic effects were excluded from the research.

Unless family history of hypodontia prompts early investigation, diagnosis tends to occur when abnormalities in the eruption of the teeth are identified. In the presence of clinical signs of hypodontia, radiographic examination is justified and this provides a definitive diagnosis (Isaacson et al., 2008). The age when a particular tooth can be accurately diagnosed as absent depends on normal average eruption dates (American Academy of Paediatric Dentistry, 2003) (Figure 2.1). Earlier diagnosis may be made if the developing tooth germ is absent on a radiograph, although hypodontia is associated with variable generalised delay in dental development (Tunc et al., 2011, Uslenghi et al., 2006).

Figure 2.1: Approximate age of dental development (figure adapted from the American Academy of Paediatric Dentistry, 2003)



Maxilla	**Tooth development (months)**	3-4	10-12	4-5	18-24	24-30	Birth	30-36
	Eruption (years)	7-8	8-9	11-12	10-11	10-12	5.5-7	12-14
Tooth		**Central incisor**	**Lateral incisor**	**Canine**	**First premolar**	**Second premolar**	**First molar**	**Second molar**
Mandible	**Tooth development (months)**	3-4	3-4	4-5	18-24	24-30	Birth	30-36
	Eruption (years)	6-7	7-8	9-11	10-12	11-13	5.5-7	12-14

Hypodontia can impact on appearance, function, daily activities and through this, oral health-related quality of life (Meaney et al., 2012, Akram et al., 2011, Anweigi et al., 2013a, Hvaring et al., 2014a, Hashem et al., 2013, Wong et al., 2006b, Locker et al., 2010, Laing et al., 2010) (Appendix Table 1). Dental appearance is cited as a major concern for people with hypodontia and a key motivator for seeking treatment (Hobkirk et al., 1994). The most common clinical feature of hypodontia is spacing in the dental arches, which may be reduced if primary teeth are retained in the place of absent permanent teeth. Hypodontia is associated with increased frequency of other dental anomalies (Garib et al., 2010, Pinho et al., 2009, Baccetti, 1998) and these, alongside hypodontia-related spacing, can lead to malpositioned and abnormal shaped teeth, uneven bone and gingival contour (Morgan and Howe, 2003a) and an over-closed appearance (Gahan et al., 2010). Qualitative research suggests increasing age causes greater awareness of dental appearance and modification in behaviour to conceal teeth (Meaney et al., 2012). Functional difficulties may present as impacts on eating (avoidance of certain foods, inability to chew, food getting stuck between teeth) and speech effects (particularly lisping with ‘s’ sounds) (Meaney et al., 2012, Akram et al., 2011, Anweigi et al., 2013a, Hashem et al., 2013, Wong et al., 2006b, Locker et al., 2010, Laing et al., 2010, Hvaring et al., 2014b). Increasing age has been found to correlate positively with greater functional limitation and physical disability (Anweigi et al., 2013a), possibly as a result of losing retained primary teeth (Laing et al., 2010).

2.1.2 Management of hypodontia

Treatment for hypodontia aims to address the issues associated with untreated hypodontia while promoting long-term dental health and minimising the long-term burden of care (Table 2.1). Other dental anomalies commonly associated with hypodontia, such as hypoplastic enamel, ectopic maxillary canine tooth position (Brin et al., 1986), microdontia (Brook et al., 2009), conical teeth, transpositions (Shapira and Kuftinec, 2001), supernumerary teeth (Varela et al., 2009) and infra-occluded primary teeth, may require additional interventions that are outside the scope of this thesis.

Table 2.1: Aims of hypodontia treatment

Holistic treatment	Manage all aspects of malocclusion Provide a positive experience of dental care
Maintain dental health	Minimise the risk of harm during treatment Support health-promoting behaviour
Improve dental appearance	Improve appearance of teeth & supporting structures Improve confidence related to teeth
Improve function	Improve ability to bite / chew Reduce food packing between teeth Reduce speech problems e.g. lisping
Improve oral health-related quality of life	Reduce impact on every day activities Improve psychological wellbeing Improve social wellbeing
Promote long-term health	Minimise the risk of future harm and complications Provide treatment that enables health-promoting behaviour
Minimise long-term burden	Minimise anxiety about the future Provide long-lasting treatment Ensure alternative treatment options in case of treatment failure Minimise future costs

Hypodontia is a developmental anomaly rather than a pathological process and people without functional or aesthetic concerns may feel there is little to be gained from intervention. In some cases, poor dental health or medical contraindications to dental treatment may preclude treatment (Morgan and Howe, 2003a, Morgan and Howe, 2003b). Provided the patient has sufficient information and support to make this choice and is doing so freely, they have the right to refuse treatment (Kleinman, 1991); however, it is important that there is opportunity to reassess the treatment decision (Rosen et al., 2005), as there is evidence that understanding and expectations change (Meaney et al., 2012). Furthermore, loss of primary teeth may increase the impact of hypodontia and cause new concerns to arise (Laing et al., 2010, Wong et al., 2006b). Where no active intervention is chosen, self-care may be required alongside support to enable adaptation to, and acceptance of, the condition (Schwartz, 2009).

Care in the primary dentition (up to 6 years old) aims to optimise dental health and development. Information provision and counselling can help to alleviate anxiety about future treatment and prepare children and family for future treatment (Gill et al., 2008). Patients and families may be offered additional preventative care and monitoring but active intervention is often discouraged, as early treatment can impact on ability to maintain oral health and increase the overall treatment burden for the patient and family (Nunn et al., 2003). For children with severe hypodontia, parents may request tooth replacement with removable prostheses to improve appearance and speech.

Hypodontia is frequently diagnosed in the mixed dentition (aged 6-12 years) when teeth fail to erupt. Referral at this time may be advised (British Orthodontic Society, 2010) but treatment decisions are often delayed until all the permanent present teeth have erupted. This allows more precise assessment of dental problems and treatment options and minimises delays between treatment phases. During the waiting period care aims to optimise dental health and development to maximise future treatment options (Nunn et al., 2003). Interim treatments may be considered for patients with concerns about dental appearance or function but this can increase overall treatment time and burden (Table 2.2).

Table 2.2: Interim treatments for hypodontia

Treatment	Purpose
Restoration of primary teeth	Improve size and shape of teeth Restore function in primary molars with mild-moderate infra-occlusion
Extraction of primary teeth	Encourage tooth eruption to promote space closure in edentulous sites
Interceptive orthodontics	Improve tooth position and inter-arch relationship
Removable / fixed prosthesis	Replace missing tooth/teeth to improve appearance and function

Comprehensive treatment tends to begin in adolescence once all permanent teeth have erupted. The ideal timing of treatment depends on the need for dental implants, as implants can only be provided once facial growth has slowed, usually 17 years old at the earliest (Morgan and Howe, 2004). Where implant treatment is planned it may be preferable to delay the start of preparatory treatment to reduce the time between treatment phases; however, people with hypodontia report waiting for treatment to start and delays during treatment contribute to dissatisfaction with care (Meaney et al., 2012). This can create a conflict; early treatment increases the likelihood of quiescent periods between treatment stages, which are perceived by patients to be delays, while postponing the start of treatment can cause frustration.

2.1.3 Comprehensive treatment strategies

The treatment strategies for comprehensive management of hypodontia are described below. These methods may be used in isolation or together in a multi-stage care plan.

- Management of retained primary teeth: maintenance or extraction
- Restorative camouflage of primary teeth or small or malformed permanent teeth
- Orthodontic treatment to alter tooth position to aid or eliminate tooth replacement
- Tooth replacement with removable, fixed or implant-supported prosthesis or tooth auto-transplantation

Primary teeth may be retained in agenesis sites and management is a choice between maintaining the tooth or extracting the tooth to facilitate spontaneous or active (orthodontic) closure of the space (Robinson and Chan, 2009b). A number of clinical factors influence the feasibility of whether a primary tooth can be maintained including number and position of missing teeth, the health and position of the primary tooth, general dental health and occlusal factors, such as crowding/spacing and inter-arch relationship. The age of the patient, amount of spacing, position of the teeth and the occlusion will determine whether the missing tooth space can be closed or maintained. Primary teeth often do not have the expected lifespan of a permanent tooth (Hvaring et al., 2014b) and the eventual loss of the primary tooth has been emphasised as a cause for worry in people with hypodontia (Akram et al., 2011).

Restorative camouflage can be used to address small and abnormal shaped permanent teeth and retained primary teeth associated with hypodontia. Tooth-coloured adhesive composite dental material can be used to alter the shape, size or colour of teeth to improve appearance and reduce spacing in the dental arch (Robinson and Chan, 2009a). Restorative camouflage may be an interim or permanent treatment option, as the material does not damage the underlying dental tissue and can be modified during growth and active treatment.

Orthodontic treatment aims to alter tooth position to correct aspects of malocclusion. Tooth movement with orthodontic appliances can be used to eliminate spaces where teeth are missing or redistribute space to optimise tooth replacement. The feasibility of orthodontic tooth movement is influenced by dentofacial characteristics, the presentation of hypodontia, occlusal factors and suitability for orthodontic treatment. Orthodontic treatment varies in length and complexity, depending on the number and position of missing teeth and the aims of treatment.

In agenesis sites without primary teeth, where orthodontic space closure is not possible or desirable, the edentulous space may be accepted, or a tooth replacement method may be chosen. This includes:

- Removable prosthesis (denture)
- Tooth-borne prosthesis (bridge)
- Implant-supported prosthesis
- Tooth auto-transplantation

Removable prostheses (dentures) consist of acrylic teeth attached to acrylic or metal baseplates, which rest on the soft tissues or other teeth. Prosthesis design is versatile and can be used to replace one or multiple teeth as a temporary or permanent solution (Bishop et al., 2007). Dentures are relatively quick, easy and predictable and can be constructed to manage severe hypodontia and deficiencies in soft tissues and vertical proportions (Durey et al., 2014b). Oral rehabilitation with partial dentures in young patients was shown to improve oral function, phonetics, aesthetics and reduce social impairment (Filius et al., 2016). The main limitations of removable prostheses are gaining adequate retention, increasing the risk of plaque stagnation and psychological adaption to a removable appliance (Bishop et al., 2007). In adolescents, appliances may require replacement due to dissatisfaction with appearance, technical complications and oral changes leading to poor retention (Hobkirk et al., 1989).

Tooth-borne prostheses (bridges) are one or more ceramic teeth attached to adjacent supporting teeth. Bridge designs are variable and influence the impact on supporting teeth and surrounding dental tissues. The choice of bridge design depends on which tooth or teeth are absent, the size of the edentulous space, the occlusion, and the size and health of adjacent teeth (Bishop et al., 2007, Durey et al., 2014b). Minimally invasive resin-bonded bridges (RBB) are the most conservative type, attaching to the adjacent tooth with a metal wing that requires little preparation of the supporting tooth crown. RBB require a favourable bite and adequate good-quality enamel for bonding to prevent bond failure. Conventional bridges cover the full surface of the adjacent teeth providing greater retention but this requires reduction of the supporting tooth unless the supporting tooth is developmentally small. Crown preparation of the supporting tooth is associated with increased risk of damage to the tooth vitality and is usually only considered in teeth that are already heavily restored.

Dental implants can be used to retain fixed prostheses including single tooth crowns, bridges or dentures. Dental implants are biocompatible titanium screws that are placed with a surgical procedure and then osseo-integrate into the alveolar bone. Suitability for implant-retained prostheses depends on adequate alveolar bone levels and the proximity of adjacent structures. Adjunctive bone grafting may be required as a separate preparatory procedure where bone volume is inadequate (Durey et al., 2014a). Royal College of Surgeons guidelines for selecting appropriate patients for implant treatment are currently under review. The 2012 version indicates the role of dental implants in hypodontia and emphasises the requirement for a specialist team with competencies in both the surgical and prosthetic elements of implant care, but acknowledges local arrangements may determine provider options (Alani et al., 2012).

Tooth auto-transplantation is the controlled extraction of a tooth and replacement within another alveolar site within the same individual (Andreasen et al., 1990). This method is advocated where a suitable tooth is due to be extracted for orthodontic purposes and there is adequate space and bone in the recipient site. The maintenance of a viable periodontal ligament enables continued dento-alveolar development and scope for future orthodontic treatment, which is advantageous in growing individuals. Treatment involves minor oral surgery to transplant the tooth, and the orthodontic and restorative components can be burdensome. Success depends on careful case selection, clinical skill and a highly motivated patient (Monteiro et al., 2019).

2.1.4 Impact of treatment on patients and families

Clinical experience indicates patient experience of treatment and the impact of treatment on everyday life varies between individuals and treatment methods (Table 2.3). Impacts may be generic or treatment-specific, for example, orthodontic treatment has been shown to cause a decrease in oral health-related quality of life (OHRQoL) for adolescents during the treatment period due to negative effect on smiling, talking and eating (de

Oliveira and Sheiham, 2004) and impacts associated with oral symptoms, functional limitations and social wellbeing (Brosens et al., 2014). Attending appointments imposes substantial costs on the patient and family in terms of travel, childcare for siblings and time off work for accompanying parents and often the ideal time for treatment falls during an important period for education (Gill et al., 2008).

Table 2.3: Potential impacts from undergoing treatment for hypodontia
Psychosocial effect of treatment
Physical effect of treatment e.g. discomfort
Impact on function e.g. eating, speech
Effect on self-care e.g. more intense oral hygiene regime, dietary restrictions
Effect on behaviour e.g. restrictions on contact sports, use of mouth guard
Time needed for attending appointments – impact on work and school
Impact on family e.g. support, attendance at appointments, anxiety
Out-of-pocket costs of treatment
Indirect costs associated with treatment

A literature search failed to identify any UK-based economic evaluations that quantify the financial impact of long-term hypodontia treatment. In the UK, treatment provided in secondary or tertiary NHS care settings is free of charge at the point of delivery while in primary care in England and Wales, NHS treatment requires co-payment by the patient except for people under 18 years old and those with specific exemptions (NHS England, 2017). In most cases comprehensive hypodontia care commences before the age of 18 years, eliminating treatment costs for the main part of treatment. In more severe cases and where local services allow, long-term care may be provided in secondary care via re-referral from a GDP, but provision of long-term care is more variable and may have an associated cost burden. For those unable or unwilling to access NHS care, or for specialist services in primary care, private treatment may be sought. The cost of private treatment varies considerably based on provider type, availability and demand, and geographical location.

Risk of adverse events or side-effects depends on the type of treatment, individual risk factors and adherence to behaviour and self-care advice. Risk is quantified in terms of likelihood of occurrence, severity and long-term consequence but individual's understanding and perception of risk can be affected by the presentation of information

(Ahmed et al., 2012). Common risks associated with hypodontia treatments are summarised in Table 2.4. The evidence regarding risks for specific treatments is considered further in Chapter 3.

Table 2.4: Risks during hypodontia treatment

Treatment failure	Ineffective treatment Technical issues – failure of components or procedures Failure to achieve treatment aims Failure to complete treatment
Physical effects	Effect of procedure e.g. discomfort, bruising, swelling Iatrogenic damage to hard and soft dental tissues
Psychosocial effects	Impact on self-confidence, self-esteem, social interactions

The estimated rate of failure to complete orthodontic treatment is high despite this being identified as a priority area of dental care by adolescents (Vazquez Fde et al., 2015). Failure rates range from 19.5% (Roberts et al., 1994) to 26.2% (Fox and Chapple, 2004) for hospital treatment and 12.9-33.6% for treatment with a specific type of orthodontic appliance (O'Brien et al., 2003). It has been shown to be difficult to predict those who are likely to successfully complete treatment prior to commencing treatment (Mandall et al., 2008). Five-year data from a Scottish hypodontia clinic found that of the 83 patients included in the audit, alterations to planned treatment were reported for 23% of patients and treatment was abandoned in 16% (Shafi et al., 2008).

Poor attendance in dentistry has been linked to socio-demographic factors and parental behavioural beliefs and attitudes, as well as factors at the provider and system level (Badri et al., 2014). Failure to complete treatment may be associated with poor decision-making and selection of the ‘wrong’ treatment (Mulley et al., 2012). Other factors associated with adherence more broadly in healthcare include health literacy, understanding about treatment, communication by the health professional and trust, concern about risks and side-effects, experience of symptoms and motivation for treatment, access to resources, cultural beliefs and educational level (Devine et al., 2018).

Due to the potential for change in dental status over time, for example due to loss of primary teeth, and the age-limited nature of some treatments, for example dental implants, often treatment continues over the life course (Stevenson et al., 2013). Worry about future treatment has been identified as a source of concern for people with hypodontia (Meaney et al., 2012, Akram et al., 2011). Almost all dental treatment requires long-term maintenance to preserve health, function and appearance. The type of treatment will determine the extent and frequency of maintenance care required, for example, repair or replacement of dental work, periodontal care, long-term retainer wear to maintain corrected tooth position after orthodontics. There has been little research to examine the long-term impact of hypodontia or to quantify costs over the life course. Long-term complications may be related to a deterioration in dental appearance, functional issues, biological complications, technical complications and the psychosocial effects resulting from complications. Evidence for complications associated with hypodontia are considered further in Chapter 3.

2.1.5 Choosing to access hypodontia care

In hypodontia care, the first decision made by adolescents and their families is whether to access dental care. Availability and access to services and understanding and ability to negotiate the healthcare system may also be important drivers. Research across dentistry has identified a number of multifaceted and complex factors that determine access (Table 2.5) (Badri et al., 2014, Hill et al., 2013, Goettems et al., 2012, Bright et al., 2015, Curtis et al., 2007). The literature search failed to identify evidence about factors that determine access to dental care specifically for people with hypodontia.

The present research focused on adolescents and parents who have opted to access care. It is acknowledged that the group of people affected by hypodontia who do not present for treatment are not represented. A thorough search of the literature failed to find any evidence to estimate the number of people with hypodontia who do not seek care. The 2013 Child Health Dental Survey estimates 80% of 12- and 15-year-olds reported attending for regular dental check-ups (Tsakos et al., 2015). It is not possible to determine whether a similar proportion with hypodontia access regular care. Generally, inequity in dental health and access to NHS dental services has a greater impact on lower socio-economic groups (Appleby et al., 2017). Unmet orthodontic treatment need is estimated to be 37% in 12-year-olds and 20% in 15-year-olds, but this increases to 40% and 32% respectively for children in areas of deprivation, measured by proportion eligible for free school meals (Rolland et al., 2016). Again, it is not possible to determine whether this inequity is reflected in access to hypodontia care.

Table 2.5: Potential barriers to accessing dental care

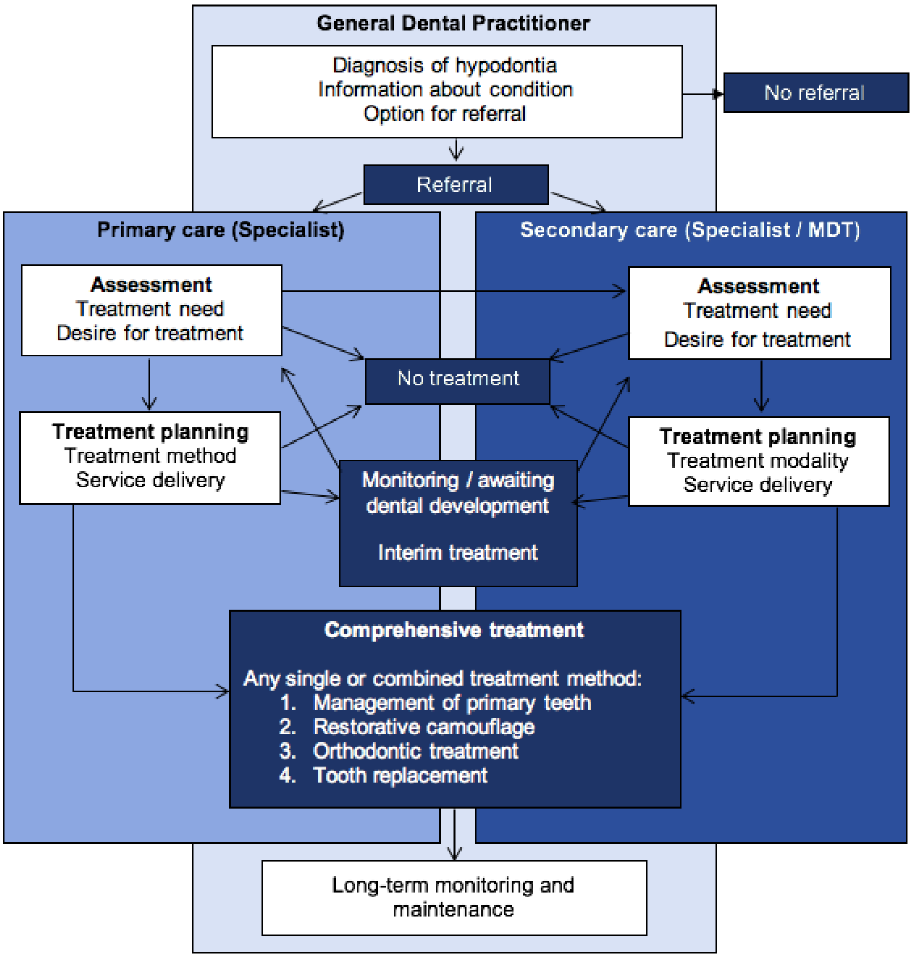
Patient factors	Perceived need for treatment Parents' education, working status, dental attendance Socioeconomic status Behavioural beliefs Perceived power and subjective norms Adverse childhood events Dental anxiety Impact of receiving care e.g. impact on family
Provider factors	Communication Professional skills Characteristics of the dentist Involvement of patient in decision-making
System factors	Availability and accessibility of services Time – waiting time to be seen; time for consultation Collaborations between professionals Referral policies Systems tailored to patient type e.g. specialist children services Cost

2.1.6 Choosing dental care for hypodontia

The decision to have treatment and the selection of a particular type of treatment usually involves the person with hypodontia, one or more member of the dental team, and any other significant party, most commonly parents or other family members. The choice of treatment depends on a number of clinical and patient factors, which may be influenced by the setting and decision-makers involved.

In the UK hypodontia is managed in both primary and secondary dental care settings (Stevenson et al., 2013). The pathway is variable but usually involves initial diagnosis by the General Dental Practitioner (GDP), who then refers the patient for further assessment, treatment planning and delivery of treatment in primary or secondary care (Figure 2.2). The chosen care pathway depends on the perceived complexity of care, the need for interdisciplinary treatments, local services and access. National commissioning guidance used for allocating NHS dental services in England highlight that hypodontia is a high priority that requires Level 3 (Specialist) care (NHS England, 2015, NHS England, 2018). Level 3a Specialist care is provided by a GDC-registered specialist in primary or secondary care, while Level 3b care is Consultant-led specialist services, principally in a secondary or tertiary care setting. Guidelines differentiate between these services based on the need for complex interdisciplinary treatment or additional medical, developmental or social needs. Interdisciplinary teams, including orthodontists, restorative and paediatric dentists, oral surgeons, clinical psychologists and genetic scientists (Gill et al., 2008), have been advocated to allow a number of professionals to work together in a cohesive way to provide optimum care (Tams and Ashley, 2013). A number of NHS secondary care providers have implemented this model for hypodontia care (Hobkirk et al., 2011).

Figure 2.2: An example of the care pathway for dental treatment for people with hypodontia



2.2 Decision-making in clinical practice

2.2.1 Individual's decision-making

Decision-making is the process individuals use to choose amongst alternatives. A well-made decision is concordant with the decision-maker's values and one that will, on balance, result in an action that confers the most overall benefit to the individual. Decision-making requires search processes to identify the alternatives from memory or an external source, evidence about whether an alternative will achieve the goal, and the goal itself. A process of inference strengthens or weakens each alternatives based on the evidence in the context of the goals (Baron, 2007). Intuitive decision-making is generally characterised as unconscious and rapid decision-making, driven by long-term memory, bias and heuristics. Analytic decision-making is a more reasoned process involving conscious thought and deliberation. A number of decision-making models exist but three most pertinent to individual decision-making in clinical practice are discussed: descriptive, normative and prescriptive (Baron, 2007, Bekker, 2009).

Descriptive models explain how individuals make judgements and decisions. The dominant paradigm is that of information-processing, which represents the individual as an active problem solver, purposively reasoning about decisions in a complex world. Examination of cognitive processes involved in judgment and decision-making allows thinking to be investigated to derive theories and models to explain how different individuals make different types of decisions in different contexts. This research used methods that allowed examination of cognitive processes to gain insight into thinking and to develop theory about how adolescents and parents make decisions about hypodontia care (see Section 4.3.4).

Normative models evaluate thinking and decision-making in terms of the goals of the decision-maker. It is premised on the belief that a fully rational individual will reach the optimal choice if they are adequately informed and able to compute accurately. Under ideal conditions individuals are expected to choose the alternative which provides most gain based on the probability of achieving an outcome and the value placed on that outcome. This assumes all necessary information about the decision options is available, individuals are motivated to evaluate all information without bias, and individuals can compute the utility and risk figures to identify the option with the greatest expected utility. Normative models underpin the preference elicitation method used in this research, which is discussed further in Section 2.3.

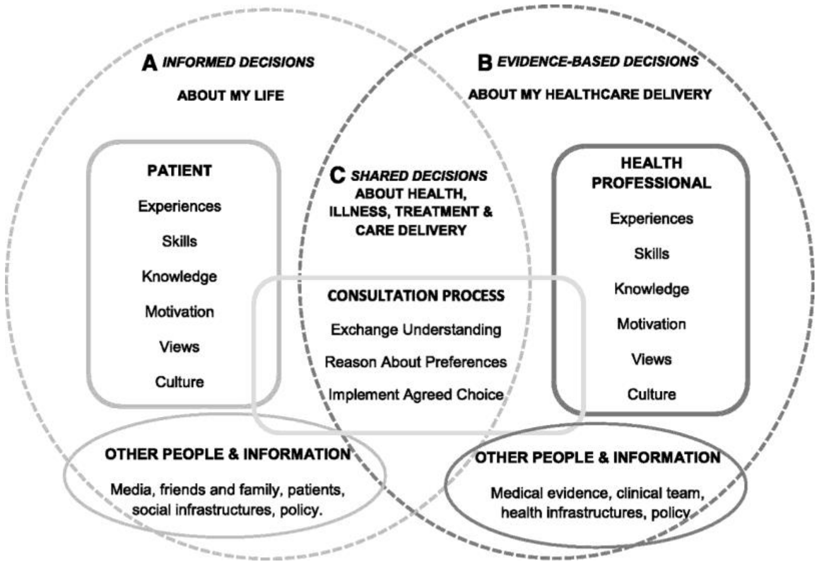
Prescriptive models describe how individuals ought to think by identifying criteria for 'good thinking' and applying methods to encourage individuals to make decisions well. This is premised on the belief that individuals can be helped to employ a decision-making process that is better for difficult decisions with serious consequences. Processing requires active open-mindedness and reasoned choice; hence it is unusual and effortful, demanding conscious engagement with information and the decision-making process. Prescriptive models underpin the concept of informed decision-making, that is, decisions should involve a thorough analysis of available information and consideration of the alternatives and consequences in an unbiased manner (Nameth, 2012, Bekker et al., 1999). Informed decisions are not judged by the consistency or coherence of the final choice but by evidence of the process. This forms the basis for clinical tools such a patient decision aids, which aim to reduce bias in decision-making and encourage better thinking. In this research, the effect of the preference elicitation instrument was examined to explore whether it changed the process for thinking about treatment.

2.2.2 Clinical decision-making

Decision-making in the clinical context has been defined as a continuous and evolving process, where data is gathered, interpreted, and evaluated in order to select an evidence-based choice of action (Tiffen et al., 2014). Decision-making historically focussed on clinician processes, which align with a paternalistic, patient-obedient model. This progressed to an informed but passive model in the mid 20th century, in which clinician choice was still dominant. Increasing value on dignity, human rights and belief in patient autonomy has driven evolution in the professional-patient relationship towards the current model, which describes clinical decision-making as a collaborative process between all important decision-makers (Blair and Legare, 2015).

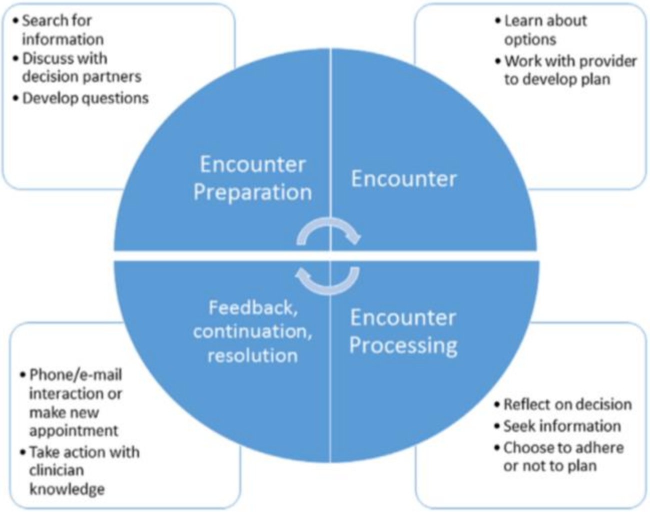
This collaborative model promotes information assimilation and evaluation by both professional and the patient to reach a mutual decision. A model presented by Bekker (2015) demonstrates how information from the patient and professional are combined in the consultation process to promote shared decision-making that includes evidence-based practice and patient values and preferences (Figure 2.3).

Figure 2.3: A model of shared decision-making based on combining patient and professional expertise in the consultation process (Reproduced with kind permission from the author – Bekker in Breckenridge et al., 2015)



The person-centred model of healthcare decisions suggests that a considerable part of decision-making occurs outside the specific clinical encounter, including time taken before to prepare and time taken after to reflect (Figure 2.4) (Clayman et al., 2017). The term person-centred was chosen to emphasise the existence of the patient as a person beyond the medical context, with wider considerations and priorities. Hence, clinical decision-making may require an iterative process to allow revisiting, revising and confirming decisions, particularly in chronic conditions with a long treatment trajectory (Agency for Healthcare Quality and Research, 2016).

Figure 2.4: The person-centred model of healthcare decisions (Reproduced with kind permission from the author - Clayman et al., 2016)



2.2.3 Decision-makers in hypodontia

Decision-making in hypodontia involves the individual with hypodontia, relevant members of the dental team and any significant person in the patient's life who may contribute to the decision and have a role in delivering the chosen care, for example, parents for children, and partners or friends for adults (Wenzel et al., 2015). Often hypodontia diagnosis occurs prior to the age of legal consent, so parents have an important legal role in consent for treatment. The concepts of informed decision-making and informed consent have become somewhat synonymous in clinical decision-making; however, a robust decision-making process incorporates more than consent (Kunneman and Montori, 2017). The distinction between the legal requirements of consent and the wider decision-making process is important for fostering inclusion of children and adolescents in decision-making (Harrison et al., 1997). Giving children an active role in healthcare decisions has a number of proposed benefits: enhancement of ownership over the treatment to improve cooperation and adherence to treatment and to encourage realistic expectations; fostering of a positive patient-professional relationship; empowerment and a sense of control; enhancement of child development through advancement of key skills (McCabe, 1996).

While inclusion of adolescents in decision-making in beneficial, it is not without challenges. Early adolescence is a period of physical, neural, cognitive and socio-economic growth. In Piaget's stages of cognitive development, the formal operational stage generally begins from 12 years old, with the development of abstract thinking and logical reasoning about hypothetical possibilities (Piaget, 1976). This hypothetico-deductive reasoning allows individuals to generate new ideas and testable hypotheses that require deductive reasoning and also results in the ability to think of potential solutions; however, Piaget suggests not all people develop formal operations and some remain in the concrete operational stage, with logical and organised but literal thinking.

Neuroeconomic research suggests important changes in decision-making processes during adolescence (Hartley and Somerville, 2015):

- Improved impulse control, working memory and complex reasoning
- Shifts in activity in neural pathways that may exaggerate response to reward and sensation seeking
- Heightened effect of positive and negative feedback on learning, with greater weighting given to rewarding outcomes than punishment
- Complicated risk perception - adolescents demonstrate understanding of the

negative consequences of risk and can comprehend probabilistic outcomes but make more risky choices compared to adults, particularly in situations where risk is learnt

- Greater uncertainty about decisions than adults with more experience
- Shift from value for immediate rewards to increased value in more substantial delayed rewards
- Context has greater influence, particularly being in a state of arousal or being with peers

Other adolescent decision-making research suggests adolescents are able to think about their options but may have limited awareness of options and lack the control required to create options. Furthermore, they may have limited appreciation of their own knowledge and lack the skills to interpret and critique information (Fischhoff et al., 1999). Compared to adults and children, adolescents are more tolerant of ambiguity in decision-making, such as uncertainty about risk and consequence (van den Bos and Hertwig, 2017).

A study examining adolescent and adult perception of decision-making found adolescents consider themselves autonomous in everyday decisions, rating themselves as having the greatest influence, followed by a close friend, then peers, with parents rated as least influential. Adults ratings for peers and parent influence on the decision-making of a '*typical 15-year-old*' were significantly higher compared to adolescents' self-ratings (Payne, 2002). In a healthcare context parents may have a greater role than in other decision-making contexts, particularly as an advocate for their child, helping to interpret information and navigate the healthcare system (Smith et al., 2015). A literature review of parent decision-making found the preferred extent of parent participation varies but most expect to collaborate with the healthcare professional and decision-making can be influenced by prior healthcare experiences, provider recommendations, other family members and community members, and family, social and cultural influences (Lipstein et al., 2012). Hypodontia has a genetic component so it is likely that a substantial proportion of parents will have hypodontia and will have already formed some beliefs about treatment based on their experience of their hypodontia. This is not an area that has yet been examined in the literature.

The dental team also have a significant influence on the decision process, but it is recognised professionals are subject to a range of biases including confirmation bias (selective information processing to support a belief), availability bias (overestimating easy to recall outcomes), regret bias (choosing to minimise anticipated regret) and framing bias (usually being influenced by negatively framed risk) (Bornstein and Emler, 2001). While there has been some discussion of biases in decision-making in dentistry (Hicks and Kluemper, 2011) to date, the only empirical research relates to racial bias when dentists are offering treatment (Patel et al., 2019). It is also important to recognise the imbalance in knowledge and power between the professionals, patient and family, which is more widely discussed in general healthcare than dentistry (Smith et al., 2015, Goodyear-Smith and Buetow, 2001).

Other parties outside the immediate decision-making setting who may contribute to the decision are peers, other people with hypodontia, and increasingly, social media. Adolescents spend considerably more time with their peers than with their parents or other adults and it is assumed that peers have a role in many different aspects of decisions and behaviour (Tome, 2012). There is a suggestion that decision-making in adolescence may be particularly modulated by emotion and social factors, for example, when adolescents are with peers or in other affective contexts (Blakemore and Robbins, 2012). The influence of peers has mostly been investigated in relation to risk taking behaviour with somewhat conflicting results. The importance of peers is potentially associated with age, confidence in social skills, individual’s resilience against peer influence and the type of friendship. Evidence suggests that rather than peers influencing decisions, the choice of peers may be determined by similarity of interests and characteristics. A study of adolescents in Sweden found the decision to have orthodontic treatment was influenced by fitting in to social norms, but often the adolescents were not fully conscious of this external influence (Trulsson et al., 2002).

Social media is emerging in healthcare as a source of information and advice, a means to understand patient experience and breakdown the patient-professional barrier and make communication less intimidating (Stones and Smith, 2018). The power of social media to influence decision-making has been demonstrated in the antivaccination movement (Smith and Graham, 2017) and the ability to rate and review care providers might become increasingly important in healthcare decisions where there is a choice of provider (Rozenblum and Bates, 2013). Although social media is a way to share experience and gain peer support (Chung, 2014), the effect of this on individual’s care choices in dentistry has not yet been investigated.

2.2.4 Shared Decision Making

Shared decision-making (SDM) is currently advocated by the NHS as the preferred approach to decision-making (NHS England, 2019). For this reason, SDM is the focus of this research but it is acknowledged that other approaches to collaborative decision-making exist. The informative model is focussed on the professional providing relevant information to allow the patient to select treatment, while the interpretative model develops the patient-professional relationship further to include elicitation of patient values and selection of treatment to realise these values (Emanuel and Emanuel, 1992). Person-centred care, including decision-making, focuses on the needs of the individual to ensure preferences, needs and values guide decisions (NHS Health Education England, 2019). Other more specific approaches have been suggested, for example, the Sharing Evidence Routine for a Person-Centred Plan for Action (SHERPA) framework, which aims to facilitate conversations between professionals and patients to support person-centred, evidence-informed, interpretative and collaborative decision-making through sharing and linking problems and planning together (Jack et al., 2018).

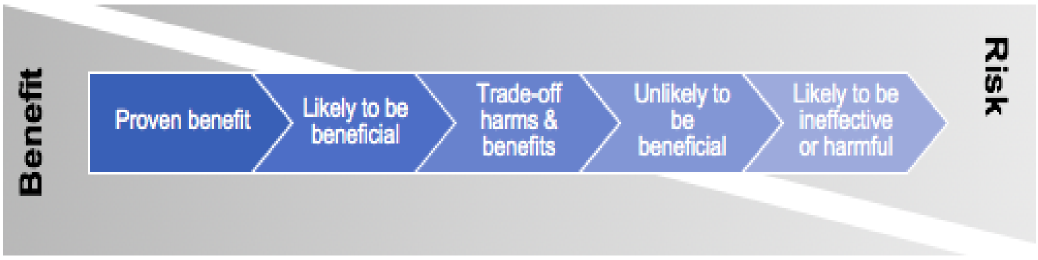
SDM was first formally defined in 1982 (The President's Commission for the Study of Ethical Problems in Medicine and Biomedical and Behavioral Research, 1982) and although different definitions have since been suggested, it is generally thought of as a collaborative process in which healthcare providers support and enable patients to make decisions about health care that are right for their individual circumstances (Da Silva, 2012) and concordant with their values and wishes (Barry and Edgman-Levitan, 2012). SDM has been conceptualised as the meeting of two experts, who are each recognised to have specific expertise and role (Table 2.6) (Charles et al., 1999).

Table 2.6: Healthcare professional and patient roles in decision-making

	Health Professionals	Patient & decision partners
Expertise	Understanding of diagnosis, aetiology and prognosis Knowledge of treatment options Knowledge of evidence base: Risks, benefits and areas of uncertainty Understanding of healthcare system	Impact of condition Own values and preferences Circumstance that may affect decision: capacity for self-care, ability to receive care
Role	Provide evidence-based information tailored to individual Assess understanding and support interpretation of evidence Support decision-making	Provide information about impact of condition, expectations from care, circumstances that affect decision Share values and preferences Participate in deliberation and decision-making

SDM is relevant at any decision point in the treatment journey where more than one option exists, including the option for no treatment (Da Silva, 2012). SDM requires an element of equipoise, that is, acceptance that there is more than one option and the absence of evidence to suggest one option is clearly superior (Elwyn et al., 2000). This is judged on the strength and quality of evidence around risk and benefit in the particular clinical context of the decision (Figure 2.5). Choice situations where evidence is equivocal have been labelled ‘preference-sensitive’ to emphasise that the decision is predominantly preference-based (Wennberg et al., 2002); however, this can be seen to imply preferences are not important to all decisions.

Figure 2.5: Variation in strength of evidence of benefit & harm from an intervention



SDM aims to optimise the decision-making process rather than change the decision itself or succeeding health outcome. In some cases, SDM may result in patients and professionals agreeing to disagree about the best course of action; however, this is considered preferable to encouraging uptake of an intervention that patients do not feel is in their best interest (Da Silva, 2012).

2.2.5 Benefits of SDM

Preference misdiagnosis has historically been overlooked as an error in healthcare compared to medical misdiagnosis, but failure to identify preference can be equally damaging to health (Mulley et al., 2012). SDM aims to select the right treatment for the individual in their particular circumstances, and if successful, SDM can be beneficial for individuals, professionals and organisations (Table 2.7).

SDM is a complex phenomenon to measure effectively and evidence for particular approaches and interventions is lacking. However, it is widely accepted that SDM is ethically sound hence future work should not be targeted at whether SDM should be done, but should instead establish how to make SDM most effective (Foot et al., 2014, Coulter and Collins, 2011).

Table 2.7: Benefits of shared decision-making to different stakeholder groups (National Learning Consortium, 2013, Da Silva, 2012, The Heath Foundation, 2016, Blair and Legare, 2015)

Patient
Involvement: Less jargon & more structure for consultation, greater involvement **Value-based:** Recognition of personal values and preferences, informed decisions consistent with personal values **Knowledge:** Increased understanding of risks, benefits and consequences of screening and treatment options, improved health education and knowledge, self-confidence in knowledge **Decision-making:** Better informed about decisions and choices, increased comfort and confidence in decisions and treatment, reduced decisional conflict **Satisfaction:** Greater satisfaction with consultations, improved experience of care **Outcomes:** Enhanced self-efficacy, better adherence to treatment, greater treatment benefits, confidence in self-care
Healthcare professionals
Supports delivery: Structure for consultation, access to relevant information, improved communication, counselling and consultation skills, better use of clinical skills **Outcomes:** Improved patient adherence to treatment/medication, better outcomes **Satisfaction:** Greater satisfaction due to better informed, more engaged and more satisfied patients **Professional development:** Helps teams consider their joint philosophy and approach to patient care and involvement, personal/professional development **Quality of care**: Improved effectiveness, safety, experience
Healthcare organisations
Prioritisation: Focus on what is important to patients, more sophisticated role for patient and public engagement, care more reflective of patient preferences and desired outcomes at individual & population level **Quality:** More standardised, better quality patient information & decision support **Support delivery:** Effective implementation of evidence-based practice, increased consistency of approaches within teams, skilled clinicians using their time more effectively, greater patient involvement in treatment decisions, development of measures and reporting on patient experience **Outcomes:** Improved patient adherence to treatment/medication, better outcomes **Satisfaction:** Increased patient satisfaction, potential reduction in complaints and litigation **Quality of care:** Improved effectiveness, safety, experience **Cost-effective:** use limited resources to provide the treatment people want and increased adherence to treatment regimes **Standardisation:** Reduction in unwarranted variation in rates of intervention

2.2.6 Barriers to SDM

Despite the drive to embed SDM into routine NHS practice a number of barriers have been identified. These are general across healthcare, rather than specific to dentistry or hypodontia. To date there have been no studies about incorporation of SDM in hypodontia, and only a few more widely in dentistry (Alzahrani and Gibson, 2018, Asa'ad, 2019, Ryan and Cunninghm, 2013).

The first barrier to SDM is professional's recognition, attitude and willingness to participate (Legare et al., 2008, Pollard et al., 2014). Although physician attitude has been found to be largely positive the extent of support varies by specialty, care setting, condition and treatment-related factors, and reported attitudes are not always consistent with the approach taken in practice (Joseph-Williams et al., 2014). The beliefs that SDM is already happening or that SDM is synonymous with informed consent are obstacles to changing practice (Legare et al., 2008, Pollard et al., 2014, Joseph-Williams et al., 2014).

Time constraints are one of the most commonly cited concerns for healthcare professionals (Legare et al., 2008). This is particularly pertinent in healthcare systems like the NHS where targets and tariffs discourage deliberation and favour rapid decision-making to promote progress through the care pathway; however, a Cochrane review of SDM interventions found no good evidence that consultation time is increased compared with usual practice (Stacey et al., 2017) and SDM has actually been advocated as a time-saving measure due to proposed improvement in understanding, compliance and adherence (Coulter and Collins, 2011, Da Silva, 2012).

Other attitudinal issues that affect SDM amongst medical professionals include: a lack of trust in SDM, often resulting from a misconception that SDM threatens professional autonomy, causes a sense of uncertainty, abandons patients to make choices alone or is too artificial and rigid to make a difference to either process or outcome; a belief that SDM does not apply to characteristics of certain patients or clinical situation; a lack of trust in the evidence around specific aspects of SDM; a belief that SDM is difficult to do and will not make a difference to outcome (Legare et al., 2008, Pollard et al., 2014, Legare and Thompson-Leduc, 2014, Legare and Witteman, 2013)

A paradigm shift in decision processes is required to shift power and control of interactions from clinicians to patients and change from evidence-based scientific practice to informed preferences (Elwyn et al., 2017). Evidence indicates that patients want to engage at a higher level that perceived by professionals (Deber et al., 2007, Chapple et al., 2003) and actually SDM is actually more beneficial to disadvantaged groups (Durand et al., 2014). The professional's role is to provide information, and recommendations where appropriate, and this addresses the concerns that patients are not equipped or able to make the right decision (Buetow and Kenealy, 2007).

2.2.7 SDM process

A number of different approaches to SDM have been proposed, in terms of steps to deliver SDM, and currently there is no consensus that one single approach is superior; however, most share common elements (Agency for Healthcare Quality and Research, 2016, Legare et al., 2011, Makoul and Clayman, 2006, Elwyn et al., 2017):

1. Fostering choice awareness and inviting participation
2. Information exchange
3. Value and preference clarification
4. Deliberation and decision

2.2.7.1. Fostering choice awareness & inviting participation

SDM commences with awareness of the need for a collaborative decision based on existence of more than one treatment option and acceptance of equipoise, and an invitation to the patient to participate. It is necessary to ensure the patient understands the problem and explicitly informed that there is decision to be made and that there is no single right choice, with reassurance that the best option will depend on individual circumstances (Makoul and Clayman, 2006). Failure of clinicians to recognise decision points where more than one option exists and implement SDM has been identified as a key barrier (Legare and Witteman, 2013).

Despite many health professionals' beliefs, the majority of patients wish to participate in healthcare decisions, often to a greater level than is realised (Flynn et al., 2006, Chapple et al., 2003); however, patients may be less keen to ask questions and participate if they fear being 'difficult' patient (Frosch et al., 2012). Methods to encourage patient participation have been shown to be an effective way to foster choice awareness and start the conversation, including the "Know your options" campaign (Agency for Healthcare Quality and Research, 2016) and Ask Three Questions (Shepherd et al., 2011).

2.2.7.2 Information exchange

Information exchange is a two-way process that aims to educate patients and families to allow meaningful conversation about the options in their own circumstance (Agency for Healthcare Quality and Research, 2016). The term exchange is used to reflect that it is more than passive information provision, where professionals clarify understanding and personalise information based on the feasibility of options in the healthcare setting and the personal circumstances of the patient, and where necessary, re-attribute incorrect beliefs (Makoul and Clayman, 2006).

Information includes details of the condition, treatment options with description of how they work, estimate of expected outcomes of different treatment, risks of treatment, service delivery and requirements for self-care (Makoul and Clayman, 2006, Charnock, 1998). The supporting evidence base should be clear along with identification of areas of uncertainty (Charnock, 1998, International Patient Decison Aids Standard Collaboration, 2005). Evidence-based, non-biased information exchange can be challenged by a lack of relevant high-quality patient-centred research.

Effective communication depends on the accessibility of information, for example literacy, numeracy and presentation of information (Legare and Witteman, 2013). Increasing the volume of information without optimising the accessibility of information does not improve understanding and can lead to information overload (Klerings et al., 2015). Despite misconceptions, SDM does not preclude professionals from providing a recommendation (Makoul and Clayman, 2006). To the contrary, professionals are experts in the condition, options and supporting evidence and where this suggests superiority of one treatment over another, recommendations would be beneficial (Legare et al., 2008, Pollard et al., 2014).

2.2.7.3 Values and preference clarification

This stage aims to identify important values that might influence preferences, understand preferences for treatment process and outcomes, and assess individual’s perception of risk. Approaches are based on prescriptive decision-making theory, aiming to improve the quality of the decision-making process options with uncertainty or risk by promoting active reasoning and reducing information bias.

Patient decision aids (PDAs) are tools that help people participate in decision-making by explicitly stating the decision to be made, the options and outcomes, and clarifying personal values. PDAs have been shown to promote more cognitive and emotional strategies during decision-making, leading to a more effective choice (Bekker et al., 2003). The International Patient Decision Aid Standards (IPDAS) Collaboration, developed in response to the growing volume of PDAs, applied a Delphi approach to develop internationally approved quality criteria to judge the content, development process and effectiveness of PDAs (International Patient Decison Aids Standard Collaboration, 2005). Guidelines for reporting decision aid evaluations have been published to promote scientific rigour (Hoffman et al., 2018). A Cochrane review of PDAs for treatment or screening identified 105 randomised controlled trials, which collectively found, compared to usual care, PDAs increased participant knowledge, improved accuracy of risk perception, improved congruency between informed values and care choices, reduced decisional conflict related to feeling uniformed, reduced indecision about personal values, reduced the number of passive decision-makers and the proportion of undecided participants, and improved patient-professional communication and satisfaction with the decision-making process (Stacey et al., 2017).

A literature search found no decision-support tools specifically for hypodontia; however, two PDAs have been developed and tested for fixed orthodontic appliances (Marshman et al., 2016). The first, which used a pre-post design with 30 patients aged 12-16 years old and parents found a significant reduction in decisional conflict, increase in knowledge and no change in expectations (Marshman et al., 2016). The second, a randomised controlled trial of 76 patients, compared a group given a different PDA with a control group who received only standard information. Although the PDA did reduce decisional conflict, the difference between the groups was not significant (Parker et al., 2017). The findings are difficult to compare directly due to heterogeneity in study design, but both promote further research in this area.

Value clarification methods (VCM) help individuals and decision partners think about the desirability of treatment options or attributes of options within a specific decision context (Fagerlin et al., 2013). Mechanisms include consideration of options and outcomes to understand what it means to have the intervention and consequences, consideration of how others value the options and whether decision-maker is similar to others, and rating, ranking or trading off features of options to understand personal preferences. Synthesis of evidence evaluating VCMs within a PDA found mixed effects and currently it is accepted that VCMs are unlikely to be harmful, but evidence of their effectiveness is lacking (Fagerlin et al., 2013).

2.2.7.4 Deliberation and decision

Deliberation is necessary to reach a decision and the time required is judged on an individual basis, with the opportunity to make or defer the decision as required (Agency for Healthcare Quality and Research, 2016, Makoul and Clayman, 2006). Deliberation aims to reach a reasoned decision based on accurate information, evaluated within own beliefs, employing trade-offs between the options. Differences may exist between actual treatment choice and patient preference, clinician preferences and guidelines (Montgomery and Fahey, 2001).

Evaluating individual's choice and the impact on outcome helps further guide SDM. This is distinct from evaluating the SDM process to understand the experience of, and satisfaction with, decision making. Understanding intentional non-adherence through collaboration with the patient provides an opportunity for evaluating the decision and whether the SDM process was effective (Clayman et al., 2017). The SURE is a validated clinical tool for screening for decisional conflict in patients based on four questions: certainty with decision; understanding of information; understanding of the risk benefit ratio; perception of adequate support to make the decision. The SURE tool has been validated in a number of clinical settings including women considering pre-natal screening (Legare et al., 2010) and for antibiotic use in respiratory infection (Ferron Parayre et al., 2014); however, the tool has not been validated in the dental setting.

2.2.8 Evaluating the quality of decision-making

Decision-making interventions can be evaluated in terms of the quality of the decision-making process (deliberation), the choice that is made (determination), the knowledge used to make the decision, and concordance between the choice and decision-maker's attitudes, preferences and values (Elwyn and Miron-Shatz, 2010).

Measurement of the decision-making process itself can include assessment of knowledge, understanding of the decision and options, or engagement in the decision-making process (Elwyn and Miron-Shatz, 2010). Measuring knowledge is based on two assumptions; that knowledge is necessary to allow deliberation, and conversely, that knowledge correlates with good decision-making. Determining a valid measure of knowledge is challenging, for example, deciding whether items should relate to options, attributes of options, possible outcomes, probabilities of outcomes, or consequences of outcomes (Elwyn and Miron-Shatz, 2010, Scholl et al., 2011).

A number of approaches have been reported for establishing engagement in the decision process (NHS, 2012, Gartner et al., 2018). The most widely recognised is the OPTION scale, a validated measure to assess the extent to which professionals involve patients in consultations, based on definition of the problem, a statement of professional equipoise, communication of options and risks, conducting or deferring the decision process (Elwyn et al., 2003). Following user feedback the coding was revised from attitudinal to magnitude-based scale (Elwyn et al., 2005). The OPTION scale has been translated into Italian, German and Dutch and an observer and dyadic version.

Numerous patient questionnaires, provider questionnaires and coding schemes have been developed to evaluate the SDM process (Scholl et al., 2011). It is outside the scope of this thesis to evaluate all tools, but two commonly used measures are outlined. The first, the Shared Decision-Making Questionnaire (SDM-Q), is a 9-item tool to measure the SDM process in clinical encounters based on nine steps identified as important in the SDM process (Simon et al., 2006). The tool has been translated from German to English, Spanish, Dutch and Hebrew and a provider version has been developed. The second, CollaboRATE, measures SDM against 3 process items, provision of information, elicitation of preferences and integration of preferences, using either a 10-point or 5-point scale (Elwyn et al., 2013a). A recent systematic review of 40 tools for measuring shared decision-making process found a lack of evidence for measurement quality and validity, and the authors recommend refinement and validation of instruments using the Consensus-based standards for the selection of health measurement instruments (COSMIN) guidelines (Gartner et al., 2018).

Evaluation of choice focuses on the perception of the decision that has been made. Most common are measures of decisional conflict and decisional regret although a number of other satisfaction, attitude and evaluation scales are reported (NHS, 2012). Decisional conflict is a state of uncertainty about the course of action to take and arises from a mismatch between intuitive and analytic decision-making processes. Decisional conflict is more likely in choices with risk or uncertainty, high gains or losses, or a need for trade-offs. Conflict can be increased by a lack of understanding, values, skills or by emotional distress or external pressure; this is the basis for the Decisional Conflict Scale (DCS) (O'Connor, 1995). The three domain tool measures uncertainty, factors contributing to uncertainty and perception of effective decision-making and testing has demonstrated acceptable psychometric properties and usability.

Decisional regret is more complex to define, as regret itself has different philosophical and theoretical origins and can be similar to other emotions, such as disappointment, guilt and sadness. In the Decisional Regret Scale (DRS) is a scale to measure the construct of regret, defined as remorse or distress over a decision (Brehaut et al., 2003). The five items include presence of regret, satisfaction with the decision, likelihood to make the same decision, whether the result was wise and whether it had resulted in harm, marked on a 5-point scale after reflection on specific treatment decisions.

2.3 Preference elicitation research

The review of clinical decision-making highlights the importance of understanding and incorporating patient preferences. The growing use of preference elicitation research methods reflects the increasing drive to consider patient preferences when deciding healthcare financing and delivery of service (Lancsar and Louviere, 2008). The use of research preference elicitation methods within a clinical setting to help decision-making is a growing area of interest (Dowsey et al., 2016, Ryan, 2017). In the present research the preference elicitation method was used to examine decision-making processes and preferences without direct translation of the findings into individual’s clinical care but to inform development of decision support tools and the suitability of this approach for application in clinical care.

2.3.1 Revealed and stated preferences

Preferences can be elicited directly or inferred from actual choice behaviour (revealed preferences) or hypothetical choice behaviour (stated preferences). Revealed preference (RP) methods are observational, using actual choices in real-life contexts to estimate preferences (Zin and Roper, 2019). The main benefit of RP is that the choice observed is consistent with real-life behaviour, so minimal assumptions are required; however, the approach is limited by a number of factors (Johannesson, 1996, Hicks, 2002): the inability to control the decision-making environment, limiting testing to existing alternatives; unawareness of the choice situation and all alternatives available, presenting challenges for establishing trade-offs; inability to test options that are not in the choice situation; correlation of characteristics between options causing bias in the estimation; data collection can be expensive and time-consuming as often each respondent is limited to providing a single response.

Stated preference (SP) methods are an experimental approach to measuring preferences where the researcher creates hypothetical choice scenarios to examine factors of interest while controlling for known confounders that might affect the choice.

This provides a degree of control over the choices and environment, allows preferences to be estimated for non-existent alternatives and enables control of collinearity between attributes and potential confounding factors (Kroes and Sheldon, 1988). The use of hypothetical scenarios is limited in that behaviour might not reflect real-life behaviour due to unrealistic options or environment, a lack of motivation and desirability bias (Kroes and Sheldon, 1988). Another recognised limitation of SPs is hypothetical bias, which can result in a tendency to overestimate preferences (Loomis, 2011). Due to the complexity of hypodontia, stated preference approach was selected for this research to provide control over the choice context, to ensure there was an explicit statement of choice and to allow manipulation of the alternatives.

2.3.2 Utility-based approaches to preference elicitation

Economic-based approaches to quantification of preferences include direct elicitation of the monetary value placed on goods or services to estimate demand or willingness to pay or choice behaviour modelling to estimate utility without direct monetary valuation. The latter method is the approach used in this research. Utility is defined in economics as a measure of preference over a set of goods or services. Utility is not directly observable but can be estimated from the choices made by consumers. Lancaster's theory of value (1966) states that all goods and service consist of a number of characteristics (attributes), and overall utility is derived from the utility of the underlying attributes. The utility maximisation theory follows a normative model of decision-making. It assumes consumer choice is based on utility maximization, where the consumer's objective is to maximize the total value derived from available resources. Utility maximisation assumes rational consumers will sacrifice attributes with a lower perceived utility for those with higher utility, hence by measuring trade-offs it is possible to estimate utility and preference. A utility-based approach allows estimation beyond monetary valuation by including trade-offs between characteristics of the process, non-health outcomes and health outcomes. This has been used to evaluate current goods or services, design new interventions and predict uptake of new services or goods (Ali and Ronaldson, 2012).

The analytic framework used for estimating utility in the Discrete Choice Experiment (DCE) context is the Random Utility Model (RUM), based on Thurstone's Law of Comparative Judgment (Thurstone, 1927), which assumes i) choice is a discrete event ii) utility towards a good varies across individuals as a random event and iii) consumers will make choices to obtain the highest utility. Therefore, utility has both a systematic and a random component; systematic attributes explain the difference in choice alternatives while random components are unidentified factors that may influence choice.

Based on the RUM, an individual’s utility for a particular good is:

$U_{ij} = V_{ij} + gij$ *w*here U_{ij} is the utility of choice *j* for individual *i*
V_{ji} is the systematic component
g_{ji} is the random component

The assumptions underlying utility-based preference elicitation may be violated where cognitive shortcuts are employed, the preference context is not understood ‘irrational’ choice behaviour is seen or respondents refuse to trade off (Ali and Ronaldson, 2012).

2.3.3 Stated preference methods

Three common stated preference approaches are pairwise comparison, best-worst scaling (BWS) and Discrete Choice Experiments (DCE). Pairwise comparison allows calculation of the relative importance of options by ranking alternatives. Options may contain single-attribute items or multi-attribute items. Paired options are placed into a grid for comparison and cells are used to select which option is preferred from the pair. Preference is converted into a score to calculate a total score and ranking for each option. In dentistry, single-attribute statements in pairwise choices have been used with adolescents to judge the relative importance of statements relating to dental problems, general health and psychological well-being (Assink et al., 1995) and dental appearance, health, body shape, school grades, friends and sports (Bos et al., 2008).

BWS was first reported in 1992 (Finn and Louviere, 1992) and has since gained popularity in health and social care as a method to compare the absolute utilities of attributes. BWS asks respondents to choose the best and worst items from a list; three approaches have been developed (Marley and Flynn, 2015):

- Object Case: relative value of objects is obtained by presenting choice sets to respondents to select the best and worst objects
- Profile Case: attribute-levels are selected from a single multi-attribute profile
- Multi-Profile Case: Selection of best and worst multi-attribute profiles

BWS assumes ordinality and this enables the impact (average of the utility across all levels) of all but one of the attributes to be estimated (Flynn et al., 2007). In a study with adolescents comparing object case BWS with a simple rating exercise both methods were shown to be able to predict actual choice but BWS showed greater sample discrimination and rating was judged to be easier to perform by respondents (Mielby et al., 2012). A 2016 systematic review of BWS in healthcare found a rapid increase in their use over the last 5 years, with a total of 62 BWS studies including 26 object case, 29 profile case and 7 multi-profile case. BWS was most commonly used with the general public, patients and healthcare professionals to value health outcomes, investigate trade-offs between health outcomes and treatment experience, and to establish professionals' preferences for screening and treatment (Cheung et al., 2016).

DCE uses choice tasks, which are scenarios with two or more alternatives, with each alternative containing multiple attributes. Respondents are presented with a series of choice tasks and asked to select their preferred alternative in each task. Preferences are estimated by modelling the choices made across multiple choice tasks by multiple respondents. DCE has been shown to produce similar preference estimates to profile case BWS (Potoglou et al., 2011). An initial systematic review assessing the use of DCE in healthcare identified between 2001-2008 found 114 studies, with 20 of these (59%) conducted in the UK (de Bekker-Grob et al., 2012). A successive review found a further 179 studies in the period 2009-2012, with increasing popularity outside the UK (Clark et al., 2014). The most recent review found 301 studies in the period 2013-2017 (Soekhai et al., 2019), emphasising the growing popularity of DCEs. The reviews found DCE has been applied widely to explore a variety of topics including the value of experience factors, the value of health outcomes, trade-offs between health outcomes and experience factors, utility weights within the quality-adjusted life-year (QALY) framework, healthcare professional's job-choices, development of priority setting frameworks, and health professional's preferences (de Bekker-Grob et al., 2012, Clark et al., 2014, Soekhai et al., 2019). DCE has been used to identify important patient-centred outcomes for use in a clinical trial (Stamuli et al., 2017) and more recently, to value process utility benefits in economic evaluation (Tinelli et al., 2016), recognising the importance of patient experience when quantifying the value of care.

2.3.3.1 Rationale for use of DCE

DCE was selected for this research because it most accurately reflected the clinical decision-making context. In hypodontia care the choice of treatment often requires trade-offs between unfavourable parts of the treatment process and the desired outcome. The ambition of the research is to drive changes in clinical decision-making, by improving communication and inclusion of patient preferences. Use of DCE provided an opportunity to examine whether trade-off tasks and presentation of comparable options helped adolescents and parents to think about the alternatives, risks and benefits. This information is important for design future decision support interventions.

2.3.4 Overview of DCE methods

DCE instruments are developed in sequential stages and best practice guidance has been developed to encourage rigour in methods (Bridges et al., 2011, Hauber et al., 2016, Reed Johnson et al., 2013). DCE requires a clear research question outlining the purpose of preference elicitation, the good or service under investigation, the population of interest and the intended use preference information (Reed Johnson et al., 2013). The key stages in the DCE include:

1. Attribute development
2. Creation of choice scenarios
3. Instrument development and testing
4. Data collection and analysis

2.3.4.1 Attribute development

Valid preference elicitation is predicated on identifying and selecting attributes that are appropriate to the research question (Bridges et al., 2011). The most suitable approach depends on the study question, population and perspective and appropriate mixed methods are recommended to ensure a comprehensive approach (Helter and Boehler, 2016, Mangham et al., 2009).

Attributes must be:

- Salient, plausible and capable of being traded (Ryan, 1996)
- Cognitively and statistically uncorrelated in choice sets (Abiiro et al., 2014)
- Not in violation of the conceptual framework for the random utility theory (Coast et al., 2012), that is, attributes must be relevant and include all that might influence decision, not too close to latent construct under investigation, not dominant to the point of being deterministic, not intrinsic to personality, and open to manipulation by intervention

Attribute-levels are the dimensions that describe the attribute, for example, the cost of a medicine might be assigned the levels £4, £10 and £20. Attribute-levels should be relevant and have sufficient range to enable discrimination, but not so extreme they lose realism and meaning (Lancsar and Louviere, 2008, Viney et al., 2002). A two-stage approach to attribute development has been suggested, with the first identifying and selecting relevant attributes and the second stage focussing on identifying meaningful wording and levels to ensure the terminology and description evoke the desired meaning for the target respondents (Coast et al., 2012). In healthcare, the reported number of attributes ranges from 2 to >10, with most being in the region of 4-6 (Clark et al., 2014, Ryan and Gerard, 2003) and a maximum of 3-4 levels (Bridges et al., 2011).

2.3.4.2 Creation of choice tasks

To create the choice scenarios, profiles (alternatives) are created by combining attribute-levels, and profiles are combined to form choice tasks. The number of potential profiles, calculated from the number of levels (L) to the power of (A) attributes, is often too large to use in full and consequently a fractional factorial experimental design is common. The selection of profiles governs the effect of the attribute-levels on the dependent variable and hence determines the indirect utility functions that can be estimated from choices. The experimental design aims to generate sufficient variation in attribute levels to examine choice (Reed Johnson et al., 2013). Profiles are selected to provide level balance (each level occurs the same number of times), utility balance (each option has a similar probability of being chosen), orthogonality (attributes vary independently and are not correlated), efficiency (smallest variance matrix), plausibility (attribute-levels combined within a profile make sense) and non-dominance (avoidance of choice sets with one profile that is superior in all attributes). Design efficiency (d-efficiency) is a function of the variance and covariance of the parameter estimates and is affected by the number of alternatives per choice task, the number of attributes and attribute-levels and the number of choice tasks (Vanniyasingam et al., 2016).

To construct the choice tasks, the choice process is considered in terms of the choice context, the nature and composition of the options and the framing around the choice questions. Ideally choice sets should be constructed to promote incentive compatibility, that is, selection of the ‘true’ choice rather than one that is seen to be desirable or strategically advantageous (Zawojsk and Czajkowski, 2017). Task construction requires consideration of the number of attributes included (full or partial profile), use of labelling of alternatives, the opportunity to opt-out or retain the status quo and the ideal number of tasks. In healthcare research, a full profile is considered good practice (Bridges et al., 2011) but a partial profile, achieved by selecting a subset of attributes for inclusion in each profile or by constraining some attribute-levels, may reduce cognitive burden for respondents and improve choice consistency and completion rates (Jonker et al., 2019).

The choice to use labelling versus generic labels depends on whether labels are likely to encourage detrimental heuristics in decision-making. Labels may convey additional information about tangible or intangible qualities making processing easier by reducing the number of attribute trade-offs required and providing a more realistic context based on existing beliefs or knowledge; however, generic terms may encourage more discerning responses by forcing respondents to read the choice options. The opportunity to opt-out or retain the status quo in the choice set is advocated to reduce the error associated with forced choice in situations where respondents may be non-consumers (Lancsar and Louviere, 2008, Ryan and Gerard, 2003).

Preference may be elicited as a simple choice between alternatives, or with a rating to indicate strength of preference. Additional information about the choice, such as ease of choice, certainty in choice, likelihood to make the same choice in real-life can be collected to aid more detailed examination of choices (Bridges et al., 2011).

2.3.4.3 Instrument design and testing

Instrument design considers the presentation of the choice tasks alongside any information about respondent demographics and experience that is required for interpretation of results or subgroup analysis. The survey is framed with contextual information, including a description of attributes and levels and an introduction to the task, ideally including an example task. Pictures, diagrams or symbols may aid comprehension, particularly for low literacy populations or to explain more technical information. Training methods have been suggested for helping respondents understand complex information, such as serious games (Vass et al., 2018b) or use of ‘cheap talk’, which is strategy for co-opting people to become part of the research team by highlighting common errors made by other people when completing the DCE choice tasks (Reed Johnson et al., 2011). Assessment of respondents’ existing experience and knowledge determines how much background information is needed to avoid assumptions being made by respondents. A ‘report card’ may be used prior to the DCE to establish which attributes and levels most accurately reflect their status quo to inform model estimation (Bridges et al., 2011).

Piloting is an important aspect of DCE development to allow validity testing of the instrument including comprehension or completeness, review of the choice experimental task complexity and cognitive burden for respondents, identification of relevant attributes that have been omitted, estimation of preference weight size and direction to improve the efficiency of the experimental design and in some cases, contribution of preliminary choice data for analysis (Greiner et al., 2014). Piloting can include quantitative examination of preliminary choice data (Janssen et al., 2018) and/or qualitative methods to gain in-depth feedback about particular aspects of the DCE instrument (Vass et al., 2017).

2.3.4.4 Data collection and analysis

As with other quantitative research methods, the sample should be representative of the target population. Calculating the ideal sample size for quantitative analysis of stated preference experiment data is complex and depends on the output of interest (attribute preference estimate, marginal rate of substitution or difference between groups), the strength of expected differences between attributes, the level of certainty required, the size of the target population, the planned DCE methodology, any planned subset analysis and the expected preference heterogeneity in the sample (Orme, 2010, de Bekker-Grob et al., 2015). ‘Rules of thumb’ for sample size for DCE include 300 respondents in total or between 50-200 per group where subgroup analysis is planned (Orme, 2010), or 20 respondents per questionnaire version (Lancsar and Louviere, 2008). Data collection methods may be undertaken face-to-face or via remote methods such as postal or online. A recent study involving adult respondents found similar results between online and paper survey administration (Determann et al., 2017), and online and face-to-face methods were found to produce the same valuation of health states with adult respondents (Mulhern et al., 2012).

Statistical analysis of multi-attribute stated preference data is based around principles of regression analysis and different models can be used depending on the choices being modelled and assumptions regarding distribution of the random component (Viney et al., 2002). Different approaches to analysis are discussed further in Chapter 5. Estimate coefficients (β-coefficients) capture the mean preference weights of the attribute-levels and the relative contribution of the attribute-level to the utility assigned to the alternative. The magnitude of difference between most and least preferred level of the same attribute indicates the relative importance of the attribute (Hauber et al., 2016). Preference weights alone are difficult to apply clinically but they can be used to order attribute-levels relative to each other.

Alternatively, the marginal rate of substitution (MRS), which is the rate at which a consumer would be willing to trade one attribute for another, can be calculated from preference estimates. Common examples of MRS in healthcare include monetary equivalence (Willingness to Pay), time equivalence (Willingness to Wait) (Marshall et al., 2018), risk equivalence (maximum acceptable risk (Boeri et al., 2018, Hauber et al., 2017), willingness to risk relapse (Rothery et al., 2016)) and travel equivalence (Willingness to Travel) (Zickafoose et al., 2015). A linear utility function assumes marginal disutility is constant; however, in reality the additional disutility over the attribute range is likely to change, so a quadratic or stepwise utility function may be required (van der Pol et al., 2014). Alternatively, preference weight ratios can be calculated by dividing utility differences resulting from an incremental change in one attribute (for example one-unit improvement in health) by the incremental utility of another (for example, an increase of one pound) but coefficients need to be adjusted to account for differences in scale.

DCE preference models can also be used to predict uptake of goods or service (Terris-Prestholt et al., 2016). As this research did not aim to create predictive models, this application of DCE preference data is not discussed further.

2.3.5 Validity of DCE

The validity of DCE methods reflects the extent to which the quantitative measures of relative importance or trade-off reflect true preferences of the respondents. The framework suggested by Janssen et al. outlines principles and measures of validity and reliability (Table 2.8) (Janssen et al., 2017). One of the most complex but useful measures is external validity, that is, whether hypothetical choices reflect actual observed treatment choices. This is one of the main concerns with stated preference methods. Clinical encounters potentially provide a valuable opportunity to examine external validity as often actual treatment choices will be required; however, hypothetical choices must adequately reflect the actual choice context and the real-life choice options must be explicit. A recent systematic review identified only eight healthcare studies that compared predicted choice from stated preference data to actual choices. Pooled estimates of data from six of the studies found 88% (95% CI 81 to 92%) sensitivity and 34 % (95% CI 23 to 46%) specificity, suggesting DCEs have reasonable predictive power (Quaife et al., 2018).

Table 2.8: Methods for evaluating Discrete Choice Experiments

Measurement reliability: Consistency or repeatability of responses	
Survey test-retest	Repeat of all choice tasks at a later date to test choice stability
Choice task test-retest	Repeat of single choice task within survey to test choice stability
Version consistency	Fixed choice task across versions to be completed by individual
Holdout prediction	Inclusion of choice task that is not part of experimental design included. Use preference estimate from model to predict choice in this task
Choice reliability: consistency in choices and agreement with assumptions about choices	
Transitivity	Preference relation between 3+ choice profiles.
Sen’s consistency	Present initial and follow on choice task that contain different number of choice profiles
Level recoding	Recode levels to test how much respondent processes number.
Measurement validity: Reflection of the accuracy and generalisability of the DCE	
Content validity	Pre-test qualitative methods & piloting Check direction of preference estimates meet prior expectations Consultation with experts
Convergent validity	Use of other instrument to allow comparison Comparison to other preferences in the literature
External validity	Comparison of predicted choice from SP with RP data
Choice validity: whether responses are as expected based on assumptions about behaviour	
Monotonicity	Within-set: Use of dominant profile in one choice task Across-set: One profile dominates across >1 choice task
Compensatory preferences	Assess attribute dominance Assess attribute nonattendance
Task non-attendance	Assess left-right preference

2.3.6 Review of relevant applications of DCE

2.3.6.1 Use of DCE in dental studies

In preparation for this research, a systematic review was conducted to understand how DCE methods have been used in dental research (Barber et al., 2018c). Studies using DCE to measure patient, public or professional preferences for any dental service, treatment or oral health state were included. Searches were completed on 22nd March 2017 and 12 suitable records were identified. Studies were undertaken between 1999-2015 with four from the UK, five from Western Europe and the remaining three from the USA. Two were unpublished PhD theses (Douglas, 2001, Zhang, 2013) and one was a commissioned reported (Lord et al., 2015). The choice of DCE was based on the desire to establish the relative importance of multiple attributes and to reflect real-life decision-making by requiring respondents to choose by making trade-offs. A summary of key features of the included studies and the quality assessment is available on request.

The purpose of the studies was to establish:

- Patient and dentist preferences for a dental intervention (caries detection device (Douglas, 2001), restorative material (Espelid et al., 2006), dental prosthesis (Zhang, 2013), sleep apnoea treatment (Krucien et al., 2013) to estimate WTP
- Patient preferences for a dental intervention (sleep apnoea treatment) to predict demand for treatment (Krucien et al., 2013)
- Public preference for attributes of tooth-whitening product and effect of risk (Arora, 2006)
- Public preference for service delivery (dental plan benefits) to predict demand (Cunningham et al., 1999, Gaeth et al., 1999)
- Patient preferences for orthodontic service, particularly willingness to travel and willingness to wait (Ryan and Farrar, 2000)
- Public valuation of oral health states for future economic evaluation (Lord et al., 2015)
- Methodological investigation of DCE using dental service delivery model (Bech et al., 2011, Kiiskinen et al., 2010, Krucien et al., 2015)

2.3.6.2 Use of DCE with adolescents

A rapid review of the literature was performed to understand how DCE methods have been used with adolescents. This aimed to establish the purpose of preference elicitation, specific experimental or survey design features used to make method appropriate for adolescents and the reported reliability and validity of methods for adolescents. The study eligibility criteria and search methods are summarised in Appendix Table 2. Searches were completed in July 2016 and updated in January 2019. The initial search identified 993 studies from which five met the inclusion criteria (de Bekker-Grob et al., 2010, Hofman et al., 2014, Marshall et al., 2016, Sung et al., 2012, Brown et al., 2014). One study was only available as an abstract (Brown et al., 2014). The updated search identified two additional studies (Forsander et al., 2018, Wang et al., 2017) and further conference abstract (Barnett et al., 2017). The key data from the studies is given in available on request.

The purpose of the studies was to elicit adolescent preferences for diabetes glucose monitoring (Barnett et al., 2017) and diabetes self-care (Forsander et al., 2018), vaccines (Brown et al., 2014, de Bekker-Grob et al., 2010, Hofman et al., 2014, Marshall et al., 2016, Wang et al., 2017) and outpatient management of low-risk febrile neutropenia (Sung et al., 2012). Specific design considerations to make the DCE method appropriate for adolescents included methods to increase validity included the use of pictures, graphs and pictograms to explain percentage and rates (de Bekker-Grob et al., 2010, Hofman et al., 2014) and face-to-face completion with a researcher present to support respondents (de Bekker-Grob et al., 2010, Hofman et al., 2014).

Tests to evaluate and manage validity included:

- Inclusion of dominant choice set with sensitivity analysis of preference estimates for those who pass or fail the test (de Bekker-Grob et al., 2010, Hofman et al., 2014)
- Additional request for respondents to rank attributes to allow a check for convergent validity (de Bekker-Grob et al., 2010, Hofman et al., 2014)
- Inclusion of 4 items with a 5-point Likert scale to measure respondents' perception of the DCE choice tasks (Hofman et al., 2014)
- Inclusion of a measure of confidence and an opportunity to provide reason for lack of certainty, with subsequent exclusion of respondents who indicated uncertainty due to lack of understanding (Forsander et al., 2018)
- Test question to assess understanding of choice task, with subsequent exclusion

of those who fail test (Forsander et al., 2018)
- Examination of dominant response for specific attribute (Marshall et al., 2016)
- Inclusion of a repeat question to check for internal consistency, with exclusion and sensitivity analysis of those who failed (Wang et al., 2017)

This information was used to inform development of the preference elicitation tool in this research (Chapter 4).

2.3.6.3 Joint preference elicitation

Different approaches to evaluation of joint or proxy preferences have previously been reported. Advocate preferences, where someone choses on behalf of another, is widely reported, for example, parents selecting preferences for their child (Brown et al., 2010, Lloyd et al., 2011, Waschbusch et al., 2011) or healthcare professionals selecting preferences for patients (Shah et al., 2015). Joint preferences have been tested by comparing aggregate preferences of independent respondent groups, for example healthcare professionals, parents/caregivers and patients (Bolt et al., 2018, Malhotra et al., 2015, Regier et al., 2012). The least used approach is examination of preferences of related respondents, such as parent-child dyads and couples (Beharry-Borg et al., 2009). The latter approach is of interest for this research.

A literature search was performed to identify stated preference studies involving joint preference elicitation from dyads. The study eligibility criteria and search methods are summarised in Appendix Table 3. Only two studies were identified that included dyadic respondents and met the inclusion criteria (Bray et al., 2016, Beharry-Borg et al., 2009). The key data is available on request. One study involved adult couples while the other included parent-child dyads. Couples, like parent-child dyads, form a natural unit for decision-making where individual preferences will shape joint decisions; however, parent-child dyads are expected to have greater imbalance in cognition, values and power.

The parent-child dyad study was a pilot that used DCE methods to elicit preferences for wheelchair services (Bray et al., 2016). The study aimed to compare preferences of children and their parents and estimate willingness to pay for different configurations of wheelchair services. The parent and child groups completed the survey independently. The individual responses were matched for nine parent-child dyads and attribute preference weights were compared. The study did not ask respondents to complete a survey together, so joint preferences were not included. Consideration was given to make the survey appropriate for adolescents, but no modifications were reported in relation to joint preference elicitation.

The couples paper examined factors influencing choice of beach on holiday with the aim of providing an analytic framework for analysing individual and joint decisions (Beharry-Borg et al., 2009). Couples were asked to complete the same DCE survey separately first then again as a couple by negotiating mutually agreed preferences. The analysis established individual preferences then used different models to test theories around bargaining models to reach joint consensus. The study provides some interesting information about use of bargaining models and estimates from individual preferences to investigate joint decision-making. The study did not report observation of negotiation process between couples, preventing the use this observed behaviour as a means to evaluate the validity of the models.

Chapter Three:

Examination of Decision-Making Practice in Hypodontia care

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Examination of Decision-Making Practice in Hypodontia Care

This chapter reports the first stage of the research, which examined current decision-making practice in hypodontia. Complementary mixed methods were used to capture a variety of experiences and perspectives. Each method is described with the key findings and what they suggest about decision-making practice. The chapter synthesises the key issues and the implications for clinical practice and future research.

3.1 Introduction

To date there has been little research to understand how decision-making is approached in hypodontia and more widely across dentistry. The first stage of the research aimed to explore decision-making practice in hypodontia to understand current practice and its limitations, opportunities for improvement and development of shared decision-making approach. Complementary methods were used to examine various aspects that contribute to the overall decision-making process and the experience of decision-making for adolescents with hypodontia and their families. The methods used were:

- Systematic review of hypodontia literature
- Analysis of patient information resources
- Naturalistic observation of clinical consultations
- Analysis of social media posts relating to hypodontia
- Interviews with adolescents with hypodontia and their parents

A preliminary scoping review of the literature was undertaken to identify and appraise current evidence for hypodontia treatment methods and to map the outcomes used to evaluate care. Systematic reviews are a recognised method for synthesising existing data to guide evidence-based practice, support development of guidelines for treatment and identify future research needs (Gopalakrishnan and Ganeshkumar, 2013). Systematic methods promote transparency and minimise bias in the selection and appraisal of evidence (Higgins and Green, 2011). Understanding the evidence base was judged to be important for establishing whether there was high quality research to support information provision about the potential risk and benefits of treatment for decision-making, and to identify areas of uncertainty.

To examine how this information is then presented to adolescents and parents to support decision-making, two approaches were used. First, written and online patient information sources were identified and examined. Information resources aim to improve health literacy by a) informing, preparing and educating patients; b) enabling more effective engagement with dental professionals and service delivery; c) encouraging participation in treatment decision making and/or self-management; and d) providing understanding about how information is used to make treatment decisions and how people can be encouraged to consider their own values (Fagerlin et al., 2013, Freeman, 1999, Winterbottom et al., 2007).

The second method involved observation of consultations between adolescents, parents and the dental team in specific interdisciplinary clinics (Hahlweg et al., 2017). This provided an opportunity to examine information provision and observe communication and decision-making practice. Measures for objectively assessing aspects of shared decision making are available (Scholl et al., 2011); however, naturalistic observation was chosen to capture normal behaviour and allow objective evaluation of processes, roles and engagement (Mays and Pope, 1995).

Interviews with adolescents and parents were undertaken to understand the experiences of people affected by hypodontia, in terms of the impact of hypodontia and its treatment, and their experiences of decision-making. A descriptive approach was taken to understand hypodontia care based in the lived experiences of participants. To provide an additional source of patient experience, a systematic search and analysis of social media posts was conducted. In the last decade there has been a substantial increase in the use of social media in healthcare by providers for health communication to patients (Campbell et al., 2016), inter-professional networking and information sharing (Ventola, 2014) and to encourage patient engagement with care providers (Liddy et al., 2017). In dentistry, social media content has been examined to explore patient experience of orthodontic treatment (Rachel Henzell et al., 2014, Noll et al., 2017), quality of life related to third molars (Hanna et al., 2017) and bullying in relation to dentofacial features (Chan et al., 2017).

3.2 Aims and objectives

This stage of the research aimed to explore current decision-making practice in the hypodontia care pathway and understand adolescent and parent experience of making decisions about hypodontia care.

The objectives were:

- To undertake a systematic review of current literature to establish the adequacy of the evidence base for supporting decision-making in hypodontia care
- To identify and appraise patient information resources to examine information provision underpinning decision-making in hypodontia
- To observe clinical consultations between adolescents, parents and the dental team to examine content, language and decision-making practice
- To explore adolescent and parent experience of decision-making in hypodontia through interviews and experience reported on social media
- To synthesise the information to form an overall view of current decision-making in hypodontia care and identify the key implications for clinical practice and future research

3.3 Systematic review of hypodontia literature

A preliminary scoping review of the literature indicated the evidence base for hypodontia treatments was poor; however, a systematic review was still judged to be an important first step in establishing evidence for hypodontia care and mapping potential attributes. The review was conducted and reported following best practice guidance (Centre for Reviews and Dissemination, 2009, Higgins and Green, 2011, Moher et al., 2009, Petticrew and Roberts, 2006). The aim was to systematically identify, appraise and synthesise studies evaluating hypodontia care with the purpose of evaluating the suitability of the evidence-base for information provision for decision-making.

3.3.1 Methods

The study was a systematic literature review of empirical studies evaluating hypodontia care. The protocol was registered on PROSPERO prior to commencement (Barber and Khambay, 2015).

Study inclusion and exclusion criteria were developed using the PICOS approach (Centre for Reviews and Dissemination, 2009, Moher et al., 2009) (Appendix Table 4).

A broad approach to study design was adopted to provide a comprehensive assessment of the evidence base (Petticrew and Roberts, 2006). Quality improvement methods, although distinct in design and purpose from research, aim to improve clinical effectiveness, patient safety and patient experience (Fereday, 2015). Audits, performance benchmarking, process mapping and other quality improvement methods were included to provide information about stakeholder priorities and expected care standards. A number of information sources were identified to ensure maximum retrieval of studies (Appendix Table 5). Search concepts were developed for hypodontia and treatment methods. MeSH terms and free headings were used (Baumann, 2016) and the terms showed good capacity for identifying appropriate studies.

An electronic reference manager EndNoteX9 (Clarivate Analytics, Philadelphia) was used to manage references and document the study selection process. For practicality, study selection was not blinded by removal of author, institution or publication details. Records from the electronic database searches were collected and imported into the reference manager. Programmed de-duplication was undertaken based on matching author, date, title and page (Bramer et al., 2016). A sample of de-duplicated records was manually checked for accuracy and no issues were identified.

Study selection was performed by the researcher and a second reviewer independently (Balvinder Khambay). References were screened initially by title using the inclusion criteria. Records were eliminated if they were judged to be irrelevant, duplicate or identified in the title as a case report (Centre for Reviews and Dissemination, 2009). Abstracts for the remaining records were retrieved and evaluated based on the eligibility criteria. For those judged to be potentially eligible or where a decision could not be reached, full text articles were obtained. Records found through grey literature searches were included at this stage. Full text articles were reviewed for inclusion and any studies excluded at this stage were indexed in Microsoft® Excel v16.17 (2016) with reason for exclusion.

A standardised data extraction form, modified from the Cochrane Public Health Group data extraction template (Higgins and Deeks, 2011), was used to systematically extract data to improve consistency and reduce bias (Centre for Reviews and Dissemination, 2009). Data extraction was checked for accuracy and consistency by a second reviewer (BK).

The following data items were retrieved for each study:

- Publication information: Author & affiliations, publication date, journal, research date and setting, source of funding
- Study characteristics: Aim, design, type
- Participants: Recruitment data, age, sex, severity of hypodontia, inclusion & exclusion criteria
- Intervention: Type, description, follow-up
- Comparator (if applicable): Type, description, follow-up
- Outcome: Outcome category, measurement tool
- Findings: Statistical methods, key findings, study conclusions

The research quality was evaluated using a modified version of the quality assessment tool developed for evaluation of studies with diverse designs (Sirriyeh et al., 2012). This allowed comparison across study designs using a single scale and determined whether the study design fulfilled the research aim. A three-level scoring system was used for each criterion to score the performance against the descriptor, as a compromise between discriminatory ability and ease of application. A free-text box was included for each scoring criterion to enable support for each judgement to be recorded (Sterne et al., 2014). The scores for the studies were displayed in a table using colour coding to facilitate comparison within and across studies.

No numerical summary measures were appropriate for combining the findings of the studies due to heterogeneity in study design, populations, interventions and outcomes measures. A narrative approach was used for synthesis in stages: 1) Summary of key characteristics of the studies 2) Quality assessment 3) Synthesis of the overall evidence base for specific treatment methods.

3.3.2 Results

Electronic searches were initially completed on 1st September 2015 and updated on 19th December 2016. The PRISMA flow diagram outlining study selection is shown in Appendix Figure 1. Fifty-six research records and eight quality improvement records were included in the review. The purpose of the research studies was evaluation of a single treatment method, comparison between methods or service delivery (Table 3.1). The eight quality improvement reports were clinical audits undertaken in NHS Hospitals to evaluate aspects of service delivery.

The studies and quality improvement reports are grouped by purpose and the intervention, design and key findings are summarised (Appendix Table 6). This summarises the outcomes used to evaluate hypodontia and the evidence underpinning different treatment approaches, including long-term outcomes and complications. The studies evaluated a range of different treatments, including management of primary teeth, orthodontic treatment and tooth replacement methods. The majority of studies (95%) were observational and a quarter did not have a control group to allow meaningful comparison of treatment effect. The most common outcomes were dental health-related and appearance. Two qualitative studies examined patient experience of hypodontia treatment and satisfaction. The audits measured aspects of service delivery including access, clinical efficiency and measures of treatment delivery (treatment time and adherence to treatment). A detailed summary of the characteristics and quality assessment of all included studies is available on request.

The key findings about hypodontia treatment synthesised from the research were:

- Retained mandibular second molar teeth survive 1-20 years in 86-100% cases but the extent of root resorption, infra-occlusion and caries is variable.
- Extraction of primary second molars at the optimal time in cases of premolar agenesis can promote spontaneous space closure and extractions are not associated with any harms (TMD symptoms and marginal bone loss).
- In mild hypodontia space closure is achievable and does not result in any harms (occlusal, periodontal, TMD).
- Placement of resin-bonded bridges (RBB) to replace maxillary lateral incisors improves quality of life compared to unrestored space. RBB last around 5 years on average, but this is increased by operator experience. Failure is most commonly due to bond failure.
- Tooth replacement with implant-retained prostheses increases quality of life through improved function. Survival rates are high but colour match can be a challenge.
- In the post-orthodontic period prior to implant placement bone volume decreases, but the impact on implant placement is less clear. Around 10% cases show relapse in root position that prevent implant placement.
- Tooth transplantation has a success rate of >95% in carefully selected cases with root formation around 50-75%. Biological complications are the most common cause of failure.
- Orthodontic space closure appears to have better patient-reported outcomes and does not cause periodontal or TMD problems.

Table 3.1: Summary of hypodontia-related research studies & audits	
Evaluate effectiveness of single intervention (n=41)	**Maintenance of mandibular primary second molars** (Bjerklin and Bennett, 2000, Bjerklin et al., 2008, Ith-Hansen and Kjaer, 2000, Sletten et al., 2003, Kurol and Thilander, 1984, Hvaring et al., 2014b, Kjaer et al., 2008)
	Loss of mandibular primary second molar +/- space closure techniques (Valencia et al., 2004, Lindqvist, 1980, Northway, 2004, Mamopoulou et al., 1996)
	Orthodontic space closure in mild hypodontia (Rosa et al., 2016, Zimmer et al., 2007, Zimmer and Seifi-Shirvandeh, 2009)
	Resin-bonded bridges (RBB) (Anweigi et al., 2013b, Allen et al., 2016, Garnett et al., 2006, Spinas et al., 2013)
	Denture for children with severe hypodontia (Hobkirk et al., 1989)
	Implant-supported tooth replacement in adults (Hosseini et al., 2011, Degidi et al., 2009, Allen et al., 2017, King et al., 2016, Zarone et al., 2006, Hosseini et al., 2013, Creton et al., 2010, Mangano et al., 2014, Hu et al., 2011, Finnema et al., 2005, Nissan et al., 2011)
	Implant-supported tooth replacement in children (Heuberer et al., 2015)
	Preparatory treatment for implant placement (Novackova et al., 2011, Uribe et al., 2013a, Uribe et al., 2013b, Eliasova et al., 2014, Beyer et al., 2007, Olsen and Kokich, 2010)
	Tooth autotransplantation for mandibular second premolar agenesis in adolescents (Slagsvold and Bjercke, 1974, Josefsson et al., 1999, Jonsson and Sigurdsson, 2004)
Comparison between treatments (n=12)	**Orthodontic space closure vs orthodontic space opening with tooth replacement for absent lateral incisors** (Armbruster et al., 2005, De-Marchi et al., 2014, Pinho et al., 2015, Pini et al., 2013, Qadri et al., 2016, Robertsson et al., 2010, Marchi et al., 2012, Nordquist and McNeill, 1975, Robertsson and Mohlin, 2000)
	Tooth-supported vs. implant-supported tooth replacement (Incici et al., 2009, Krieger et al., 2009, Dueled et al., 2009)
Evaluate treatment experience (n=3)	**Treatment experience** (Tams and Ashley, 2013, Meaney et al., 2012, O'Keeffe et al., 2016)
Audit of service delivery (n=8)	**Access to services in Scotland** (Borrie and Cord, 2014)
	Efficiency of assessment clinic (Walker and Dyer, 2009)
	Treatment delivery (Crawford et al., 2010, Borrie and McIntyre, 2008, Parvizi et al., 2010, Shah and Gwilliam, 2014, Wazani and Sood, 2016)
	Record keeping (Shah and McKaig, 2016)

Appraisal of the study purpose, design and methods identified a number of key issues:

- Lack of clarity in the aim and study design making it difficult to judge appropriateness of the chosen method
- Lack of detail regarding setting limiting the scope to determine generalisability
- Absence of sample size calculation
- Lack of detail about sample selection, increasing the risk of bias in selection
- Limited presentations of hypodontia examined, which created difficulties for applying the evidence to individuals
- High variability in intervention, making direct comparison difficult
- Lack of control group in many studies, limiting meaningful translation of results into likely clinical effect
- High variability in outcomes and outcome measures with few relevant patient-reported outcomes
- No measures related to service delivery, for example access to care and costs

Overall the limitations in the evidence base suggest a lack of patient-centred evidence to support clinicians when discussing treatment options with patients. The inability to combine data from different studies means there is uncertainty about effectiveness across the different treatments. This was a combined effect of high variability in the types of hypodontia studied (number and location of missing teeth) and heterogeneity in research design and outcome measures. Many of the outcomes used are difficult to translate into information that would be accessible to adolescents and parents. Overall the evidence-base was judged to be of insufficient quality to address uncertainty in decision-making about immediate and long-term outcomes and possible adverse effects of hypodontia treatment.

3.4 Analysis of patient information resources

Patient information resources (PIRs) for people with hypodontia were examined to explore the information given to support patients to understand and make decisions about hypodontia care. The aim was to identify and appraise patient information resources (written and online) for hypodontia and its treatment. The purpose was to establish whether information resources are able to help adolescents and parent prepare for decision-making about hypodontia treatment.

3.4.1 Methods

Cross-sectional survey using mixed methods in two stages.

1. Identification of patient information resources through:
    a. A questionnaire to general dentists and three specialist dentist groups
    b. A systematic search of online information
2. Analysis of the content and quality of written and online resources about hypodontia

To identify written patient information resources used by dentists, a sample of general dentists across Yorkshire and Specialists across the UK were contacted. The sample was randomly selected from a publicly available list using a random number generator (Appendix Table 7). Each dental professional was sent information about the study and a short questionnaire, which was developed specifically for the research. The questionnaire was tested for face validity through preliminary completion under observation by clinical colleagues. No formal testing process to assess different psychometric properties was feasible within the research timeframe (Bolarinwa, 2015). Further details of the questionnaire methods are reported in the peer-reviewed publication associated with the study (Barber et al., 2018b). The questionnaire asked dentists about their use of written and online information resources and their opinion on the quality of resources that they use. Copies of leaflets and details of websites used for patient information were requested for the quality appraisal and a stamped addressed envelope was provided.

The systematic search of online information resources was completed on 14th February 2016 to identify resources for quality assessment. Recommended online search methods were followed (Agency for Healthcare Research Quality, 2017). Appendix Table 8 details the search strategy used for data acquisition. The search terms were entered successively into each search engine by the researcher. The first 10 pages of each search, which included 100 hits, were screened for websites. Those meeting the inclusion criteria were 'bookmarked' in the web browser at the time of searching and reviewed for suitability against the eligibility criteria. Details of the eligible resources identified through the dentist questionnaire and systematic online search were indexed in Microsoft® Excel v16.17 (2016) for data extraction and quality assessment.

A data extraction form was used to record:

- Demographics: publisher, year
- Design features aiding literacy: style, text layout, use of images, literacy aids, usability, and accessibility
- Content: purpose, condition, treatment options, treatment consequences and risks, health service, value clarification, decision-making components
- Quality judgment: accuracy, transparency, conflict of interest

Content and quality assessment was conducted independently and in duplicate by two reviewers (SB and HB) and agreement on the final score was reached by consensus. The quality assessment criteria was developed using guidance from DISCERN (Charnock, 1998), IPDAS (International Patient Decison Aids Standard Collaboration, 2005) and tools from similar studies (Hanna et al., 2015, Wong et al., 2003, Winterbottom et al., 2007). The Flesch-Kincaid Grade Level and the Simple Measure of Gobbledegook (SMOG) were used to assess readability levels using an online tool (www.online-utility.org). For pragmatic reasons, authors of the resources were not contacted to establish whether patient input was sought during development.

3.4.2 Results

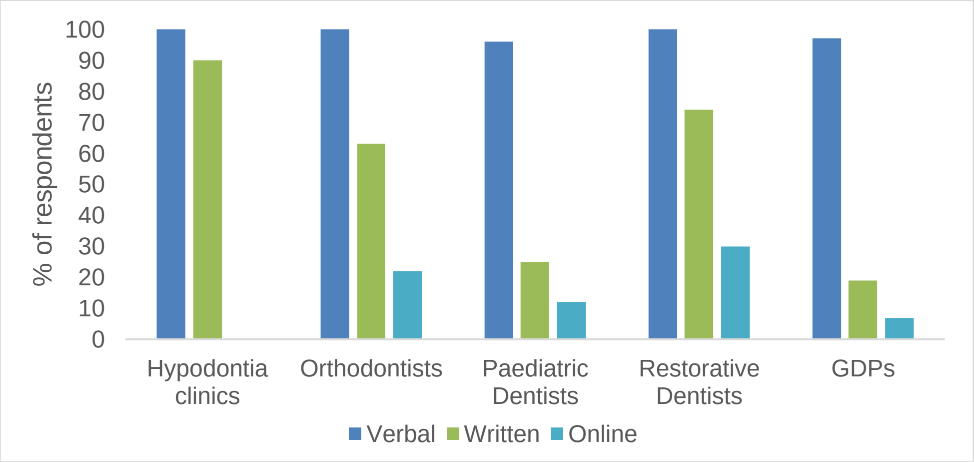
The questionnaire was completed by 297 dental professionals, of whom 269 provided hypodontia care. The response rate was 49% and varied by specialty: orthodontists (56%), hypodontia clinics (55%), paediatric dentists (46%), general dental practitioners (44%), and restorative dentists (32%). Most were NHS or mixed NHS/private dental practitioners, working in primary and/or secondary care at a range of levels. Only eleven respondents returned information leaflets, of which all but two had been previously identified, and no respondents reported directing patients to online resources.

The systematic online search identified 122 potentially relevant websites; 90 were excluded as duplicates and nineteen were excluded after full review (Appendix Figure

2). Thirteen online resources fulfilled the selection criteria. In total, 18 leaflets and 13 online resources were identified for inclusion in the quality assessment. Detailed summary of the resources and quality assessment is available on request.

There was considerable variation in the provision of written information by speciality and provision of written information, particularly online, was low (Figure 3.1). Only 19% of respondents felt current information resources were fit for purpose, while the same proportion felt they were not fit for purpose, and a further 61% were unsure. The majority of respondents (92%) indicated further information resources would be useful.

Figure 3.1: Provision of different types of information by survey respondent



Information resources varied in their purpose and scope (Table 3.2). The majority aimed to prepare patients for consultation or a procedure and only one specifically stated a role in decision-making. The majority of leaflets covered a single treatment, while online resources covered the condition, multiple treatment options and information about service delivery.

Table 3.2: Purpose and scope of patient information resources for hypodontia

		Leaflets (n=18)	Online (n=13)
Purpose of information	Preparation for consultation/procedure	13	13
	Aftercare to aid adjustment or coping	4	0
	Decision-making	1	0
	Patient experience / advocacy	0	0
Scope of resource	Condition, service & treatment	2	11
	Condition only	0	0
	Service only	1	1
	Treatment only	15	1

The quality assessment identified a number of issues in PIRs:

- The purpose of information and target audience was rarely explicitly stated
- There was no statement to suggest people affected by hypodontia had been involved in co-developing the resource, although no additional information about this was sought
- No resources supported decision-making in terms of discussing the decision to be made, the importance of shared decision-making, the patient's role, factors to consider and methods to aid preference elicitation
- In resources about hypodontia, information about the impact of hypodontia, reasons for treatment and timeline and future consequence was absent
- In resources about treatment, information about hypodontia to ensure adequate background understanding was omitted. Information did not adequately explain who treatment is suitable for, alternative treatment options including no treatment, an explanation of how treatment works and information about the health service
- Risks were not described for all side-effects and alternative methods to aid understanding were not used, such as more than one numerical or visual method
- Design features were not selected to optimise design e.g. use of non-white pale background & matt paper, font selection, navigation aids to improve usability
- Images did not always have a clear purpose and there was no evidence that patients were involved in their selection
- Literacy aids were absent e.g. descriptions for numerical data, flow diagrams for processes, diagrams for technical procedures
- Accessibility was limited by non-tailored resources and a high average reading age
- Credibility was limited by a lack of authorship and credentials and statement regarding sources of evidence and areas of uncertainty

The limited availability and use of information resources, and the lack of comprehensive, accessible information within available resources presents challenges for decision-making. Only one resource stated a role in helping with decision-making about treatment, but this was not a decision aid-type resource and had no features to help active decision-making. The resources examined were not deemed to be adequate to allow preparation for decision-making in terms of explaining what is involved in clinical decision-making, the decision to be made, providing the options in balanced and evidence-based way and highlighting areas of uncertainty.

3.5 Observation of Clinical Consultation

Hypodontia clinics provide patients and families with information about hypodontia and its treatment and enable inter-professional discussion around diagnosis and treatment planning (Tams and Ashley, 2013). Clinical consultations were observed to examine information provision, communication and current decision-making practice. The aim was to understand how decision-making is approached by the dental team, to examine the role of different participants and identify evidence of shared decision-making.

3.5.1 Methods

The study involved naturalistic observation and analysis of interdisciplinary clinic consultations for people with hypodontia. An inductive thematic approach was taken to understand the experience of those involved in the clinical consultation (Peat et al., 2019).

Consultations in interdisciplinary clinics in Leeds Dental Institute and St Luke's Hospital, Bradford between August 2016 and April 2017 were observed. In accordance with commissioning guidelines (NHS England, 2015, NHS England, 2018) the clinics serve people with hypodontia of any severity requiring interdisciplinary care. Patients at any stage of treatment can be referred for advice and decision-making. Eligibility criteria for the study is given in Table 3.3. The dental teams varied in composition but usually included Orthodontists, Paediatric Dentists, Restorative Dentists and Dental Nurses.

Table 3.3: Eligibility criteria for observation of clinical consultations

Inclusion criteria	Exclusion criteria
Adolescent ▪ Person with hypodontia (any severity) ▪ Aged 12-16 years inclusive ▪ People undergoing monitoring, pre-treatment planning, at any stage of active treatment or follow up **Parent** ▪ Person with parental responsibility for adolescent with hypodontia	▪ People with teeth missing for other reasons ▪ Hypodontia associated with a syndrome that is likely to have a significant impact on receiving dental care ▪ Significant medical history likely to impact on dental care ▪ Adolescent / parent unwilling or unable to provide valid consent for participation

A purposive sampling approach was desirable to obtain a sample with diversity in experience (Starks and Trinidad, 2007). A maximum variation sampling approach was selected to allow experiences to be compared and contrasted to identify similarities and differences in experience (Palinkas et al., 2015). A sampling framework was developed based on clinical knowledge about characteristics that might be important in clinical decision-making for hypodontia: age, gender, ethnicity, severity of hypodontia and stage of treatment. Observations were conducted until data saturation was reached. To establish whether the patterns observed in the Yorkshire clinics were representative of practice more widely, hypodontia clinics in other hospital units (Sheffield, Manchester and Liverpool) were observed. Due to time restrictions it was not possible to obtain ethical approval for audio-recording but notes were made about the clinic procedures and observations were discussed with the dental teams at each site.

Information about the research and an invitation to participate was given to participants and parents to obtain assent and consent respectively. For clinical observations consent was obtained from the dentists involved in the consultation. Participants were assigned a study number to enable demographic data to be recorded for participants (age; sex; ethnicity; postcode as a measure of socioeconomic status; presentation of hypodontia; stage of treatment; parent present).

The consultation was audio-recorded and additional field notes were taken without any active participation by the researcher. Audio-recordings were transcribed contemporaneously into Microsoft Word v14.7.7 (2011) by the researcher, reviewed with the field notes and anonymised. The framework analysis method was used to collect, manage and interpret qualitative data (Ritchie and Spencer, 1994, Smith and Firth, 2011) (Table 3.4). A coding framework was used to develop themes while maintaining an accurate representation of participant data. An iterative approach was used to allow data to be re-examined for developing themes and a reflective journal was kept for transparency in decisions (Noble and Smith, 2015, Nowell et al., 2017).

Table 3.4: Thematic framework method used for analysing qualitative data

1. Separation of transcript into inter-professional dialogue and dialogue between the adolescent, parent and dentist
2. Development of coding matrix:
    a. Dialogue divided into units based on meaning of conversation (meaningful units)
    b. Codes developed to describe dialogue
    c. Similar codes grouped to develop categories
    d. Development of matrix with codes in columns and cases in rows
3. Refinement of initial thematic framework through discussion with supervisor Professor Bekker
4. Reflection on original data and analytical stages to ensure accurate representation of participant accounts

3.5.2 Results

Five consultations were observed across the two units. The number of observations conducted was lower than expected; however, it was evident that the clinical consultations followed a distinct format and the information given was similar in most cases. This was supported by the visits to other units, where a similar approach to consultation was followed.

Demographics of the participants included in the observations are summarised in Table 3.5. The female: male ratio reflects the gender skew reported for hypodontia. No consultations were observed that involved adolescents at the end of treatment; this may reflect clinical experience that suggests treatment is often not completed until beyond 16 years old. The dental professionals included in the three different interdisciplinary teams consisted of four orthodontists, one paediatric dentist and two restorative dentists.

Table 3.5: Demographics of participants involved in clinical consultation observations

Gender	Female	3	Male	2		
Age	12-13y	3	14-16y	2		
Severity of hypodontia	Mild	1	Moderate	3	Severe	1
Stage of treatment	Pre	2	Mid	3	End	0
Parent attending	Mother	4	Father	1		

Analysis of the consultations suggested three main purposes:

i) Information exchange between the dental team, patient and family
ii) Inter-professional discussion amongst the dental team
iii) Decision-making and treatment planning

In all units, pre-clinic meetings by the dental team were used to discuss cases and formulate possible treatment plans. Dialogue within the consultation was largely inter-professional discussion and information provision from dental professionals to the parent to facilitate understanding of the condition and treatment options. Parental contribution was predominantly confirmation of the accuracy of patient information, while adolescents contributed little other than to outline their concerns and symptoms. Good communication practices were observed throughout the consultations but low levels of active participation by the parent and adolescent were observed.

The decision to be made was stated in most consultations, often with an indication of time scale and acknowledgement of the complexity of the decision. The option of more than one treatment was routinely highlighted but the option and consequence of no treatment was rarely discussed to the same extent. In most cases, the clinical presentation and lack of evidence for specific treatments meant treatment decisions were ones of clinical equipoise, where it would be appropriate to elicit patient preferences when choosing the best option for the patient; however, the importance of patient preferences was rarely stated and the concept of collaborative decision-making was not discussed at all.

Information was provided about the individual's clinical presentation and treatment options, typically with some discussion of potential risks and benefits and the impact of treatment. Information leaflets were often provided to supplement the information provided verbally. Clinicians attempted to elicit patient understanding but there was often little engagement from the patient and no methods for testing understanding were used. There was evidence that parents were unfamiliar with dental language and there were deficiencies in knowledge of treatment options and process. Evidence-based estimates of treatment effectiveness were provided for some treatments but generally scarcities in high quality evidence and a lack of patient-centred methods for explaining research data were reported to be a barrier to further comparison of treatment effectiveness.

Little dialogue in the consultation was attributed to elicitation of patient and family preferences and values. The impact of hypodontia and motivation for treatment were sought during the consultation, most commonly in terms of physical outcome from treatment. Although the importance of preferences in the decision was mentioned, no tools were employed prior to or during the consultation to assist identification of patient and family values and preferences.

Few mechanisms associated with shared decision-making were identified. The key issues identified in current decision-making practice included:

- Inadequate professional awareness of shared decision-making practice
- Patients' lack of awareness of the decision to be made and their role in decision-making
- Patients' lack of knowledge of about hypodontia and lack of familiarity with dental terms, making information exchange challenging
- Language was an obstacle due to inadequate adjustment by health professionals to simplify technical language
- Patients not being informed of all possible options
- Failure to establish patients' and families' preferred level of engagement
- Lack of effective information exchange, including establishing patient knowledge and understanding
- Absence of methods for eliciting patient preference and values
- Organisational barriers in the structure of the care pathway – for example, limited number of opportunities for information exchange, tariff systems in the hospital that favour progress through the pathway
- The time point for agreeing a treatment decision driven by hospital care pathway rather than patient circumstance

3.6 Interviews with adolescents with hypodontia & parents

To capture the experience of people affected by hypodontia, interviews were undertaken with adolescents with hypodontia alone, and with a parent. The aim was to explore adolescent and parent experiences of hypodontia and dental treatment and to understand their experience, beliefs and expectations around decision-making for hypodontia.

3.6.1 Methods

The study used semi-structured interviews with adolescents with hypodontia and their parents to elicit their perspective on hypodontia, treatment and decision-making. An inductive thematic approach was used. Interviews were conducted across two sites, Leeds Dental Institute and Bradford Hospital. Adolescents with hypodontia and their parents were identified using the same eligibility criteria as for the observation of clinical consultations (Table 3.3). Purposive sampling was used to encourage a range of experiences. A preliminary sample size of 14-20 participants was planned with additional recruitment if necessary to achieve data saturation in terms of no new data emerging within the interview and within the emerging themes (Saunders et al., 2018).

Potential participants were identified by clinicians involved in providing dental care for people with hypodontia during treatment clinics, assessment clinics for new referrals and interdisciplinary clinics. Information was provided about the research and for those who agreed, a telephone number was taken to contact one week later to determine willingness to participate. At this point a date was agreed for the interview. Assent and consent were obtained on the day of the interview. Participants were assigned a study number to enable anonymised demographic data to be recorded using a standard data capture form (age; sex; ethnicity; presentation of hypodontia; stage of treatment; parent present).

Interviews were conducted in a private room without disturbance or distractions. The first half of the interview involved the adolescent alone and the parent was invited to join for the second half of the interview. The interview was audio-recorded and transcribed by the researcher. A topic guide was developed to provide a basic structure for the interview (Hancock et al., 2007). The preliminary topic guide was developed using theory around disease perception and clinical knowledge in four concepts (Table 3.6). Leventhal’s Illness Representation Model (1997) was used as a framework to develop questions for understanding patient’s beliefs and expectations about illness. Potential impacts from untreated hypodontia were identified from generic (Slade and Spencer, 1994) and condition-specific (Akram et al., 2011, Hvaring et al., 2014b) quality of life measures. Questions in the management concept were informed by the revised version of the Illness Perception Questionnaire (IPQ-R) (Moss-Morris et al., 2002).

Table 3.6: Concepts in topic guide for semi-structured interviews

Concept	Areas to explore
Condition	Identity Timeline Consequences Cause Control / cure
Management	Individual's role Role of others People involved in treatment Treatment options
Health service delivery	Perceived impact of treatment Important factors in health service delivery Potential barriers Difference in healthcare settings/ providers
Decision-making	Factors in decision making People involved in decision-making Sources of information

An inductive thematic approach was taken using a framework method, as previously described in Table 3.4 (Ritchie and Spencer, 1994, Smith and Firth, 2011). Themes developed around understanding of hypodontia, treatment options, expectations around treatment and outcome, and how decision-making happened.

3.6.2 Results

Eight interviews were undertaken involving a total of 16 participants (8 adolescents and 8 parents) across two units. Contemporaneous data transcription and analysis identified data saturation had been reached at this point, as no further themes were arising. Participant characteristics are summarised in Table 3.7.

Table 3.7: Characteristics for the interview participants

		N=		N=		N=
Gender	Female	6	Male	2		
Age	12-13y	3	14-16y	5		
Severity of hypodontia	Mild	4	Moderate	1	Severe	3
Stage of treatment	Pre	2	Mid	3	End	3
Parent attending	Mother	7	Father	1		

Parents largely perceived the decision-making process to be led by the dental team, with their role being to support their child to ensure the treatment selected was appropriate (Table 3.8).

Table 3.8: Parent's perception of their role in decision-making

"(The Consultant) ultimately looks at her case and makes all the big decisions" (SI01, Mother)
"We are more or less in their hands..... they've not forced us down any particular path but you've got to go on the best advice they have and they're doing it day in day out, so if they think there's two or three options but this one is the most suitable you really are led by your clinicians to say this is the best option for you so we have no reason to say it isn't" (SI03, Father)
"All the head people got together to decide what they were going to do...I'll just go with whatever, she knows her own mind, she can make her own decisions I'll just be there to support her whatever she decides to do" (SI04, Mother)
"I think it would be a case of someone telling us the options and probably going away and researching it a little bit and then sitting down with (patient) and work out what's the best and also taking advice from the consultants because they're the ones that will see on a daily basis" (SI06, Mother)

This was reflected in the attitudes of adolescents, who indicated an advocacy role of parents, and strong guidance from the dental team. Parents and adolescents did not appear to identify their right to determine their level of involvement in decision-making and their equal role in treatment choice (Table 3.9).

Table 3.9: Adolescent perception of decision-making
"I'd ask them both (parent and dentist) and see what they would recommend because I might get the wrong thing and regret it in the future so I'd get everyone's advice" (SI06)
"I would just ask my mum and dad if they knew a little bit" (SI03)
"The dentist has always been really open to her asking questions and she was quite shy, well she was in the beginning but she isn't now, she was quite shy but before we came she would tell me what she wanted to ask and I would ask it but now you just generally just ask" (SI01, Mother)

Recall of the treatment options given during treatment planning was variable (Table 3.10). The option to have no active intervention was commonly dismissed by adolescents, who reported a strong desire for treatment (Table 3.11).

Table 3.10: Adolescent perception of treatment options
"I only went (to specialist practice) once and he made it very clear you have to do this or you have to do that but he didn't really give me the option.... when I came here (hospital) there was more options" (SI01)
"I think (the dental team) told us the different options and we just talked about which was probably the best to take." (SI02)
"I don't know if I had that many options at the start, I don't think I did" (SI06)

Table 3.11: Discussion of option of no treatment
"No... but I definitely would've changed (my teeth)" (SI01)
"No I've always wanted my teeth to be good so I think I've always wanted treatment, I've never really liked my teeth so I think it's good that I will get the help" (SI03)
"No I was always wanting to get them sorted and wanting to go and get what I could done, as soon as it happened I wanted it doing" (SI04)
"Well we had a choice but I wanted teeth that would make me look, teeth that would be more normal" (SI05)
"I can't quite remember but I knew that I didn't have a choice because my mum told me she really regretted not getting braces so I should get them" (SI08)

Participants demonstrated variable understanding about treatment options and there was evidence of misunderstanding, even for those near the end of treatment. For some treatments, participants sought additional information from other sources, most commonly the internet (Table 3.12).

Table 3.12: Understanding options and seeking further information
*"It was quite clear and they weren't too complicated either… I thought it was all going to be more complicated but it's really easy for me to understand...I don't know why I thought it might be more difficult but it's good" (*SI01)
"I got really freaked out and I went on yahoo answers to see if fake teeth look real, I just searched fake teeth" (SI03)
"We've no experience of braces or the bridges so it will be new to us" (SI03, Father)
*"I don't know really, having been there before I've sort of got some sort of idea of what's involved" (*SI05, Mother)
"When they first told me I didn't really understand but I think my mum understood so that's all that really mattered" (SI08)
"I looked at stuff (information) for blocks (functional appliances) because I didn't know anyone that had had them so I went on YouTube and tried to see if anyone else had the same thing or videos of how it works" (SI01)
"I think we got some kind of pamphlet from the post I think or we just picked them up so I looked at some of those" (SI06)

When treatment preference was discussed, participants referred to their motivation for treatment or expected treatment outcome, most commonly in relation to physical outcome or wellbeing related to dental appearance. Sometimes discussion of preference appeared to be understood as choice of treatment (Table 3.13). No participants reported using a decision aid or value clarification tool.

Table 3.13: Adolescent preferences for hypodontia treatment
"Yes, in regard to my missing teeth, it was whether I wanted my teeth moving or just reshaping I think that was about it" (SI01)
"They asked me what my plan would be and obviously, I told them for my teeth to be straight" (SI04)

Previous information provision about treatment options appeared to have developed beliefs about certain treatment options, most commonly dental implants, which may not accurately reflect the evidence base (Table 3.14).

Table 3.14: Beliefs about treatment methods
“I think (implants) is the best solution rather than dentures, isn’t it?” (SI02, Mother)
“(Implants) get screwed onto the bone, yeah that sounds terrifying but apparently I’ll have the nicest teeth in my family” (SI06)
*“I just had the impression that (implants) was the best and permanent option because I don’t know a lot about implants” (*SI02, Mother)
*“Our dentist told us, he says it’s better for him if he gets braces on” (*SI07, Mother)

One adolescent nearing the end of treatment demonstrated greater understanding of their own willingness to accept treatment side-effects for improved outcome (Table 3.15).

Table 3.15: Adolescent understanding of willingness to make trade-off
*“If it was going to have the same result but one was painful and one wasn’t painful I would pick the one that wasn’t as painful, but if one had a really good result and one that was ok but the one that had the really good outcome was more painful, I would still pick that one” (*SI01)

Participants were generally satisfied with their experience of treatment; however, there was little reflection on the experience of decision-making and satisfaction with the choices that were made. One participant near the end of treatment highlighted how her willingness to accept the treatment decision changed over the treatment period, suggesting an initial dissatisfaction with the treatment choice (Table 3.16).

Table 3.16: Adolescent and parent satisfaction with treatment
“I have always been happy with this treatment” (SI02)
“They’ve told me that I’m probably going to have my braces done... then I’ll probably get a bridge.... I’m happy, I’m really happy” (SI03)
*“We’ve done a lot of research on it and asked the questions that we think are right and we’ve had nothing but good experiences” (*SI03, Father)
“When I first got told I should get braces, I was like hmmm I don’t really want them.... but now I think they’re worth it” (SI08)

Overall the interviews found evidence of some misunderstanding about hypodontia and its treatment, a perception that decisions are largely made based on recommendations of the dental team with little integration of patient preferences, and minimal reflection on the decision-making process.

3.7 Review of social media

To supplement the interviews, adolescent and parent perspective was sought through examination of the experience of hypodontia reported in social media posts. Use of social media as tool to explore patient experience is an area of growing interest (Greaves et al., 2013) with potential to enable exploration of patient experience and opinion in real time and in a non-healthcare setting that may provide a more realistic patient perspective (Hanna et al., 2017). The aim was to explore experience of hypodontia care and decision-making from the perspective of the public through a review of social media posts.

3.7.1 Methods

The study followed a cross-sectional survey design in two stages. First a systematic search of social media platforms was conducted to identify eligible records, then relevant records underwent qualitative analysis of the content.

The 10 most popular UK social media websites were considered as possible data sources (Facebook, YouTube, Twitter, Instagram, Google+, SnapChat, LinkedIn, Tumblr, Reddit, Vimeo). Websites that share only video content were excluded (YouTube and Vimeo), as it was outside the scope of this study to analyse video content. SnapChat is a mobile telephone application-based network so it was not possible to access content. LinkedIn was excluded because it is a platform aimed at professional networking. Six sources were included: Facebook; Twitter; Instagram; Google+; Reddit; Tumblr.

Key words relating to hypodontia and common treatment methods were listed based on terminology and consultation with patients and dentists in the Leeds Dental Institute (Appendix Table 9). Scoping searches were performed to test the sensitivity and specificity of the key words. Non-specific terms for hypodontia and all terms for treatment methods returned a high number of irrelevant posts. '*Hypodontia*' and '*oligodontia*' provided a good number of relevant posts and consequently only these terms were included for the search.

Searches were performed by the researcher. Each individual post was treated as a record and the records were indexed into Microsoft® Excel v16.17 (2016) based on source and search term. Duplicates were recorded and removed. Information was recorded for each record: date of posting; information about the user, where available (gender, location, affiliations); word content; any other content e.g. photograph, video. The records were categorised by two reviewers independently and in duplicate to minimise bias using a coding system based on the post theme, user type and target audience to enable relevant records to be identified. Records for content analysis were selected based on pre-defined selection criteria (Appendix Table 10). Word content of eligible records was extracted into Microsoft® Excel v16.17 (2016). Content was divided into meaningful units to develop a coding index, then coded data was summarised and synthesised to develop a thematic framework (Table 3.4).

At the time of the study, the University of Leeds Dental Research Ethics Committee advised no ethical approval was required to collect and analyse data from social media. This was based on no use of private data, no access to private groups by the researcher, no interaction with human subjects and removal of the user name from posts; however, the use of social media as a research resource is increasing and the ethics of using public posts as research data is evolving (Moreno et al., 2013). A subsequent review of best practice guidance for social media use in research highlights that it is not possible to effectively anonymise non-aggregated raw data, as individual posts can be retrieved by searching the social media platform using key words or phrases, and guidance suggests if verbatim quotes are to be used, the user should be contacted for permission (Social Media Research Group, 2016). It was not feasible to contact all the users retrospectively, so all verbatim quotes were removed from the thesis.

3.7.2 Results

Searches were conducted in May-June 2017. One social media site (Instagram) reported more search results than were accessible to view; this was attributed to user privacy settings. It is likely that similar privacy restriction on Facebook limited access to personal posts. After exclusion based on duplication, irrelevance or language 470 records were included (Appendix Table 11).

The 470 records were coded based on author, type of post and theme of post (Table 3.17). The majority of records were captions associated with photographs on Instagram or ‘tweets’ from individuals or dental professionals on Twitter. The most common purpose of posts was to share experience, opinion or information. Instagram was particularly popular with a few users who posted multiple times to share their experience of their journey through hypodontia care. Google+ records were predominantly records consisting of links to other sites or articles. Facebook provided only one useful record. Of the 470 relevant posts, 167 fulfilled the inclusion criteria for the content analysis.

Table 3.17: Relevant records identified from the systematic search of social media (*eligible for content analysis)

		Facebook (n=30)	Twitter (n=168)	Instagram (n=168)	Google+ (n=65)	Tumblr (n=22)	Reddit (n=17)
Author	*Individual affected by hypodontia**	2	53	90	2	22	15
	Dental professional/provider/community	16	63	75	42	0	2
	Academic institute	2	22	0	11	0	0
	Individual/group with commercial interest	8	30	3	12	0	0
Type of post	*Text post**	1	55	0	5	19	17
	*Caption for photograph / video**	14	20	168	7	0	0
	Photograph / artwork / video content	0	1	0	1	3	0
	Link to other content	13	92	0	54	0	0
Theme of post	*Sharing experience / opinion**	1	47	90	1	18	3
	*Information gathering**	1	6	1	1	1	14
	*Information sharing**	23	109	61	60	3	0
	Advertising	3	6	16	5	0	0
Number of records eligible for content analysis		**1**	**41**	**90**	**1**	**19**	**15**

Analysis of social media content provided information about understanding of hypodontia, the impact of hypodontia, experience of treatment, expected and actual outcomes, and information about decision-making. Understanding of hypodontia was largely about diagnosis, aetiology and its consequence and impact. Experience and effect of diagnosis varied, with some reporting it factually while others demonstrated a more emotional reaction. The explanation for the condition commonly related to failure of tooth development, often giving a genetic or familial basis. The most commonly discussed consequence of hypodontia was retention of primary teeth. The eventual failure of primary teeth resulted in symptoms, such as mobility and pain, and “gaps” that lead to a perceived need for treatment. Numerous posts demonstrated the overwhelmingly negative psychosocial effects of untreated hypodontia including impacts on behaviour, such as smiling, wellbeing and social interactions, and concerns about stigma, bullying and poor understanding by others.

Treatment was discussed in terms of types of treatment, length and cost. Many people perceived treatment to be long and complex. Cost was cited as a barrier to care, but mainly by North America-based users. UK users highlighted uncertainty about the impact of hypodontia on eligibility for NHS treatment. Hypodontia treatment was reported to have both positive and negative psychological impacts; people expressed excitement at treatment progress, but also negative experiences at certain points of treatment. Social impacts of treatment included missing out on social events, feeling self-conscious in front of people and experiencing negative reactions from others, often as a consequence of physical effects such as lisping, post-operative complications and discomfort.

Outcomes from treatment were framed as expectations or actual effects. The expected outcomes included physical effects of treatment, hopes to be ‘normal’ or a ‘life-changing’ effect. Actual outcomes included ‘perfect dentition’, improved appearance and most commonly, greater confidence and happiness; however, there were also some negative experiences including the lack of permanence of treatment and need for more treatment, biological complications and technical complications.

Discussion about decision-making related to seeking advice about treatment need and the decision to have treatment. There was evidence that people felt uninformed about the need for treatment, options for treatment and the ‘best’ treatment, and individuals had different reasons for choosing to have treatment. Users sought advice from others and for a minority of users, there was a sense of distrust around the advice received from dental professionals.

3.8 Discussion

3.8.1 Understanding of hypodontia and treatment

The clinical consultations, interviews with adolescents and parents and social media posts suggested that shared decision-making is challenged by poor understanding of hypodontia and treatment, even for those with parents or siblings with hypodontia and those experienced in treatment. Interview participants demonstrated familiarity with dental terminology but a lack of clarity of the meaning of technical terms, suggesting that participants had adopted language to facilitate discussion, but this masked an underlying knowledge gap. Misunderstanding of medical terminology is common (Bagley et al., 2011, Dua et al., 2015) and there has been a drive to reduce jargon (Royal College of General Practitioners, 2014) but health literacy is complex. The capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions depends on knowledge, skills, understanding and confidence of the individual, as well as organisational factors associated with the healthcare system and decisional context (Roberts, 2015).

Information exchange occurs primarily in clinical consultations in hypodontia, but effective exchange is challenged by a number of factors: failure to establish knowledge and a divergence in perspective between the adolescent and parent and the dental team; the volume and complexity of information delivered; uncertainty in information; and the current structure of the pathway. The consultations demonstrated the communication barrier caused by the difference in knowledge and perspective between professionals and lay people. Clinicians failed to elicit pre-existing knowledge from adolescents and parents, and inadequate attempts were made to modify the use of complex dental terms. The resulting effect of a language complexity needs addressing in any future decision support tools.

The interdisciplinary clinical consultations suggested patient information leaflets were routinely used to supplement verbal information, yet the majority of patient information resources lacked high quality, relevant and comprehensive information. Outside the interdisciplinary clinic the use of written patient information reported by dentists was low, particularly general dentists. This limits the capacity for patients to understand their condition, options and prepare for decision-making. A recently published study reports the development and testing of a website to provide information about hypodontia and available treatments (Bharmal and Johal, 2018). The authors report that the website increased knowledge and the majority of respondents found the website useful but the website is not currently available to allow quality appraisal. The study does not report whether the target user group was involved in development, whether best practice guidance was followed and the questionnaire to test knowledge was not validated.

To improve the accessibility and effectiveness of resources, information needs to be presented in a format that increases lay understanding and engagement, provides clarity about sources of information and areas of uncertainty and employs techniques to encourage evaluation of values and reasoning about treatment (Coulter et al., 2006, Charnock, 1998, International Patient Decison Aids Standard Collaboration, 2005). The scope for new approaches in hypodontia, such as interactive online methods to deliver personalised health information (Kaufman, 2010) and online support groups to empower patients (Bartlett and Coulson, 2011), require investigation.

A minority of social media users highlighted distrust in information provided by dental professionals, which might be a reflection of the uncertainty in the evidence base for hypodontia. Social media was used as an avenue for seeking additional, and possibly more accessible, information by people with hypodontia. A key role of professionals in SDM is provision of evidence-based information, yet in hypodontia the lack of high-quality studies, limitations in generalisability and variability in outcomes and outcome measures make translation of research evidence into practice challenging. The absence of patient-centred outcomes reflects previous findings more widely in dentistry (Tsichlaki and O'Brien, 2014, Gwaltney, 2010). Clinical indicators may be used in research as proxy markers for long-term dental health and success, but the underpinning causal model needs to be supported. Heterogeneity in the results across different studies and outcome measures makes an overall estimate of treatment effect difficult. Uncertainty in evidence can be challenging to communicate due to differences in defining and measuring uncertainty, as well as variability in individuals' response to uncertainty (Politi et al., 2007).

The systematic review identified very little UK-based health service research to provide evidence about access to care, quality of care and cost. This limits the scope for understanding the effect of the UK dental care system on the health and wellbeing of hypodontia patients, and the impact of social factors and organisation structures. The interview respondents did not discuss access and generally, the quality of care was perceived as high. This is likely to reflect the sampling method and possibly a bias in those participating in interviews. Social media users did allude to access and cost concerns, but many of these posts arose from people in non-UK healthcare systems so it is difficult to draw conclusions about people's understanding and ability to navigate the NHS hypodontia care pathway.

3.8.2 Approach to decision-making

Observation of hypodontia clinics locally and more widely demonstrated that consultations follow a pattern, reflecting the drive to standardise care to reduce variation in practice and improve the quality of care (Lavelle et al., 2015). Clinics were reported as an opportunity to determine treatment choice; however, framing the choice as 'treatment planning' rather than decision-making emphasises the clinician-led perspective. Effective clinical decision-making has been characterised as pattern recognition, critical thinking and application of evidence, active listening and effective information provision, sharing and reflecting on experiences and effective use of team skills and expertise (Effective Practitioner, 2019). This clinician-centric definition does not recognise the role of the patient and it might be that rephrasing the purpose of consultations from information provision and treatment planning to an opportunity for shared decision-making could create a paradigm shift.

Pre-clinic meetings were used to identify viable treatment options and reduce inter-professional discussion during the appointment; however, the meeting appeared to exclude patients from much of the discussion about options and justification for excluding certain options. This reflects findings of a study in cancer (Hamilton et al., 2016), which articulately describes challenges posed by pre-clinic multidisciplinary team meetings. The desire to conceal professionals' disagreement about options results in the patient being excluded from much of the decision-making conversation. To address this, limited or selected information about the patient can be presented by the clinical team during the multidisciplinary team (MDT) pre-clinical meeting, which can steer the discussion in a particular direction. Finally, when the patient is then presented with the options agreed in the MDT meeting, there is a high risk of bias in the framing of the treatment choice. While the aim of the pre-clinic meetings is to improve pathway planning and ultimately patient experience, patients and their families in oncology report increased distress and/or frustration from their perceived lack of involvement, representation and communication and uncertainty about who to approach for answers to questions (Moremont et al., 2017).

Observations of consultations and the subsequent discussions with the teams involved suggest professional awareness of SDM is low. Currently, SDM does not feature in undergraduate or postgraduate dental training and it is not explicitly defined as a required competency in the General Dental Council's Scope of Practice (General Dental Council, 2013). A lack of familiarity with SDM manifested as failure to invite participation, a lack of explicit statement about the choice and few opportunities for patients and family members to contribute their experience of hypodontia, values and expectations and few mechanisms to promote engagement. The role of the patient and family was not defined and interview participants demonstrated a lack of choice awareness and ambiguity about their role in decision-making. Previous research suggests professionals need to actively engage patients in consultations using patient-centred methods (Street et al., 1995), although more recent research highlights the complex interplay on personal, professional and contextual factors in participation (Street et al., 2005). Education and training is required to promote awareness, promote a positive attitude and teach SDM skills beyond simply good communication; however, consensus about the most effective methods of training is required (Legare et al., 2012).

The desire for further experience-based advice from other people with hypodontia identified on social media suggests a need for additional decision support that has not been adequately provided in the clinical decision-making process. The use of social media networks may also reflect a desire to connect with others with shared experiences, which has been found for adolescents with other long-term conditions (Peat et al., 2018). The extent of discussion about options on social media also emphasises the degree of deliberation that occurs outside the clinical encounter, as suggested in the model of person-centred decision-making (Clayman et al., 2017). The current pathway focusses on the dental encounter, with limited attempts to present options in the wider context of the adolescent's life. No methods were used to establish adolescent and parent values and preferences and there was little recognition of the need to 'trade-off' treatment options in terms of process, outcomes and long-term implications. There are no existing decision-support tools specifically for hypodontia, although two PDAs have been developed and tested for fixed orthodontic appliances (Marshman et al., 2016, Parker et al., 2017). This research suggests a need for evidence-based clinical decision support tools.

3.8.3 Strengths and limitations

Establishing current practice is important to understand trends and attitudes, identify variation in practice, identify where improvements can be made and understand challenges to implementation and uptake (NICE, 2017). The methods, although not exhaustive, were able to provide useful information about current decision-making practice and experiences of adolescents and parents.

3.8.3.1 Evidence synthesis methods

The systematic review of hypodontia literature provided an opportunity to map the existing evidence and understand the strength of the evidence base and areas of uncertainty. Rigour was demonstrated through publication of the review protocol prior to commencing the study (Barber and Khambay, 2015), use of comprehensive search methods for identifying records based on scoping searches, explicit pre-defined eligibility criteria for selecting records, standardised data extraction by more than one reviewer independently, standardised quality assessment using validated tool (Higgins and Green, 2011, Centre for Reviews and Dissemination, 2009, Petticrew and Roberts, 2006). The review has been published in a peer-reviewed journal (Barber et al., 2018a). While the review fulfilled its purpose for this research, the breadth of the review question, challenges in evidence synthesis and the low quality of included studies make it difficult to provide an evidence-based summary of treatment effectiveness to inform clinical practice. The review has, however, formed the basis for recommendations about future hypodontia research.

The methods used in the analysis of patient information resources were not novel but this is the first such study in hypodontia and the comprehensive identification of resources and objective quality assessment was important to understand patient literature for hypodontia. A number of limitations are acknowledged: the questionnaire to obtain information for dental professional was not validated, few information resources were returned by professionals so it is possible other patient resources were omitted, there is likely to be a self-selection bias in those who responded and resource authors were not contacted to establish patient contribution. On balance, it is not expected these limitations would significantly affect the findings but these are considerations for future similar studies. The greatest limitation of the study was failure to seek adolescent and parent opinion about the quality and usefulness of information resources. This information would be valuable to inform development of future resources; however, it would require a method that encouraged in depth examination of patient opinion to guide worthwhile improvements in resources. A co-production approach to patient information resource development for hypodontia has been included in the recommendations for future work.

Social media provided an opportunity to understand patient experiences in real-time and with an honesty that might be restricted during formal interviewing. In this study social media provided a useful additional perspective to other methods, however, the use of social media is not without challenges. First, and most importantly, were the ethical considerations discussed in Section 3.7. Consideration also needs to be given to the validity and generalisability of conclusions drawn from social media data; there is a risk of misinterpreting information when taken out of its original context and the perspectives provided are likely to be limited. Social media users are not representative of the general population (Mellon and Prosser, 2017), users are more likely to share negative experiences or opinions (Primack et al., 2019), and the so-called ‘echo chamber’ effect can amplify certain perspectives (Garimella et al., 2018).

3.8.3.2 Qualitative methods

General principles of qualitative methods were followed to promote transparency, credibility and rigour (Tong et al., 2007) and to enhance the truth value, consistency and neutrality of findings (Noble and Smith, 2015); however, some limitations that affect reflexivity require consideration. The researcher is a dental professional with experience of treating adolescents with hypodontia who at the time of data collection was new to qualitative methods; these factors increase the risk of the researcher unconsciously influencing participants during data collection, particularly in the interviews, and introducing bias during data analysis. Divergence in the roles of interviewers in clinical and research roles has been emphasised as a challenge for new researchers, particularly where the clinical situation requires elicitation of specific information to facilitate care decisions and the research requires freedom to explore topics of importance to the participant (Hunt et al., 2011).

The researcher is female, White and in her mid-30s; this did not visibly affect interaction with the interview participants but may have influenced who opted to participate in the interviews. Two participants for the interviews were known to the researcher through previous clinical contact, so these participants were aware that the researcher is a dentist and part of the care team. Initially there was concern this may reduce the candidness of interviews but actually familiarity appeared to put the adolescent at ease to discuss their experience more openly. The teams involved in the clinical observations were all known to the participant and were aware of the research topic, and there was a chance this might influence behaviour; however, attendance at the clinic prior to the research commencement and observation during the research identified no noticeable behaviour change.

Steps were taken to reduce bias including formal training prior to commencement of the studies, support and guidance from experienced supervisors and use of reflective practice. A reflective journal was kept to promote transparency in decisions (Noble and Smith, 2015, Nowell et al., 2017) and clinical colleagues were selected to discuss emerging themes and concepts to provide an opportunity for triangulation; however, more extensive peer debriefing with an independent researcher would improve the credibility of the findings and in future this would be strongly considered (Nguyen, 2012). Transcripts were not returned to participants due to time and feasibility constraints and data coding was undertaken by the researcher alone although discussed and checked with the supervisory team. This may also reduce the credibility of the results.

Observations of consultation were limited by the small number of participants, reducing the generalisability of findings. The number of participants was adequate for the purpose of this research, as the method was combined with other approaches; however, there would be value in repeating the observations with more participants and across more sites. It may also be worthwhile for a non-dental researcher to participate in observations to provide an alternative perspective; this was not feasible in this study due to time restrictions. A formalised, objective measurement tool could be used, such as a communication measure (Epstein et al., 2005) or the OPTION scale for measuring involvement (Elwyn et al., 2003).

3.8.4 Implications for clinical practice

Adolescent and parent understanding of hypodontia and its treatment is inadequate

Limitations in adolescent and parent understanding around hypodontia and its treatment, even for those experienced in treatment, impacts on the scope for adolescents, parents and professionals to participate in meaningful discussion around treatment options and outcomes from treatment. Information resources are not adequate to prepare people for consultation and there is evidence that people are seeking information and support from alternative sources, such as social media, to address knowledge gaps and help navigate care. Better information is needed including an improvement in the evidence base to address uncertainty about treatment options, and high-quality, evidence-based accessible patient information to support preparation for decision-making.

The current approach to clinical consultation does not promote collaborative decision-making

Adolescent and parent participation in decision-making in hypodontia care is low. This appears to be a combined effect of failure by dental professionals to recognise and define the choice context and decision-maker roles, absence of mechanisms to promote engagement and consideration of patient values and preferences, low professional awareness of collaborative decision-making processes and organisational barriers. Adolescents and parent seem unaware of the opportunity to participate in decisions and the value of their expertise. Changes in practice are required to promote a shared decision-making approach, including training and awareness-raising for dental professionals, evidence-based tools and processes to encourage patient participation, evidence-based approaches for integration of patient preferences into decision-making and methods to encourage reflection on the decision-making process to encourage continued learning and development.

3.8.5 Implications for future research

Research to improve information to support decision-making:

- It would be beneficial to use a stakeholder consensus-building method to development of a core outcome set for hypodontia to be used in research. This would improve the quality and relevance of the evidence base, particularly inclusion of patient-reported outcomes (PROMs).
- A careful examination of methods for improving professional-patient communication is required, particularly presentation of complex information, such as uncertainty and risk, to adolescents and parents. This aligns with the Oral and Dental Health Research priorities (James Lind Alliance, 2018).
- Evaluation of patient information resources from a patient and family perspective and understanding of adolescents' preferred method for information provision is required to inform co-development of information resources with adolescents and parents, for adolescents and parents.

Research to further examine decision-making practice in hypodontia care:

- Further examination of the purpose and content of clinical consultations for people with hypodontia in other settings is warranted to triangulate the findings from this research and increase generalisability.
- Examination of professionals' awareness and attitude to shared decision making is an important first step in identifying barriers to implementation of changes.
- Evaluation of shared decision-making education and training for dental professionals, patients and families is required to identify the best approach.
- There is value in the development and testing of evidence-based decision support tools including methods to promote choice awareness, methods to promote engagement, decision interventions e.g. PDAs and VCMs.

Application of methods to other areas of dentistry:

- These methods can be replicated in other areas of dentistry to understand patient experience of decision-making in other contexts.
- Examination of decision-making practice in other dental care settings, such as primary care dental practice, is required to identify scope for improvements in processes, pathways and to identify education and training needs more broadly across dentistry.

Chapter Four:

Preference Elicitation Instrument Development and Testing

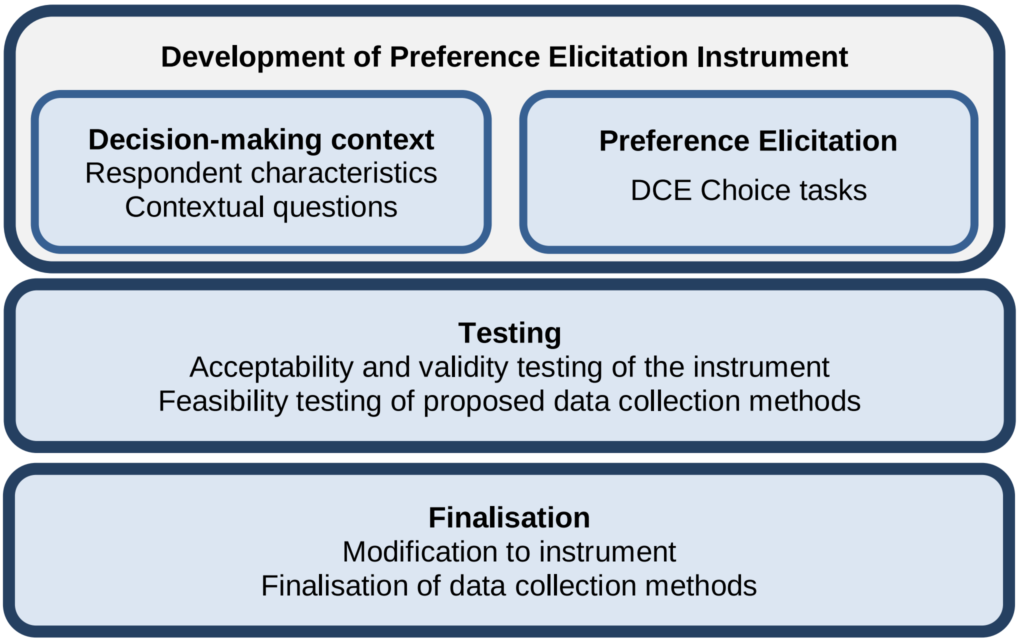
Chapter Four:
Preference Elicitation Instrument Development and Testing

The previous chapter examined hypodontia care to understand the decision context and current practices in decision-making. This chapter focuses on the development and testing of a preference elicitation instrument for use with adolescents, parents and adolescent-parent dyads. The first part of the instrument aimed to establish the decision-making context, including respondent characteristics and experiences of hypodontia, dental care and making decisions. The second part included Discrete Choice Experiment (DCE) choice tasks to elicit preferences for attributes of hypodontia care. The suitability of the tool and proposed method for data collection methods were tested in a pilot study using a combined qualitative and quantitative approach. The results of the pilot study were used to modify the preference elicitation instrument. The key implications for clinical practice and future research are outlined.

4.1 Introduction

This stage of the research developed and tested a two-part instrument consisting of questions to establish participant experience of hypodontia and decision-making to give the context to the research, and Discrete Choice Experiment (DCE) choice tasks for preference elicitation (Figure 4.1). The overarching purpose of this research was to measure adolescent and parent preferences and decision-making to provide information to improve clinical decision-making practice. The ambition was to obtain evidence that could guide patient-centred information provision and identify important aspects of treatment to incorporate into future decision support tools. Preference elicitation was not aiming to determine which specific treatment is ‘best’; this is a clinical decision that will depend on individual patient characteristics and service factors. For this reason, alternatives in the choice tasks were not intended to represent existing or proposed treatments, but instead provide information about which aspects of treatment are most important and how adolescents and parents decide between options.

Figure 4.1: Development and testing of the preference elicitation instrument



The literature review (Chapter 2) and the methods used to examine current decision-making practice in hypodontia (Chapter 3) provided some information about the choice situation, the treatment options available, how options are presented to adolescents and parents. This is important for conceptualisation of the choice process when designing the preference elicitation tool (Lancsar and Louviere, 2008). The intention of the first part of the instrument was to further examine the decision-making context from an adolescent and parent perspective to understand the factors that might influence choice. Understanding which individual characteristic and parts of previous experience affect decision-making and preferences is important for translating the findings into clinical practice.

DCE was the chosen preference elicitation method for this research because the trade-offs required accurately reflect the complexities of decision-making in hypodontia. DCE provided an opportunity to examine whether trade-off tasks help adolescents and parents to think about the alternatives, risks and benefits. The steps for DCE development are outlined in Section 2.3.4. Valid preference elicitation is predicated on including attributes that are important and relevant to the target group and are able to answer the research question (Abiiro et al., 2014). The mixed methods described in Chapter 3 facilitated identification of potential attributes of hypodontia care from a range of different perspectives. Hypodontia treatment is complex so it was not possible to include all attributes in the DCE tasks, so a selection process was required to choose those most relevant and important. A number of attribute selection methods were considered (Helter and Boehler, 2016). Some of the more complex quantitative methods, such as the repertory grid technique, laddering and Delphi method were excluded due to lack of evidence about their suitability for adolescents. The nominal group technique (Hiligsmann et al., 2013) was excluded because preliminary work found adolescents were reluctant to attend group sessions. The chosen method included a quantitative rating and ranking survey combined with a qualitative consultation approach involving adolescent-parent and expert stakeholders (Coast et al., 2012, Janssen et al., 2018, De Brun et al., 2018, Ryden et al., 2017). Consultations also provided an opportunity to simultaneously develop appropriate framing and investigate attribute-levels proposed from clinical and research evidence.

To date, DCE has been primarily used with adults so information about design decisions specifically for adolescent respondents is scarce. To manage this contributors from the target group were involved in instrument development and testing phases where possible (Jones, 2018). In DCE development piloting is key for testing the number and validity of attributes and levels, the construction of choice tasks and proposed preference elicitation method and other non-DCE components of the instrument (Bridges et al., 2011). ‘Think Aloud’ feedback (Koskey, 2016) has been successfully used in DCE pilots with adults (Ryan et al., 2009, Whitty et al., 2014, Cheraghi-Sohi et al., 2007, De Brun et al., 2018) and adolescents (Diorio et al., 2012). It has been shown to support assessment of internal validity and acceptability (Whitty et al., 2014) and provide more detailed information about respondent’s cognitive processes during task completion, which can explain what may appear to be irrational choices (Ryan et al., 2009, Cheraghi-Sohi et al., 2007).

In this study the pilot study provided an opportunity to preliminarily examine decision-making and preferences to formulate hypotheses for testing in the subsequent stage. Think aloud methods was used to assess the validity of the instrument and approach and explore the decision-making processes underlying choices. The preference data collected was also examined to support decisions about inclusion and exclusion of attributes and to guide improvements to the DCE experimental design (de Bekker-Grob et al., 2015). A number of approaches are available for analysis of preference data; this is discussed further in the subsequent chapter.

4.2 Aims and objectives

The aim of this stage of the research was to develop and test an instrument for measuring adolescent, parent and adolescent-parent dyads' preferences and decision-making in hypodontia care.

The objectives were:

- To develop a preference elicitation instrument using best available evidence, principles of DCE design and input from key stakeholders
- To test the validity and acceptability of the preference elicitation instrument
- To test the feasibility of using the preference elicitation instrument to examine decision-making processes
- To use the findings to finalise the preference elicitation instrument and survey methods for large scale data collection
- To summarise findings to identify the key implications for clinical practice and future research

4.3 Methods

The preference elicitation tool was developed and tested using mixed methods. A rigorous and evidence-based approach was taken to optimise the instrument and methods for use with adolescents and adolescent-parent dyads.

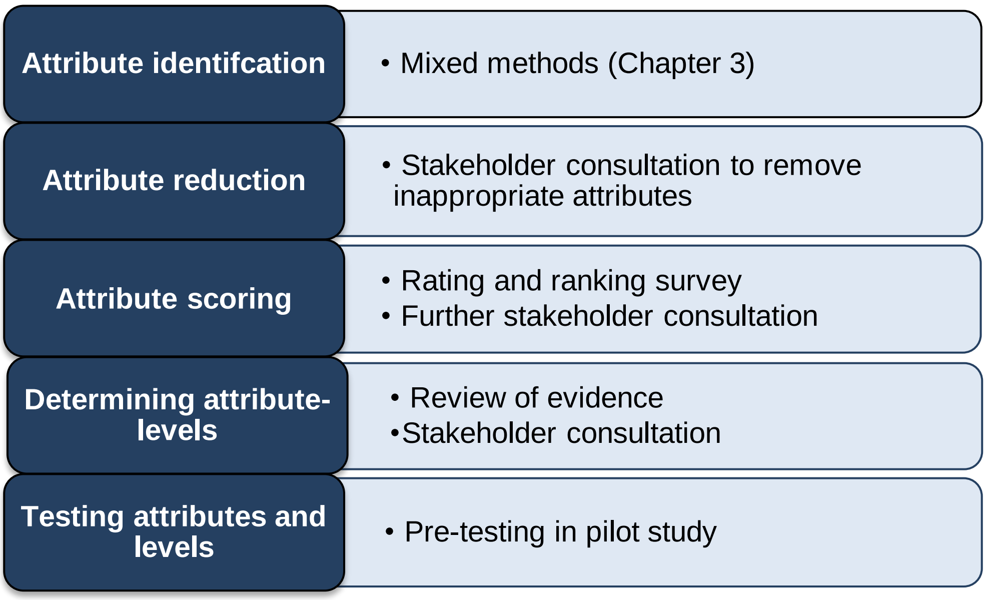
4.3.1 Identification of relevant contextual questions

The first part of the tool included questions to understand the background in which decision-making and preference elicitation were occurring. This included questions about respondent characteristics and factors that may influence decision-making. Evidence gained in Chapter 2 and 3 were used to develop questions, with further literature searching and PPI contributor input as needed.

4.3.2 Attribute development

Attribute development used combined quantitative and qualitative methods to select attributes that were important and relevant to the target respondent group (Helter and Boehler, 2016). Based on evidence from previous DCEs in healthcare (Clark et al., 2014, de Bekker-Grob et al., 2012, Soekhai et al., 2019) the goal was to include no more than six attributes with 2-3 levels in the final DCE Choice tasks. Attribute development was undertaken in stages (Figure 4.2).

Figure 4.2: Stages in attribute development



Attributes were identified from the data gathered in Chapter 3. The mixed methods were chosen to facilitate comprehensive attribute identification from different stakeholder perspectives. Attributes were extracted separately from each study and indexed in Microsoft® Excel v16.17 (2016). Attributes were synthesised to develop a framework for grouping attributes with a common theme.

Preliminary attribute elimination was based on criteria developed from the literature (Coast et al., 2012) to eliminate:

- Duplicates: attributes with sufficient overlap to be judged to describe the same concept
- Inappropriate: attributes that do not fulfil the conceptual framework for the random utility theory – those that do not describe hypodontia care, that are not relevant to the target respondents or that are judged to be intrinsic to personality
- Ambiguous: attributes where the evidence base is inadequate or ambivalent
- Indiscriminative: attributes related to generic issues that could occur equally with any treatment

Attribute reduction involved consultation with stakeholders. Three pairs of PPI contributors were included in the consultation to ensure validity, relevance and importance of attributes (Klojgaard et al., 2012):

- Siblings with hypodontia: Martha (15-year-old female, missing four posterior teeth and mid-treatment) and Beth (13-year-old female, missing two anterior teeth and pre-treatment)
- Adolescent-parent dyad: Natasha (14-year-old female, missing two anterior teeth, post-orthodontic treatment) and her mother
- Adolescent-parent dyad: Abbie (15-year-old female, missing two anterior teeth, post-treatment) and her mother

The expert group included clinicians who provide dental care for people with hypodontia, who advised on the suitability of attributes in the specific context of hypodontia and its management, and methodologists with expertise in stated preference experiments, decision-making and engaging with patient contributors. These stakeholders were included to check the plausibility and accuracy of attributes and levels for future translation into clinical practice (Helter and Boehler, 2016).

An iterative approach was used to refine the raw data into a long list of attributes relevant to hypodontia care. During planning it became apparent that it was not feasible for contributors to meet collectively due to practicality issues around location and school and work commitments; instead pairs of contributors attended together to promote discussion and provide some support for the adolescent to make the consultations less intimidating. Attributes were presented to each group and discussed to explore their meaning, relevance and perceived application to hypodontia care. The meetings with PPI contributors were used to explore the perceived relationship between attributes within and across attribute concepts. Notes were made throughout the consultations and used to revise and modify the attribute framework. There was strong agreement in the opinions expressed by the stakeholder groups despite the consultations being largely conducted separately. A decision log was used to record and justify decisions.

Two methods were used to reduce the long-list of attributes to the target range of 6-8 for piloting: 1) Cross-sectional survey to rate and rank attributes (Salkeld et al., 2003, Morgan et al., 2000); 2) Further stakeholder consultation. The cross-sectional rating/ranking survey was undertaken with adolescents with hypodontia and parents of adolescents with hypodontia. The survey was intended to guide further consultation around attribute selection rather than definitively selecting the attributes. Participants were recruited from a Hypodontia Support Facebook group where members had previously indicated that they would be happy to provide advice or participate in research about hypodontia. At the time of the survey there were 98 members from around the world but only those in the UK were eligible to participate. Due to the scarcity in reporting of attribute selection methods there is little precedent for establishing ideal sample size for surveys for ranking or rating attributes. The survey aimed to recruit as many people as possible, with at minimum sample size of 30 participants.

A post describing the research and inviting participation was added to the group's homepage with a link to the online survey. Inclusion was limited to UK-based adolescents (aged 12-16 years) and parents of adolescents with hypodontia. Participation was voluntary and anonymous. Consent for participation was explicitly sought at the start of the questionnaire. A £5 e-voucher was offered to each participant who completed the survey. Respondents were required to provide an email address for the e-voucher to prevent multiple completions. The survey was live for 4 weeks with reminder posts placed at the end of the first, second and third week.

PPI contributor input was sought during development of the rating/ranking survey, particularly regarding response format, phrasing for describing the attributes and the level of burden. Adolescent contributors strongly felt that they would find it difficult to rank all attributes and reported that they would prefer to rate the attributes individually or pick most and least important from a list. Rating of attributes independently carries a risk of central tendency scoring leading to poor discrimination (Wolf et al., 2016), so two complementary response formats were used in the questionnaire:

- Rating exercise using a 5-point scale (1=not important, 5 =very important) for 16 attributes related to during treatment process and 11 attributes related to treatment outcome
- Ranking exercise to select the 6 most important attributes for treatment process and 6 most important attributes for treatment outcome

The questionnaire was converted into an online format using an online survey tool (OnlineSurvey, Jisc). Basic demographic information was collected (age, gender, location, severity and location of hypodontia, treatment experience, respondent group (adolescent, parent, dyad)). Logic functions were used to create adolescent and parent versions. The online survey was tested by patient and parent contributors prior to distribution to ensure attribute phrasing was valid, the format was user friendly and there were no difficulties in the process (e.g. accessing the link, completing the survey).

Data was collated in Microsoft® Excel v16.23 (2016) and summarised using three methods:

1. The 5-point Likert score for rating was used to categorise individual's response for each attribute as Not Important (for scores 1-2) or Important (for score 3-5). Aggregate scores summarised the number of times the attribute was rated as important.
2. A mean rating for each attribute was calculated from the 5 -point score
3. The number of times an attribute was ranked in the Most Important

The findings from the rating/ranking survey were used to facilitate further consultation with stakeholders to reach a short list of attributes for piloting. The consultations were also used to finalise the description of the attributes and the range of attribute-levels. This focussed on identifying meaningful wording and levels to ensure terminology and descriptions evoked the desired meaning for the target respondents (Coast et al., 2012). Levels were defined using information obtained during attribute identification combined with clinical information. A Microsoft® Excel spreadsheet was used to index the data describing potential levels and findings were synthesised to present to stakeholders for discussion. Stakeholders checked their relevance, realism and range.

4.3.3 DCE choice task development

DCE choice tasks were developed following best practice guidance, with consideration of task construction, experimental design, the method for preference elicitation and the design of the instrument as a whole (Bridges et al., 2011, Hauber et al., 2016, Marshall et al., 2010, Reed Johnson et al., 2013, Lancsar and Louviere, 2008) (Table 4.1). Evidence from previous DCE studies was considered in design choices and a supervisor with experience in DCE provided advice (David Meads). The rationale for design decisions was recorded throughout to provide transparency.

The contextual questions and DCE choice tasks were combined to create the preference elicitation tool for testing in the pilot study. Information to describe the survey and attributes and instructions for completing the choice tasks were developed with PPI collaborators. The preference elicitation instrument was programmed and administered by the survey company Accent (http://www.accent-mr.com) following design instructions provided by the researcher.

Table 4.1: Considerations in in DCE choice task development

Choice Task construction	Full or partial profile Number of alternatives Inclusion of opt-out Labelling
Experimental design	Choice and properties of experimental design Number of tasks
Preference elicitation	Motivation and explanation Elicitation format Qualifying questions
Instrument design	Layout and design Level of burden

4.3.4 Pilot study to test instrument and data collection method

The pilot aimed to determine whether i) the contextual questions were able to elicit meaningful information about factors that are important to decision-making; ii) the attributes were important and relevant to participants; iii) DCE choice task design promoted choices that reflected genuine preferences; iv) the instrument was acceptable; and v) the data collection method was feasible. This information was used to modify the instrument and methods for the subsequent stage. The pilot also provided some preliminary information about decision-making and preferences.

The pilot involved a cross-sectional survey using think aloud methods with some verbal probing (Koskey, 2016). The sample population was adolescents (aged 12-16 years) with hypodontia of any severity at any stage of treatment. Parents or other family members who were important in decision-making were also recruited. The pilot was conducted in two hospital Orthodontic Departments in Yorkshire (LDI and SLH). The target sample size for the pilot was 20 participants, including 10 adolescents and 10 parents. This was approximately 10% of the expected final sample size, which was expected to allow adequate feedback on the preliminary survey without impacting on the number of potential participants for the main study (McGrady et al., 2018). Each participant answered eight choice task questions, providing a total of 160 observations. This was judged to be adequate to provide quantitative data for statistical testing to select the final attributes and estimate prior parameter estimates to inform the experimental design (de Bekker-Grob et al., 2015).

Potential participants were identified by the treating clinician. Brief verbal information was provided alongside the written study information sheet and permission sought for the researcher to make contact via telephone or email 1-2 weeks later. The researcher contacted the potential participant to further explain the research and if the participant agreed, a convenient date and location was arranged for survey completion. Consent was obtained at the time of data collection. The adolescent and parent chose whether they wished to complete the survey together or separately.

Study information sheets and consent forms were developed for the pilot study with input from adolescent-parent contributors. The survey was presented to the study participants using a laptop in a private room. Participants were asked to complete the survey whilst providing ‘think-aloud’ feedback (Koskey, 2016). The researcher made notes based on the verbalised thoughts and asked questions where clarification was needed or there were opportunities to gain further feedback on aspects of the survey. The interviews were audio-recorded to allow retrospective clarification of field notes during analysis if required, however, audio-recordings were not transcribed. The participant was encouraged to ask for help if they could not understand the survey and any uncertainties were recorded. Survey responses were automatically collected from the survey, collated and anonymised by Accent.

After the pilot survey had been completed participants were asked specific questions relating to instrument design (layout, language and content), attributes and levels (relevance, importance and any missing attributes), explanation and presentation of the choice tasks and level of burden (time taken to complete survey, complexity of the DCE task). The field notes and any additional information from the post-survey interview were reviewed contemporaneously to allow iterative modification and testing of the survey. The choice tasks were not modified throughout the pilot. Feedback was organised into categories to create evidence to address the aims of the pilot (Table 4.2).

Table 4.2: Categorisation of data from *'Think aloud'* feedback in the pilot study

Suitability of method	Scope to elicit preferences from adolescents
	Scope to promote discussion between dyads
Instrument design	Layout, presentation and usability
	Content and language in each section
	Acceptability and relevance of contextual questions
	Level of burden
Construction and understanding of tasks	Presentation of DCE Choice tasks
	Understanding of Choice tasks
Preference elicitation	Elicitation format
	Use of qualifying questions
Validity of attributes and levels	Understanding of attributes and levels
	Importance of attributes

Preference data generated from the survey responses was analysed using conditional logit model with main effects and dummy coding. The conditional logit (CL) relates choices to attribute-levels defining the alternatives based on the assumption that error terms are independent and identically distributed across observations and that there is no preference heterogeneity across respondents (Adams et al., 2015). While these assumptions may not be true, the model was judged to be adequate for the pilot based on a previous pilot study (Bray et al., 2016). Preference weights (regression β-coefficients) for each attribute-level were calculated for adolescents, parents and both groups combined. A simple additive linear model was used that assumed no interaction between attributes and an equal effect across levels (Ryan et al., 2001):

$$\Delta B = \beta_1 \text{Discomfort} + \beta_2 \text{Appointments} + \beta_3 \text{Wait} + \beta_4 \text{Problems} + \beta_5 \text{Function} + \beta_6 \text{Appearance} + \beta_7 \text{Cost} + \text{error}$$

> where β is the preference weight associated with the attribute level within an attribute, compared to the reference attribute level.

β-coefficients were reported relative to the reference level, with 95% confidence interval. Preferences were considered statistically significant when the p value for an attribute level was <0.05. The sign and magnitude of the β-coefficients indicated the direction and strength of preference respectively.

- Preference weight >0: attribute-level is preferred over the reference level of the same attribute
- Preference weight <0: reference level is preferred and the attribute-level results in loss of utility

Preference heterogeneity between parents and adolescents was examined by segmenting the sample based on respondent group.

4.4 Results

4.4.1 Questions to establish the decision-making context

Demographic data questions were included to allow description of the sample and identification of possible characteristics that may influence preferences. Age, gender and ethnicity were judged to be of interest, as clinical experience suggests these factors may be important in decision-making behaviour and preferences. The number and location of missing teeth was requested to allow classification of the presentation of hypodontia, as quality of life studies have found mixed results for the impact of hypodontia severity and specific missing teeth (Appendix Table 1).

In the adolescent version, participants were asked their plans for education after completion of school as an indicator of level of education and entitlement to free school meals as an indicator of income. In the parent version, participants were asked their educational level and household income bracket and the number of adults and children in household to enable income data to be estimated. Household income provides context for interpreting willingness to pay estimates (Rowen et al., 2018). It was hypothesised that level of education may contribute to variance in preferences, but this has not yet been examined for decision-making in the dental setting.

Understanding of the health condition is suggested as a contributing factor in decision-making processes and for satisfaction with decision-making (Sepucha et al., 2013). To establish respondents' knowledge questions were developed based on the domains of the Illness Perception Questionnaire (Weinman et al., 1996). Questions and answers were devised from the content of patient information resources and using existing clinical knowledge. For ease of scoring, a dichotomous answer format was used (True / False) with an opt-out (Don't know) to minimise guessing (Appendix Table 12). PPI contributors confirmed the content and readability of questions. Based on previous approaches for self-rating health (Cavallo et al., 2015), adolescents were asked to rate their general health and dental health on a 4-point scale (Excellent / Good / Fair / Poor). Parents were asked to rate the health of their child. No information was given about the meaning of general health, dental health or the rating categories.

To inform questions about beliefs about dental treatment, existing questionnaires were examined. The Dental Beliefs Survey (Kvale et al., 2004) was not appropriate as it measures interpersonal relationships between the patient and professional. Other attitude and belief measures used in healthcare were explored (Webel, 2015) and after discussion with PPI contributors, the Beliefs about Medicine Questionnaire (Horne et al., 1999) and Beliefs about Surgery Questionnaire (Francis et al., 2009) were used as a basis to develop questions to explore beliefs about dental treatment. Four statements were selected based on stakeholder guidance and participants were asked to indicate to whether they agreed using a dichotomous scale (True / False) (Table 4.3).

Table 4.3: Questions to assess beliefs about dental treatment
Dental treatment is the only option for my child’s missing teeth
Having dental treatment now for missing teeth will reduce my child’s need for more dental treatment in the future
I worry about the risks of dental treatment for missing teeth
Dental treatment for missing teeth can result in new dental problems

Existing tools for measuring beliefs about decision-making were reviewed (Section 2.2.8). The Decisional Conflict Scale (DCS) is a validated 16-statement questionnaire that aims to measure individual’s comfort with a decision for choices that involve risk, loss, regret or challenge to personal life values (O'Connor, 1995). The DCS has been shown to be valid for measuring decisional conflict in surrogate decision-makers, which included parents on behalf of children (Pecanac et al., 2018). The SURE tool is a shorter alternative to the DCS, which was developed and validated for use in clinical practice (Legare et al., 2010, Ferron Parayre et al., 2014). The tool contains four simple questions relating to feeling certain, informed, supported and aware of own values, with a dichotomous response option. These questions were selected for inclusion in the survey alongside a question to identify important people in decision-making (Table 4.4).

Table 4.4: Questions to assess beliefs about decision-making

The following people help me choose treatment for my missing teeth:

- Parents / guardian and family
- Dentist
- Friends
- Other people with missing teeth

I feel sure about the best choice for me (Yes / No)
I know the benefits and risks of each option (Yes / No)
I am clear about which benefits and risks matter the most to me (Yes / No)
I have enough support and advice to make a choice (Yes / No)

Dental anxiety, defined as an adverse psychological response to a dental stimulus that is perceived to be harmful but that is not immediately present, is estimated to affect approximately 5-20% of the population to some extent (Seligman et al., 2017). Dental anxiety has been identified as one of the strongest determinants of access to dental care (Longman and Ireland, 2010). It was hypothesised that anxiety for general dental treatment and hypodontia-specific treatment may affect preferences. A number of common dental anxiety measures were considered. Corah's Dental Anxiety Scale (DAS) was the first such scale developed (Corah, 1969) and later validated (Corah et al., 1978), with four questions about psychological and physiological reactions to dental situations. Subsequent versions including Modified Dental Anxiety Scale (MDAS) (Humphris et al., 1995) and the Modified Child Dental Anxiety Scale (MCDAS) (Wong et al., 1998) have been shown to be valid and reliable screening tools. The Dental Fear Survey (DFS), first published in 1973, contains questions to assess a broader range of dental fear components including anticipation and avoidance, physiological responses, and fear of specific dental situations (Kleinknecht et al., 1984). These tools were judged to be too long; however, the tools were used to inform discussions with PPI contributors and clinicians to devise two simple statements about anxiety. These statements were provided with a 5-point scale (Strongly Agree – Strongly Disagree) (Table 4.5). The term 'normal dentist' was used to refer to a general dental practitioner based on language in the interviews, observation of clinical consultation and feedback from PPI contributors.

Table 4.5: Questions to assess dental anxiety
I feel anxious about having any dental treatment with my normal dentist
I feel anxious about having dental treatment for my missing teeth

Hypodontia has been shown to impact on oral health-related quality of life (Meaney et al., 2012, Akram et al., 2011, Anweigi et al., 2013a, Hashem et al., 2013, Wong et al., 2006b, Locker et al., 2010, Laing et al., 2010), so a measure for assessing pre-treatment concerns was included. A hypodontia-specific tool for measuring impact of hypodontia and its treatment (Akram et al., 2011) was reviewed and although it was not possible to include the full questionnaire due to its length, one question from each domain was selected (Table 4.6). PPI contributors directed the selection of the question. In accordance with the original questionnaire, a 3-point scale (A lot / A bit / Not at all) was used.

Table 4.6: Questions to assess the quality of life impact of hypodontia
Please mark how much your teeth affect you day to day based on: Confidence to smile Ability to chew hard food Confidence to eat in front of other people Ability to speak – for example without a lisp Confidence to talk in front of other people Other people’s reaction to your missing teeth Ability to clean your teeth Worry about your teeth in the future

Familiarity and knowledge of care and service and previous treatment experience have been shown to be important in decision-making (Coulter et al., 2008, Bowling and Ebrahim, 2001). Participants were asked to report their stage of treatment (Not yet started / In treatment / Completed treatment) and previous dental treatment for their hypodontia. A list of common treatments was given, using descriptions recommended by PPI contributors (Appendix Table 13). In addition, parents were asked if they had hypodontia and any treatment received for this.

4.4.2 Attribute development

Synthesis of the data from Chapter 3 created an exhaustive list of attributes. Attributes related to 11 concepts: five concepts related to treatment process (time; service configuration; cost; impact of treatment; risk during treatment) with a total of 16 attributes and six concepts related to treatment outcome (psychosocial; appearance; function; dental health; harms; long-term) with a total of 26 attributes. The attributes, descriptor and source for each attribute is given in Appendix Table 14. The systematic review identified mainly clinical indicators. No attributes relating to possible psychosocial outcomes (quality of life / wellbeing) or risks of treatment were identified from the clinical consultations, while the interviews did not identify any attributes relating to risks during treatment.

Attribute reduction resulted in a long-list of relevant attributes (Table 4.7). This included 16 attributes relating to treatment process and 11 relating to treatment outcome. The decision log summarises the reason for including and excluding attributes and who drove the decision (Appendix Table 15).

Table 4.7: Summary of attributes following attribute reduction

Service delivery attributes	
Treatment duration	Overall treatment time; Length of appointment
Appointment schedule	Timing of appointment; Frequency of appointment
Time	Waiting time to start treatment
Service configuration	Travel time
Cost	Indirect costs of attending appointment
Impact of treatment	Discomfort during treatment; Effect on eating; Effect on speaking; Effect on hobbies; Effect on social interactions
Risk during treatment	Type of problem during treatment; Likelihood of problems; Timescale of problems; Effect of problems
Treatment outcome	
Psychosocial	Self-consciousness; Confidence; Other people's reaction; Feel of teeth
Appearance	Normalisation compared to peers; Gaps and straightness; Colour and shape
Function	Ability to eat; Bite
Long-term	Future treatment need; Future treatment cost

The attribute rating/ranking survey was completed between 23rd October – 19th November 2017 by 18 participants. The response rate was lower than hoped for. A number of members in the Facebook group offered to complete the survey but they were from the USA and Canada where the healthcare system is substantially different to the UK and it was judged this may skew the ratings of attributes. No surveys were abandoned incomplete suggesting the survey was acceptable to respondents. The results from this small sample were insufficient to determine final attribute selection; however, they were adequate to guide further stakeholder consultation and it was felt that the ethical approval required for recruiting additional NHS patients for the survey would cause a significant delay to the research.

Parent respondents included two fathers, eight mothers and a guardian. All adolescents and the majority of parents completed the survey alone, with only 3 parents reporting completion with their child. A spread of ages was reported by the adolescents and parents of adolescents with hypodontia (Table 4.8).

Table 4.8: Characteristics of respondents in the attribute rating survey

		Self-reported by adolescent	Reported by parent on behalf of child
Age (years) of the person with hypodontia	<13	0	3
	13	0	1
	14	2	1
	15	2	3
	16	3	3
Gender of the person with hypodontia	Male	2	4
	Female	5	7
Severity of hypodontia	Mild	2	1
	Moderate	2	1
	Severe	3	9

For the adolescent affected by hypodontia, the representation of male: female reflects hypodontia sex tendency. The majority of participants reported that they or their child had severe hypodontia. This does not match the most common presentation of hypodontia but may be a reflection of the use of a support group for sampling, if adolescents and families affected by more severe hypodontia are more likely to seek online support. Most participants reported some previous treatment experience, most commonly examinations with their own dentist or a hospital team, orthodontic treatment or tooth replacement with a denture. All participants were from England.

The survey scores are summarised based on the number of times each attribute was rated as important, the mean rating score and the number of times the attribute was ranked in the most important (Appendix Figure 3). For attributes relating to treatment process, the greatest differences in both rating and rankings between parents and adolescents were noted for travel time, discomfort and effect on speech. Fewer differences were observed in the rating and rankings between parents and adolescents for attributes relating to treatment outcome. Overall parents' mean importance ratings were higher than adolescents and generally, scores were higher for outcomes relating to treatment outcome than treatment process.

The subsequent stakeholder consultation process was used to select the most relevant attribute from each concept to provide coverage of the important components of hypodontia care. Stakeholders were presented with aggregate scores from the survey and the proposed categorisation of attributes to explore the validity of attribute categories. The stakeholders confirmed the preferred attribute from each category. Throughout the attribute selection process the language used to describe the attributes was assessed and revised. PPI contributors were instrumental in determining the most appropriate approach for describing attributes, including examples to aid understanding. The decision log for the selection of attributes for piloting is summarised in Appendix Table 16.

The evidence base provided little data to determine attribute-levels and much of the information was based on clinical knowledge and stakeholder advice. Explanation of the levels chosen is given below:

- **Discomfort during treatment:** Levels reflected the information provided in patient information resources and clinical consultations and experience reported on social media. Examples to aid understanding were suggested by PPI contributors.
- **Waiting time:** Levels were based on current service delivery in the NHS. A 3-month waiting time represented the usual wait for routine treatments in hospitals where there are no waiting list issues. A one-year waiting time reflected treatments with increased waiting time due to budget and service issues, while a three-year wait suggested treatment where there may be reasons for prolonged delay, such as a need for dental development or significant service delivery issues.
- **Treatment schedule:** Evidence regarding treatment time was largely based on audit results and clinical experience. A short treatment with few appointments

reflected minimally invasive treatment, such as restorative camouflage or denture construction without orthodontic treatment. Longer treatment times and more appointments represented more complex treatments.

- **Problems:** Framing risk is recognised as a complex area that can affect preference estimates (Veldwijk et al., 2016, Howard and Salkeld, 2009). Different approaches to framing risk, such as presence or absence of risk and the use of numbers or words to describe risk were discussed. Adolescents and parents were keen to avoid numbers and reported that wording such as ‘chance’ or ‘possibility’ were more useful.
- **Appearance:** Appearance was the most challenging attribute to frame and label due to variation in the interpretation of descriptions. To reduce complexity, two levels were chosen; the first level represented an appearance where treatment had little effect (small gaps) or resulted in a suboptimal appearance (colour disharmony) and the second level represented an optimal outcome.
- **Function:** Few outcomes used in the research provided useful evidence for determining the levels and framing for function in terms of bite. After discussion with stakeholders, two levels were selected and described as a relative change (same / improvement).
- **Future cost:** Levels were difficult to determine due to an absence of UK-based evidence about long-term costs of hypodontia care. With input from clinicians, NHS prices and an estimate of expected treatment burden were used to calculate approximate long-term costs (Appendix Table 17), which were averaged to give an estimate of cost per year.

The final attribute descriptors and levels for piloting are given in Table 4.9.

Table 4.9: Description of attributes and levels for piloting

Discomfort during treatment Dental treatment for your missing teeth might sometimes be sore or cause discomfort when you are eating and talking.	0	There will be little or no discomfort during treatment
	1	Discomfort during treatment will be moderate and may require painkillers
	2	Discomfort during treatment will be severe and may stop you seeing friends
Appointment schedule This is how long dental treatment will take overall and how often you will have to go to appointments.	0	You will have to see the dentist every 2 weeks for 2 months
	1	You will have to see the dentist every 2 months for 3 years
	2	You will have to see the dentist every 2 months for 5 years
Waiting time This is how long you have to wait to start treatment due to waiting lists or because your teeth need to grow	0	Waiting time to start treatment is 3 months
	1	Waiting time to start treatment is 1 year
	2	Waiting time to start treatment is 3 years
Problems during treatment During your dental treatment there is a chance you will have problems. This is the type of problems that may be caused by dental treatment.	0	There is a chance dental treatment will cause minor problems that mean you need to see your dentist e.g. to have an X-ray of your tooth
	1	There is a chance dental treatment will cause moderate problems that make your treatment longer e.g. an infection needing a filling
	2	There is a chance dental treatment will cause severe problems e.g. damage to a front tooth that means the tooth needs removing
Function How your teeth feel and bite after treatment	0	After your dental treatment, your teeth and bite feel much better
	1	After your dental treatment, your teeth and bite feel the same
Appearance How your teeth look after treatment based on the colour and shape of the teeth and if there are any gaps	0	After treatment, your teeth are straight without gaps and the colour match of the teeth is good
	1	After treatment you might have small gaps between your teeth or some teeth might look slightly grey or yellow
Future cost In the future you will need to see a dentist to have your dental treatment checked. The cost given is the average amount you have to spend each year	0	It will **not cost anything extra** to maintain the health and appearance of your teeth
	1	It will cost you **£50 a year extra** to maintain the health and appearance of your teeth
	2	It will cost you **£250 a year extra** to maintain the health and appearance of your teeth

4.4.3 DCE choice task development

4.4.3.1 Task construction

Task construction generally aims to create a choice context that accurately mimics the real-life situation; however, this was challenging due to variability in clinical presentation of treatment options by different dental teams and limitations in adolescent and parent understanding of treatment options.

Composition of choice sets

Profile format: A full profile is considered good practice although it is recognised that a partial profile can reduce cognitive burden for respondents (Bridges et al., 2011). For the pilot, a full profile was selected to test whether adolescents were able to consider all attributes or if simplifying heuristics were employed.

Number of alternatives: Due to the inherent complexity of DCE choice tasks, all means to reduce complexity without impacting on research design were preferred. For this reason, paired alternatives were chosen to minimise the complexity of the choice tasks. It was accepted paired alternatives would limit the total number of alternatives included in the study and potentially increase error variance (Greiner et al., 2014).

Opt-out: Dental treatment for hypodontia is elective and in some situations the options may not be judged to be sufficiently beneficial to warrant treatment so provision of an opt-out was desirable; however, evidence suggests that offering an opt-out may lead to high levels of non-response from respondents who find trading-off between options challenging (Veldwijk et al., 2014). It was not possible to provide a status quo treatment as there is no single agreed ‘gold standard’ treatment approach. Previous research stages demonstrated adolescents and parents have poor understanding of their own treatment, so the option to ‘Keep current treatment’ was excluded on the basis respondents were not able to compare this accurately (Campbell and Erdem, 2019). For the pilot a two-stage approach was taken, where respondents were forced to choose between the DCE paired alternatives in the Choice Task but afterwards a *‘No Treatment’* option was offered.

Labelling: Generic labels were chosen (Treatment A and Treatment B) because treatment options did not reflect exact treatment options, labels might encourage detrimental heuristics and estimation of attribute values and marginal rates of substitution rather than prediction of demand was planned (Blamey et al., 2000).

Experimental design

A full factorial design resulted in 2^2 x 3^5 = 972. This was too large to use in full and consequently a fractional factorial experimental design was chosen. The experimental design was selected to optimise design efficiency (d-efficiency), which maximises the precision of estimated parameters for a set number of choice tasks (Janssen et al., 2018). No priors were used in the pilot experimental design. Attribute levels were assumed to be categorical and uniformly distributed. Statistical modelling software Ngene v1.2.1 (ChoiceMetrics) was used to create and test the efficiency of different experimental designs (Vanniyasingam et al., 2016).

Evidence for the optimum number of choice tasks is scarce, but previous health applications have used between 8-28 tasks (Clark et al., 2014, de Bekker-Grob et al., 2012). Eight choice tasks were included for testing in the pilot survey to assess how respondents coped. This was expected to generate adequate data for preference weights estimates to inform the final experimental design. Blocking was used to increase the total number of Choice tasks for testing with 32 Choice tasks across 4 blocks. Approximate level balance (Appendix Table 18) and utility balance was checked. Examination of the attributes suggested no apparent correlation or dominance across choice sets.

Preference elicitation

The purpose of the research was explained in the opening section to provide motivation for completing the survey. An explanation of the DCE task was provided immediately before the choice tasks with a description of the tasks and instructions how to answer the questions. No questions were included to test comprehension of the attributes and levels. An example task was then provided for the respondent to complete.

The preference elicitation format was a choice between the options (Treatment A or Treatment B) followed by three qualifying questions about strength of preference, confidence in choice and the option of *'No Treatment'* (Figure 4.3). A 5-point rating of strength of preference was included after the simple choice between the alternatives to provide greater information about preference. Difficulty of choice was measured using a 5-point scale to assess how easily respondents could distinguish between the options in the choice set. The purpose of the question was to measure choice certainty, rather than perceived consistency between hypothetical and real choices (Greiner et al., 2014). These additional qualifying questions were informed by other DCE studies with adolescent respondents, which included measures to assess choice certainty (Forsander et al., 2018) and perception of the DCE tasks (Hofman et al., 2014). No questions were added to elicit reasons for the choice difficulty rating or selection of no treatment. This would help identify whether difficulty was due to poor understanding or genuine choice difficulty (Forsander et al., 2018) and whether no treatment reflected misunderstanding or a protest bid (Greiner et al., 2014); however, it was felt additional questions about rationale would add to the survey burden without sufficient benefit.

Figure 4.3: Qualifying questions for choice tasks

How much do you prefer Treatment (x)?

Not a lot A lot

☐ ☐ ☐ ☐ ☐

How confident are you in your answer?

Not confident Very confident

☐ ☐ ☐ ☐ ☐

You said you prefer Treatment (x). Do you prefer this or no treatment at all and keeping your teeth as they are?

☐ Treatment (x)

☐ No treatment

4.4.3.2 Instrument design

The pilot instrument contained four parts:

- Introduction: Purpose of the research (motivation for completion); Information about the survey; Instructions about eligibility and screening questions; Email address for thank you voucher
- Respondent demographics: Age in years, gender, ethnicity, geographical location, number and location of missing teeth, rating of general and dental health; Estimate of level of education and household income
- DCE Choice tasks: Explanation of the DCE task with example; Description of attributes and levels; Practice question; 8 Choice tasks
- Decision-making questions: Knowledge; Beliefs about dental treatment; Beliefs about decision-making; Anxiety; Impact of hypodontia on quality of life; Experience of dental care

The introduction was included for testing in preparation for online data collection where there would be no researcher present. The introduction included instructions about who could complete the survey and screening questions. Consent was requested from the participant, and for those under 16 years old, respondents were asked to confirm their parent/guardian was happy for them to complete the survey.

The preliminary instrument was longer than intended for the final instrument to enable different content to be tested. Adolescent-parent contributors indicated that the maximum time they would give to complete an online survey was around 20 minutes, but this would increase to 30 minutes if there was an incentive.

4.4.4 Pilot study

The pilot study was conducted in January-February 2018. The pilot included 20 participants from across two hospital sites (Table 4.10). One adolescent-parent dyad and two adolescent-sibling dyads completed the pilot survey to provide joint preferences. The remaining adolescents and parents completed the survey separately; in these cases, the adolescent completed the survey first and although their parent was present, they were instructed to select their own preferences without consulting their parent. The parent then completed another version of the survey with different choice tasks. The parent group included six mothers and one father.

Table 4.10: Characteristics of pilot study respondents

Participant	Adolescent	12	**Site**	Leeds	14
	Parent	8		Bradford	6
Adolescent with missing teeth – self-reported or reported by parent:					
Age	12 years	2	**Gender**	Male	6
	13 years	4		Female	14
	14 years	2			
	15 years	7			
	16 years	4			
Severity of hypodontia	Mild	10	**Stage of treatment**	Pre-treatment	1
	Moderate	7		Mid-treatment	8
	Severe	3		End of treatment	1

4.4.4.1 Contextual questions

Two adolescents opted not to answer whether they received free school meals. Verbalised feedback suggested this was because they did not receive free school meals and were not aware that it was possible. For the household income question, five out of the eight parents opted not to answer and verbal feedback revealed parents regarded it as irrelevant and intrusive.

The questions to measure knowledge demonstrated uncertainty in the answers and respondent feedback suggested this was due to ambiguity in the questions. The majority of respondents selected 'Unsure' for all questions (Appendix Figure 4). For the questions regarding beliefs about health, 16 respondents rated their general health as excellent, three as good and one as fair. For dental health, 13 selected excellent and 7 selected good. Verbalised feedback suggested adolescents were not sure how to answer this question and that despite being in objectively excellent health, most would have given a lower score if they were not able to ask their parent for clarification:

> *"How do I rate my health… I'm OK I guess. Fair? Good? Not sure about excellent. Mum what would you say?"* (Adolescent, P5)

Beliefs about dental treatment responses are provided in Table 4.11. The verbalised feedback suggested some adolescent respondents did not understand the statement and their answer did not appear to accurately reflect their opinion when probed further. For example, the results appear to suggest the majority of people do not think dental treatment is the only option for missing teeth but when questioned further, respondents were not able to explain what other options they were aware of, as typified below:

> *"Errr dunno really, there must be something else… I'm not sure"* (Adolescent, P8)

Table 4.11: Responses for beliefs about dental treatment (%)

	Yes	No
Dental treatment is the only option for missing teeth	40	60
Having dental treatment now for missing teeth will reduce the need for more dental treatment in the future	45	55
I worry about the risks of dental treatment for missing teeth	25	75
Dental treatment for missing teeth can result in new dental problems	75	25

4.4.4.2 Validity of attributes and levels

Understanding of attribute relevance and validity were assessed using qualitative feedback supported by analysis of preference data. Think aloud feedback confirmed that there were too many attributes in the choice task to allow respondents to consider the full profile, as demonstrated in these comments:

“It’s really hard, there is too much to think about, I keep getting confused”
(Adolescent P2)

“I think I keep forgetting to look at one because there are too many”
(Adolescent P5)

Describing the attribute-levels adequately without information overload presented challenges. It was apparent that without comprehensive description of the levels for ‘Discomfort during treatment’ and ‘Problems during treatment’ respondents were unclear about the meaning of mild, moderate and severe. Describing each attribute and its levels individually on separate screens improved understanding by forcing respondents to read through the descriptions, but respondents reported this made the survey too long and they lost interest.

“Oh gosh, that’s so much to read. Can I skip it?” (Parent, P4)

“This is going on forever… I’m getting bored” (Adolescent, P9)

Respondents did not identify any attributes that they felt were missing and generally all attributes were considered to be important and relevant. When asked which attributes they judged to be more or less important, differences were found between adolescents and parents (Table 4.12). Parents were less consistent in their perception of the least important attributes. Two mothers of adolescents who were near the end of treatment or post-treatment commented that they felt their child’s focus had changed throughout the course of dental treatment; during treatment there was more importance placed on process, particularly discomfort, but now the priority had shifted to outcome.

“She didn’t used to be bothered how good it looked as long as it was normal but now I think she really cares. Now she’d keep her braces on forever if it made her teeth perfect” (Mother, P8)

“At the start she just wanted it all done quickly and with no pain but now she is used to it she seems to think more about how it will be at the end and that’s what is important” (Mother, P11)

Table 4.12: Respondent's perceived importance of attributes from pilot study

	Attributes judged to be more important	Attributes judged to be less important
Adolescent	Appearance Bite after treatment Problems during treatment	Cost of treatment Waiting time Appointment schedule
Parent	Appearance Problems during treatment	Variable between individuals Bite after treatment (two parents)

Estimates of preference weights for the whole sample (160 observations) and by respondent group are given in Appendix Table 19 and summarised in Figure 4.4. Attribute signs met prior expectations (loss of utility for levels expected to be less desirable) for all attribute-levels except waiting time. The greatest utility loss was observed for appearance and severe problems, both of which were statistically significant (p<0.05). Verbalised feedback supported that appearance was very important, particularly for adolescents, and this was often the first and sometimes the only attribute that was considered. Both parents and adolescents reported a strong desire to avoid problems, although parents expressed greater concern and a better understanding of the impact of problems.

> *"I always look at that one (appearance) first. That's the most important by miles"* (Adolescent, P2)

> *"I could tell he always thought about how it looks first, but for me I think the most important thing has to be any severe problems"* (Mother, P6)

Reduction in utility for *'No change to the bite'* was significant for adolescents but not for parents. This reflected verbalised feedback; function was reported to be important by most adolescents but parents found it less important. One mother specifically highlighted "bite" had only become a consideration during treatment when the dentist discussed it frequently but she did not feel it had really been a concern pre-treatment.

> *"I'm surprised about bite. You didn't used to ever say that was a problem, it was always about the braces being sore. Maybe it's because your dentist here talks about the bite a lot"* (Mother, P10)

The final attribute, '*future cost'* was the most problematic attribute. Despite modifications to the wording in the description, respondents frequently mistook it to be the cost of current, rather than future, hypodontia treatment. It was also apparent that many respondents were not aware that in reality, many types of hypodontia treatment carry costs in the future. Adolescents showed little interest in cost and did not consider it in their choices. This is illustrated by respondent comments:

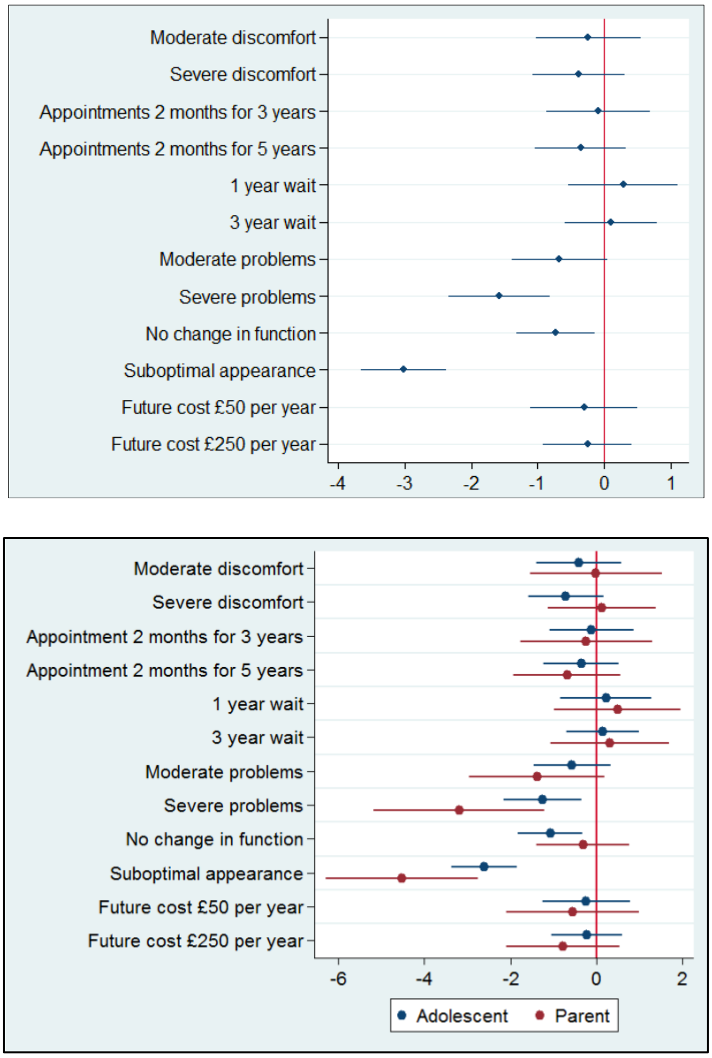
> *"I'm not looking at that (cost). It doesn't matter cos dad will be paying for it. And it's free anyway, we don't have to pay"* (Adolescent, P6)
>
> *"hmmm I'm not sure. What does future cost mean? I don't really think about that much"* (Adolescent, P4)

As a result, the future cost attribute was judged to be irrelevant to adolescent decision-making and it was removed during finalisation of the DCE choice tasks.

Figure 4.4: Preference weights estimated from preference data from pilot study, given as mean estimate and 95% CI

a) Whole sample (observations n=160)

b) **Segmented by respondent group (observations adolescent n=96, parent n=64)**



Preference weights for attribute-levels are given relative to the omitted level for that attribute, which has a value of zero (red line). Reference levels are:

- Discomfort: none/mild
- Appointment schedule: 2 weeks for 2 months
- Waiting time: 3 months
- Problems: none/mild
- Bite: improved
- Appearance: improved
- Future cost per year: £0

4.4.4.3 Suitability of choice tasks

Both adolescents and parents had difficulties in understanding the purpose and method of the choice tasks. Where the balance of utility in the alternatives was more similar, some respondents were resistant to make a choice. The ability to perform the choice tasks appeared to be related to respondent's ability to understand the concept of trading off attributes, rather than age. Adolescents in particular asked for ways to help process the task.

> *"It's hard. Can I just pick the bits I like out of each one instead?"* (Adolescent, P8)

> *"It would be much better if you could just highlight the good bit in each one, then you could count them up and decide like that"* (Adolescent, P15)

Responses to the questions about strength of preference and choice difficulty did not reflect verbalised feedback. Respondents generally did not perceive a difference between strength of preference and certainty so tended to answer both the same, and many adolescents could not articulate what they thought the questions meant. Examination of responses to the strength of preference and certainty questions for each choice task demonstrated a strong correlation (Spearman's correlation 0.69, p<0.001), suggesting it was not necessary to include both.

Revisions were made during the pilot until the response elicited was consistent with respondent feedback. Strength of preference was changed to a two-part decision, where choice was first selected (Treatment A or Treatment B), then the respondent indicated how much they preferred this choice (A bit or A lot). Choice difficulty was reworded to "How easy was it to pick?" with a 3-point response scale (Very easy / Quite Easy / Difficult). These were judged to be easier to understand but examination again demonstrated a strong correlation between strength of preference and ease of choice (Spearman's correlation 0.67, p<0.001).

'No Treatment' was selected in 11 of the 160 observations (7%) and in most cases, no treatment was only selected once or twice by an individual suggesting it was being used correctly. One adolescent (male, 15 years, severe hypodontia and mid-treatment) picked no treatment in four of the eight tasks. Verbal feedback found he was avoidant of problems and discomfort and not motivated by the appearance of his teeth, which might alter his perception of the risk-benefit of treatment.

4.4.4.4 Acceptability of tool & method

Feedback about the appearance, layout and wording of the survey was used to revise and retest the instrument design until respondent feedback was satisfactory. The content of the survey, mechanism for gaining consent and demographic questions were acceptable to the adolescents and parents and no concerns were raised about potentially sensitive or inappropriate topics.

The perceived level of burden of the overall survey varied between respondents and appeared to be largely related to how complex the respondent found the DCE choice tasks. To reduce the overall burden different formats were tested for providing information about the survey, the choice tasks and the attributes and levels. Following respondent feedback, the background colour changed for each choice task to confirm that the screen had moved onto a new task and progress was being made. Based on respondent feedback, colour coding was used throughout the survey to aid navigation through the sections. Time taken to compete the survey was measured for each iteration until the final respondents were able to complete it in under 30 minutes, and most in a much shorter time.

The pilot study tested the scope to use the instrument to observe joint preference elicitation between dyads to gain insight into decision-making processes and negotiation. Findings suggested during joint completion respondents discussed the attributes and options and debated the trade-offs between attributes. In most tasks the dyad quickly reached agreement, but when the choices were judged to be more balanced or one respondent had a strong preference for a certain attribute, there was considerable debate and negotiation to reach a decision. The method provided information about how agreement was reached and the rationale behind decisions, suggesting the proposed method was suitable for examining the negotiation process between adolescents and parents.

4.5 Finalisation of preference elicitation instrument & methods

The findings from the pilot were used to finalise the preference elicitation instrument and the main data collection methods. A summary of the changes made between the pilot version and final version is given in Appendix Table 20.

The final instrument contained five parts (Appendix Table 21). The full survey is included in the electronic supplementary file. To improve usability a mapping screen and colour coding were added to aid navigation of the survey. The questions about household income and free school meals were omitted as the cost attribute was removed, so there was no intention to calculate willingness-to-pay. Questions to measure knowledge of hypodontia and dental treatment and beliefs about general health, dental health and dental treatment were removed, as the pilot suggested the questions were not valid. Questions about dental treatment experience were simplified and re-worded to improve the accuracy of self-reporting treatments.

The number of attributes was reduced from seven to six to reduce the cognitive burden of tasks. Think aloud feedback and additional probing highlighted the inclusion of both frequency of appointments and total treatment time was too complex and simplifying techniques were used by adolescents, who tended to only consider treatment time. As a result, appointment schedule was revised to focus on total treatment time only (Table 4.13). For the remaining attributes, the description of the attribute and the number and dimensions of attribute-levels were not changed.

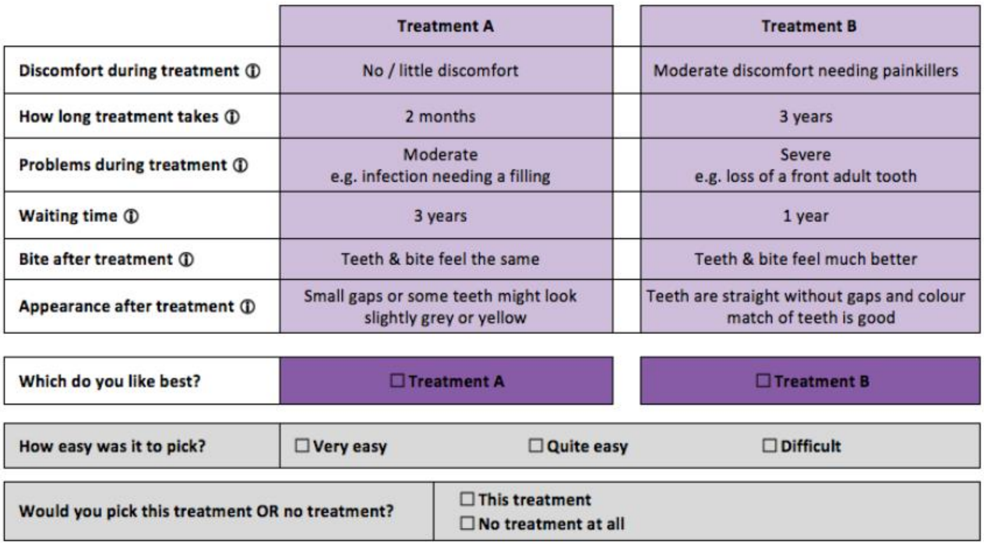
Table 4.13: Description of revised attribute for final preference elicitation instrument

How long treatment takes in total This is how long dental treatment for your missing teeth will take overall.	0	Dental treatment for your missing teeth will take 2 months to be completed
	1	Dental treatment for your missing teeth will take 3 years to be completed
	2	Dental treatment for your missing teeth will take 5 years to be completed

To address difficulties with explaining the attributes and levels, an interactive animation was used (Appendix Figure 5). A shortened version of the attribute-level was given in the profile and respondents could access the full description by pressing an information button to open a pop-up box. During the pilot, different explanations of the choice tasks and the concept of trading-off were tested until a successful approach was identified (Appendix Figure 6).

Paired alternatives and no labelling were maintained. The option of *'No Treatment'* was retained as a two-stage decision, with the forced choice first then the option for no treatment after. Additional information was added to the introduction to explain that 'No Treatment' meant accepting the teeth as they are and having no dental treatment at all. An example Choice Task is given in Figure 4.5. The qualifying questions were reduced to a single question to establish perceived difficulty of choice using a 3-point categorical scale, which was understood more easily by adolescents.

Figure 4.5: Example of DCE choice task format



	Treatment A	Treatment B
Discomfort during treatment ⓘ	No / little discomfort	Moderate discomfort needing painkillers
How long treatment takes ⓘ	2 months	3 years
Problems during treatment ⓘ	Moderate e.g. infection needing a filling	Severe e.g. loss of a front adult tooth
Waiting time ⓘ	3 years	1 year
Bite after treatment ⓘ	Teeth & bite feel the same	Teeth & bite feel much better
Appearance after treatment ⓘ	Small gaps or some teeth might look slightly grey or yellow	Teeth are straight without gaps and colour match of teeth is good
Which do you like best?	☐ Treatment A	☐ Treatment B

How easy was it to pick? ☐ Very easy ☐ Quite easy ☐ Difficult

Would you pick this treatment OR no treatment? ☐ This treatment ☐ No treatment at all

Reducing the number of attributes from seven to six reduced the number of possible profiles in a full factorial design to 324 (3^4x2^2) but it was not feasible to use a full design, so a fractional factorial design was used. Eight choice tasks were presented but this included one repeat task to test reliability (see Section 5.1). Blocking was used to increase the total number of choice tasks to 28 across 4 blocks.

To improve the efficiency of the experimental design preference weight estimates from the pilot were used (Rose et al., 2008) (Table 4.14). For waiting time, very small negative values were used to reflect the expected direction of preferences while recognising the insignificant preference estimates found for waiting time in the pilot study.

Table 4.14: Preference weight estimates used to improve efficiency of experimental design for final DCE survey

	Discomfort	Appointment	Wait	Problems	Function	Appearance
Level 0	Dummy level (0)					
Level 1	-0.24	-0.09	-0.00001	-0.67	-0.37	-2.61
Level 2	-0.38	-0.36	-0.0001	-1.58		

A d-optimising efficiency, MNL main effect design plan was used with Ngene v1.2.1 (ChoiceMetrics). No logic rules were used, as there was no clinical suggestion of a relationship between attributes. The large preference weight for appearance in the pilot study resulted in a design with overlap in the appearance attribute between alternatives in 18 out of the 28 choice tasks, reflecting the dominance of appearance. Constraining appearance had the additional benefit of reducing the number of attributes for consideration in some tasks. The final design was checked for dominance and plausibility.

The preference elicitation survey offered a valuable opportunity to compare hypothetical treatment choice in the DCE choice tasks to actual treatment choice to estimate external validity (Janssen et al., 2017, Quaife et al., 2018). The accuracy of preference elicitation methods to identify true preference is important for future design of preference support tools. Participants were asked to self-report their planned treatment based on four common treatment options (Figure 4.6). The risk of inaccuracies in self-reporting treatment choice and unknown availability of services for individuals were recognised as potential limitations of this approach.

Figure 4.6: Options to self-report planned dental treatment for hypodontia

	Yes	No	Not sure
White fillings to change the shape or size of your teeth	☐	☐	☐
Braces to move the teeth	☐	☐	☐
‘Bridge’ type of false tooth (a false tooth glued to other teeth)	☐	☐	☐
Dental implant (a metal screw placed in the jaw to hold a false tooth)	☐	☐	☐

4.6 Discussion

4.6.1 Development and validity of attributes

Attribute development methods that accurately capture the attributes that are relevant and important to the target group is fundamental to preference elicitation research. The volume and diversity of attributes of hypodontia care identified by the attribute identification methods emphasised the complexity of hypodontia care, but also the divergence in perspective between professionals and patients. The systematic review (Section 3.3) provided a number of clinical indicators, largely related to the mechanics of service delivery and dental health outcomes. This contrasted with patient-centred methods, where service delivery was framed more in terms of practicality and impact on everyday life. For example, access attributes from the systematic review focussed on how services are structured to deliver MDT care, while the interview participants highlighted practical aspects were important to their experience, such as familiarity with location and ability to park. Dental health outcomes found in the review that were not replicated by any other method included: technical measures of appearance, such as the dental aesthetic parameters; mastication and occlusal function; proxy measures of success, such as periodontal indices; and alveolar bone volume. These measures, although potentially important, are difficult to translate into attributes and levels that are readily understood by adolescents and parents.

Qualitative methods were included to represent the perspective of the target respondent (Coast et al., 2012, Abiiro et al., 2014) and to provide a richer and more nuanced description of attributes. These methods identified the psychosocial impact of treatment, the perceived impact of treatment on family and attributes relating to bullying and teasing that were missed by clinician-led methods. This emphasises the potential discordance between clinician priorities and how adolescents and parents perceive hypodontia and treatment. The clinical consultations did not provide any attributes relating to risks of treatment and there was little discussion of risks of treatment in the interviews. This suggests patients may be unaware of the risks of treatment; however, it is highly unlikely risks were not discussed in interviews because they are not important, as the ‘severe problems’ attribute-level was found to be significant in the pilot DCE analysis.

Attributes relating to behaviour change, self-care and the impact of treatment on everyday life arose primarily from patient information resources. This raises possible concerns about the mechanisms used to communicate information to patients, with a reliance on patients being able to self-educate about their role in treatment and possible side-effects. Discomfort is subjective and clinical experience suggest the daily impact of treatment varies across individuals and treatment methods. This presented challenges for determining an appropriate approach to describing discomfort and defining attribute-levels was challenging. Examination of hypodontia practice (Chapter 3) identified these areas are often avoided in clinical discussions. PPI contributors were crucial for recommending examples to illustrate the levels.

Time-related attributes arose from a number of sources and were included in the pilot DCE to reflect treatment burden and adverse psychological effects of treatment process. Piloting suggested time attributes were easily understood and were considered important by both adolescents and parents, which supports previous research highlighting the importance of treatment burden and delays in treatment (Meaney et al., 2012, Akram et al., 2011). Attribute development methods and piloting suggested that discomfort, problems and appearance evoked a more emotional reaction than time and function attributes. It has been suggested that choice behaviour may be influenced by emotion due to interplay between cognitive ability and affective states (Arana et al., 2008). This study did not attempt to quantify the emotional impact of the attributes, but this is an area that potentially warrants further investigation to explore the implications for clinical decision-making.

Appearance and bite were challenging to define due to differences in individual's interpretation of the attributes and their own experience. Clinical indicators arising from the systematic literature review were not relevant to the target audience, but subjective or experiential descriptions were prone to bias in interpretation. For appearance, a plausible clinical description based on important aspects of dental appearance (straightness, spacing and colour harmony) was developed rather than simply terms such as good/excellent or same/improved. Use of images to aid description of the qualitative attribute was considered but previous DCE research found images can introduce bias (Hurtubia et al., 2015) and evidence about patient perception of clinical photographs is scarce. The variability in presentation of hypodontia meant it was likely to be difficult to find an image that was relevant to all respondents. The size of the preference estimate for the appearance attribute suggests a strong desire to avoid a compromised appearance and this was supported by verbal feedback.

For biting function, a description of the individual's perception of function was used, rather than function relating to a specific task, such as eating. Due to the subjective and variable nature of the attribute, relative levels were used (same/improved). It is possible that an alternative description of function, for example describing function as a restriction on biting ability, would result in a different preference estimate. Empirical testing of this would require comparison of estimates derived from two versions of the DCE Choice tasks with different attribute descriptions (Janssen et al., 2017), which was outside the scope of this research. The chosen description of function was selected based on stakeholder feedback and piloting indicated it was valid; however, clinicians may feel that understanding the relative importance of limited function might be more useful for counselling during decision-making.

The qualitative feedback was fundamental to the decision to remove the ‘Future cost’ attribute, where preference data was ambivalent. Verbal feedback indicated adolescents had little concept of the future or the concept of monetary values, and as a result they did not have a consistent reaction to the cost attribute. Adolescents often assumed their parents would pay for any future treatment, so they considered cost to be irrelevant. Some adolescents indicated that they struggled to think about the future beyond finishing school, or going to university, so this made it difficult to think ahead. Research with teenagers and young adults found strategic planning and the ability to plan ahead increases with age and depending on the complexity of the task, some adolescents struggled to control impulses to make well thought out decisions (Albert and Steinberg, 2011).

4.6.2 Instrument development and testing

The validity of the questions developed to establish the decision-making context of preference elicitation varied. Knowledge questions did not appear to accurately establish respondent knowledge of hypodontia and its treatment, with the majority of respondents answering ‘Don’t know’. This could be due to genuine deficiencies in knowledge, a lack of interest or motivation, ambiguity in the questions or answer options, reluctance to give an incorrect answer. Assessment of key knowledge about the nature and outcome of options is included as one construct in the decisional quality construct (Sepucha et al., 2013); however, it is recognised that knowledge is difficult to measure accurately, and more importantly, knowledge does not necessarily equate to good decision-making.

Few adolescents rated their own general and dental health as excellent and cognitive feedback suggested ambiguity in the question and answers. A study of Norwegian teenagers found self-ratings of health are associated with a broad spectrum of medical, social and personal factors (Breidablik et al., 2008), and it seemed adolescents were unsure how to interpret the question. Similarly, verbalised feedback for the questions to establish beliefs about dental care demonstrated confusion and the answers given did not accurately reflect how respondents felt. In comparison, the questions about decision-making, quality of life and dental anxiety worked well. The difference appeared to be due to the wording of the questions, familiarity and understanding of the concepts, and pre-existing awareness of their response to the question. Decision-making beliefs, hypodontia-related quality of life impact and dental anxiety were considered important for decision-making and required further examination in the subsequent stage.

Decisions regarding task construction and experimental design were guided by evidence and tested in the pilot but it was outside the scope of the pilot to empirically test different DCE design choices, such as the optimum number of attributes, levels, alternatives and choice tasks. The pilot indicated two alternatives in a choice task was sufficiently challenging, possibly due to the number of attributes to consider, so it is expected that more alternatives would have increased the complexity of task. Respondents' opinion regarding their preferred number of tasks varied; those interested in the task and those whose understanding of the tasks improved throughout completion reported eight tasks was acceptable, while those who struggled with the tasks reported eight was burdensome. This suggests optimum design features might depend on the respondent's aptitude for the task. This requires further investigation to inform future decision support tools.

The questions to quantify strength of preference and confidence in choice appeared correlated and a number of respondents demonstrated that they considered the questions to be similar. Strength of preference provides scope for adding weighting to preferences, while confidence in choice relates to expectations about choice behaviour. Two opposing hypotheses have been suggested for the effect of choice certainty on preferences. The first hypothesis assumes that some degree of task difficulty causes more reasoned response, so increased weighting is given to respondents who indicated tasks were difficult (Regier et al., 2014). The second hypothesis assumes task difficulty results in use of heuristics that violate assumptions of DCE, so estimates will be less precise (Lundhede et al., 2009, Dekker et al., 2016) and respondents who indicated tasks were difficult should be excluded from the analysis. For this research it was important to include a measure to understand how respondents perceived the choice tasks, as this information is useful for designing clinical preference elicitation tools.

4.6.3 Preferences and decision-making behaviour

Cognitive interviewing through think aloud methods with some verbal probing in the pilot provided preliminary ideas about how adolescents, parents and dyads make decisions when faced with alternatives that require trade-offs between attributes. The results suggested respondents fall into three groups based on their approach to the choice task: those who weighed up individual attributes in each alternative to reach a decision; those who tended towards selection based mostly on one or two attributes; those who found the task difficult and picked without a clear reasoning process. There was no obvious relationship between age, stage of treatment and approach to task. The mode of decision-making appeared to influence the perceived cognitive burden and enjoyment of the task. Some respondents asked for a method to make task easier and there was evidence of the use of simplifying heuristics for those finding certain tasks difficult; however, this did not necessarily result in selection of a choice that was not concordant with preference. This is illustrated in a comment from a respondent:

> *This one's hard, they're too similar, I don't know which to pick. I can't decide. OK I'm just going to look at how good it looks 'cos that's what's important"*
>
> (Adolescent, P3)

Older adolescent participants, who tended to be further into treatment, and the parent respondents generally expressed greater interest in their choices and reported that completing multiple tasks helped them understand their preferences.

With reference to observation of dyads completing the survey, the method was not necessarily a tool for exposing thought processes but more a tool to allow observation of DCE task completion together to understand interaction. Parental presence was beneficial for establishing areas where adolescents were struggling with the task as parents were often able to explain misunderstandings. Completion of the DCE choice tasks stimulated conversation between parent and child and provided a common language for discussion. Negotiation between dyads tended to lead to more consideration of that attribute in future options and there was a convergence towards certain attributes. Preference elicitation between dyads appeared to promote greater consideration of options.

Adolescents and parents appeared unaware of their knowledge gaps, but presenting matched options helped a systematic approach to analysing the information. The use of matched options for direct comparison of alternatives and to highlight important information is the basis for option grids (Elwyn et al., 2013b). These short summary tables are presented on a single side of paper using concise, accessible and clear language to enable rapid comparisons of options. Grids are constructed using questions that patients frequently ask purposefully limited to the most important information, usually a maximum of 6-8 questions, for two to three treatment options. Option grids are designed for use in face-to-face clinical encounters to initiate conversation (Elwyn et al., 2013b). An evaluation of option grids in breast cancer, tonsillectomy and head and neck cancer found grids standardise the provision of information, facilitate patients' understanding of treatment options and consultations, assist patients to visualise the difference between treatment options and act as an aide-mémoire for experienced staff and a training tool for junior staff (King et al., 2012).

The option of *'No Treatment'* was selected relatively infrequently, except by one respondent who showed low motivation towards treatment. Opting out may be a genuine choice or may arise as a result of a preference to retain the status quo regardless of the attributes of the alternatives, a perceived difference between the opt-out and other alternatives, avoidance of difficult trade-offs, poor understanding of the choice context, preference for the consequence of inaction rather than the wrong action, or as a strategic or protest response (Campbell and Erdem, 2019). Prior to piloting there was concern the opt-out might be used to avoid difficult decisions but this was not found, possibly due to the two-stage decision process (Veldwijk et al., 2014). The low number of people opting for no treatment suggests a strong desire to have treatment regardless of benefit, or an artificial tendency towards treatment caused by the hypothetical nature of the choice option. Further exploration of the effect of an opt-out with adolescents is warranted.

Preliminary findings about preferences suggest when the alternatives require a compromise between process and outcome, respondents tend to accept undesirable process attributes for a gain in outcome. Trade-off between function and appearance was more challenging for some respondents, but in most cases appearance was dominant. The main concern for process attributes appeared to be severe problems.

4.6.4 Strengths and limitations

4.6.4.1 Attribute development methods

Attribute identification methods aimed to capture attributes that are important to adolescents and parents with relevance to current treatment options and service delivery. Policy documents were not searched (Abiiro et al., 2014) as the purpose of the research was to examine adolescent and parent preferences and decision-making, rather than inform policy changes. Different attribute identification methods successfully contributed different attributes and it is unlikely other approaches would have identified anything further. The final framework of attributes was judged to adequately describe the latent construct and a broad range of descriptors were maintained to capture the meaning of the concepts (Coast et al., 2012).

Choosing an appropriate method for selecting attributes from the vast array identified was challenging, but attempts were made to ensure transparency and robustness in the approach. The attribute selection process was improved by inclusion of stakeholder consultation but focus groups and more structured methods may have been beneficial (dosReis et al., 2016). Despite the lack of focus groups, there was good agreement between the contributors about what was important and how they understood attributes; however, this could be reflection of the participants being treated within the same geographical area and healthcare system. The contributors were all female and undergoing treatment, so this may have skewed their experience and opinion. The consultation process highlighted that strong opinions, particularly of dental professionals, had scope to strongly influence attribute selection. This is important to acknowledge, as it means attribute selection may not have represented all views and there may be excluded attributes that are important to other stakeholders. The consultations involved a limited number of stakeholders and there is likely to have been a self-selection bias in those who agreed to participate.

The attribute rating/ranking survey was limited by a small number of respondents and associated limitations to representativeness. While social media has potential to be a useful recruitment tool (Social Media Research Group, 2016), it is difficult to accurately estimate reach and likely participation. The Facebook group used for recruitment had 96 active and engaged members but subsequent posts by users confirmed many were not UK-based, which was not evident from profiles. Alternative recruitment was considered but an open survey was unlikely to reach the target group and risked completion by ineligible respondents, while recruitment of NHS patients was not feasible within the timescale due to the need for additional NHS ethical approval.

4.6.4.2 Approach to instrument development and testing

Development of the choice tasks for the preference elicitation instrument followed best evidence where possible, but the absence of information for DCE with adolescents meant some decisions relied on testing in the pilot. For example, there is little guidance about optimal approaches for introducing attributes and tasks, for choosing questions to qualify strength of preference and certainty. Significant time was allocated to identification and selection of attributes but on reflection, it may have been beneficial to include more time for testing aspects of DCE design, particularly the validity of different methods for introducing the choice tasks and attribute-levels, measures for assessing understanding of the attributes and different approaches for offering an opt-out.

For the background questions about the decision-making context, the approach taken (identifying possible areas of interest, developing measures based on current evidence, and testing these in the pilot) was satisfactory. Ideally, pre-existing validated measures would have been used but there were few suitable tools available. Decision-making is a complex process and a vast number of questions to understand the decision-making context could have been included, such as experience of service to date, attitude towards different dental professionals (Caldow et al., 2007) and emotional state (Arana et al., 2008). The questions selected provide adequate information about the decision-making context without adding significant burden.

Cognitive interviewing provided valuable insight into the potential discrepancy between findings from qualitative and quantitative methods and the processes used by respondents to reach a decision. This is supported by other studies using a combined approach to DCE data collection (Ryan et al., 2009, Cheraghi-Sohi et al., 2007, Whitty et al., 2014, McGrady et al., 2018). Verbalisation of thought processes has been shown to help understanding and reflection of adolescents where a simple “How” or “Why” might not (Koskey, 2016), and this was reflected in the respondents who showed better understanding of their own preferences after completing the tasks. The method also suggested that for some respondents, some of the assumptions underpinning DCE methods were being violated (Ali and Ronaldson, 2012), for example, the use of cognitive shortcuts or refusal to trade-off by some respondents, and inadequate understanding of task or attribute levels. Attempts were made to rectify these issues in the modifications for the final survey, to improve the quality of future preference data. Overall, the pilot method successfully provided information that supported modification of the preference elicitation instrument, verified the proposed data collection method, and provided preliminary findings about decision-making and preferences in hypodontia care.

4.6.5 Implications for clinical practice

Hypodontia is a complex condition and adolescent and parent perception of hypodontia care may differ from dental professionals' perception

Attribute identification methods emphasised that hypodontia care is complex with numerous diverse attributes, and decision-making will require difficult trade-offs between these attributes. The attribute development process suggested adolescent and parent perception and understanding of attributes of hypodontia care is different to that of professionals. Differences in the meaning of dental terms, familiarity with procedures, individual's perception of risk and nuance in language can present challenges for effective communication. Differences in perception and understanding of aspects of hypodontia care delivery and outcomes are important to identify for clinical decision-making. The preliminary results also suggest there is a difference in the value placed on different parts of care by individuals, so it is important that clinicians establish what is important to individuals to allow appropriate information provision and delivery of preference-based care.

Preference elicitation methods may help adolescents and parents think about treatment

Preference elicitation using a method that required trade-offs between attributes appeared to help some people think about treatment by encouraging systematic consideration of attributes, identifying 'hidden' attributes of treatment, supporting direct comparison of options, promoting reflection on values and providing a common language for discussing important attributes. A similar approach might be beneficial in clinical practice and further work to explore this is indicated.

It is also important to recognise that some participants found the DCE Choice tasks difficult and for these people there was evidence of increasing indecision throughout the tasks. In these cases, this type of trade-off task might not be beneficial to support clinical decision-making and other approaches may be required. This was examined further in the next stage of the research.

4.6.6 Implications for future research

Research about decision-making:

- The belief that knowledge improves decision-making is a key component in informed decision-making so there is value in examination of the relationship between adolescent knowledge and decision-making ability in dentistry. Future research to develop and validate a measure of knowledge for adolescents which can be used clinically, or used in research to examine the relationship between knowledge and decision-making, may be beneficial.
- Development of a standardised and validated tool to measure beliefs about dentistry, similar to those developed for surgery (Francis et al., 2009) and medicine (Horne et al., 1999), would be valuable. This would allow the relationship between beliefs and decision-making behaviour to be examined.
- Use of DCE choice tasks to further examine the decision-making process in hypodontia and more widely across dentistry is warranted; this supports the next stage of the research.
- Research to examine the best approaches and tools for eliciting preferences from different types of decision-makers is necessary, for example, those who find trade-off tasks useful compared to those who find them too challenging. This would inform decision support tools to engage decision-makers with different needs.

Research to improve the design and application of DCE for use with adolescents:

- Selection of valid attributes is fundamental to DCE, so future research is warranted to examine the impact of different approaches to attribute selection, attribute framing, particularly presentation of complex information such as risk percentages and rates, and subjective information such as experiential outcomes and the presentation of attributes.
- DCE has been used predominantly with adults so there are areas of DCE survey design that require further evaluation with adolescents. This includes the effect of different numbers of attributes, levels and choice sets, the effect of an opt-out, and the effect of the preference elicitation format.
- Think aloud feedback and verbal probing provided an opportunity to examine the rationalisation process and consistency of choice behaviour with the utility theory (Ali and Ronaldson, 2012). For adolescents in particular, this approach has real value alongside preference modelling to provide richer data with which to examine DCE methodology.

Chapter Five:

Preference Elicitation Survey

Chapter Five:

Preference Elicitation Survey

This chapter describes use of the preference elicitation instrument in a cross-sectional survey to investigate adolescent and parent preferences and decision-making in hypodontia care. The two data collection methods are described, which included an online survey and a smaller qualitative study where the survey was completed under observation. The approach for analysing preference data is detailed. Results are reported in terms of response and sample characteristics, observed decision-making behaviour and preferences for hypodontia care. The preference elicitation instrument and data collection methods were evaluated in terms of validity for preference elicitation with adolescents and dyads, ability to examine decision-making, and perceived usefulness as a decision-making tool. The chapter concludes with the key implications for clinical practice and future research.

5.1 Introduction

The final investigative stage of the research examined adolescent and parent preferences and decision-making behaviour in hypodontia using the preference elicitation instrument developed in Chapter 4. The purpose of this stage was to understand approaches to decision-making and preferences, providing evidence that is useful to shared decision-making in clinical practice. It is evident from work in the previous chapters that there is a large volume of complex information in decision-making for hypodontia, so information that can help guide patient-centred, relevant information provision is beneficial. Understanding the aggregate preferences of adolescents and parents and identifying factors that contribute to differences in preferences is clinically relevant. Establishing the role of trade-off tasks and understanding how adolescent-parent dyads interact will inform future decision support interventions. Finally, determining whether DCE is valid and reliable in this group of respondents is important for future preference elicitation research.

The preference elicitation instrument was applied using two approaches. The first involved face-to-face survey completion by adolescents and parents individually and then together under observation to allow examination of decision-making processes. The second was an online survey across England and Wales to collect population level preferences from a larger sample. Observation of preference elicitation by adolescent-parent dyads was used to examine choice negotiation in addition to providing further preference data. The use of complementary qualitative methods alongside quantitative preference elicitation methods has been reported as a way to gain greater insight into the agreement between cognitive processes and proposed decision-making theory (Cheraghi-Sohi et al., 2007, Ryan et al., 2009, Kenny et al., 2003). Observations were used to examine experience using descriptive approach(Rodriguez and Smith, 2018), then this information was used to develop types of decision-makers.. Information about different types of decision-makers and the correlation to treatment choice is clinically relevant for designing and testing future decision support interventions.

Based on the pilot, it was expected that there would be different approaches to joint preference elicitation, as individuals showed different types of behaviour in the choice tasks. Behaviour appeared to be based on whether the respondent already had strong preferences for a certain attribute and how easy they found trading-off in the tasks (section 4.6.3). A study of parent-child decision-making in the management of chronic disease found various ways dyads collaborate, including discussing opinions and sharing information (Miller, 2009). Importantly, the participants considered collaboration was beneficial, regardless of who makes the final decision. Collaborative decision making was found to be influenced by factors associated with the parent, child and decision context, in terms of the seriousness of the decision and the current symptoms.

The pilot also suggested the trade-off tasks were useful for some people, to help them think about treatment and their preferences; however, this was not the case for everyone and some people did find the tasks challenging and appeared to look for ways to avoid trading-off (section 4.4.4.3). The three dyads who were observed in the pilot study used the tasks as a prompt to talk about what was important to them and what they were willing to compromise on. Based on the observed behaviour in the pilot, it was hypothesised that the majority of dyads would find the tasks a useful tool for talking about their treatment preferences, but there may be others who struggled with the trading off. A randomised controlled trial comparing three value clarifications methods, including DCE, to choose whether to undergo prostate specific antigen screening found the different methods resulted in different preferences; however, this did not affect the decision to undergo screening (Pignone et al., 2013). This suggests even if DCE tasks change the decision process, it may not ultimately affect choice. A literature search found two protocols for studies that plan to use DCE as a decision tool in the clinical setting (Ryan, 2017, Dowsey et al., 2016)I; however, no other completed studies were found.

Online DCE surveys are common (Clark et al., 2014, de Bekker-Grob et al., 2012, Soekhai et al., 2019), but there is some evidence that interview completion provides lower preference estimates with smaller standard errors than self-complete methods (Snowball and Willis, 2011). Authors of previous DCE studies involving adolescents advocated face-to-face data collection to allow clarification of information where needed (de Bekker-Grob et al., 2010, Hofman et al., 2014). Due to the relatively small potential sample an online data collection method was chosen to provide a wider representation of people affected by hypodontia and a greater number of respondents to increase the precision of estimates; however, the observed survey provided an opportunity to compare preferences between the two data collection methods to determine the suitability of online DCE methods with adolescents.

Hypotheses regarding preferences for specific respondent groups were developed from clinical experience, existing literature and results of the pilot study. The first hypothesis was that adolescents are less tolerant of a long waiting time and more tolerant of severe discomfort than parents. The pilot study and clinical experience suggested adolescents are keen to start treatment and willing to ‘put up’ with discomfort to get the best outcome. On the other hand, parents reported a desire to avoid discomfort as a result of being protective of their child. Some parents also alluded to having to ‘listen to complaining’ if treatment caused discomfort or prevented participation in regular activities. The second hypothesis was that parents have a stronger preference to avoid severe problems than adolescents. Neuroeconomic research suggests adolescents and parents may process risk differently (Hartley and Somerville, 2015), which may lead to a different understanding of long-term implications of problems. Verbal feedback in the pilot study supported the suggestion that although severe problems were important to both groups, parents were more influenced by problems during treatment than adolescents.

Previous studies suggest there is a change in understanding with age and experience of treatment (Meaney et al., 2012). It was hypothesised that as older adolescents are generally further into treatment and have already invested time and raised expectations, they may demonstrate a stronger preference for an improved appearance. On the other hand, younger adolescents are likely to be more treatment-naïve and may be apprehensive of the unknown, leading to a stronger avoidance of discomfort. This was supported by comments from parents in the pilot study, who felt their child’s preferences had changed over the course of treatment (section 4.4.4.2). Evidence suggests severe hypodontia may be associated with greater functional impairment (Wong et al., 2006a), while those with mild and moderate hypodontia predominantly reported concerns with appearance (Chapter 3). For this reason, it was hypothesised there would be a difference in preference for outcomes, with greater value placed on improvement in bite by those with severe hypodontia.

There is little explicit evidence about the impact of dental anxiety and hypodontia-related quality of life impact on decision-making for dental treatment. Following consultation with stakeholders it was agreed that more anxious respondents may be keen to delay treatment and less willing to undergo treatment that carries a risk of severe discomfort and problems. Evidence from the methods examining hypodontia care (Chapter 3) and cognitive feedback from the pilot study (Chapter 4) suggested those with higher quality of life impact from hypodontia are keen to undergo treatment regardless of the possible adverse effects. The interviews suggested a strong desire to start treatment soon, even where this might make treatment longer.

In addition to preference examination, the effect of choice behaviour on preference estimates was examined in terms of selection of the opt-out and rating of task difficulty. Different approaches have been advocated for analysing opt-out responses depending on the framing of the alternative (Campbell and Erdem, 2019). For this study, the characteristics of those who selected no treatment were examined to provide information that might help clinicians identify patients who might be less keen to undergo treatment. Choice task difficulty ratings were used to identify types of respondents who might find information processing or trade-off tasks difficult, as this is important to inform future decision support tools.

The proposed use of DCE with adolescents and adolescent-parent dyads is relatively new so methods to test the validity and reliability of the survey were included to increase confidence when translating the findings into clinical practice. Content validity was tested through further respondent feedback to build on the preliminary findings from the pilot. For the observed survey, respondents were asked which attributes were most important in a post-survey interview (de Bekker-Grob et al., 2010, Hofman et al., 2014) as an approximate measure of convergent validity. Test-retest of a choice task and the whole survey was used to examine agreement between choices (Wang et al., 2017). It is acknowledged a test-retest method is limited by the potential learning effect gained from completing choice tasks and temporal instability, both of which may cause a change to preferences (Ozdemir et al., 2010). The complexity of clinical decision-making for hypodontia has been demonstrated throughout this research and it was expected that this might present challenges for extracting revealed preference data to test external validity. On balance it was felt that the potential to examine actual treatment choice was a valuable opportunity, so this information was sought.

5.2 Aims and objectives

The aim of the survey was to examine the decision-making process between adolescent-parent dyads and measure the preferences of adolescents and parents, individually and together, for hypodontia care.

The objectives were:

- To explore the decision-making process for adolescents and parents individually and together, including how joint preferences are negotiated and whether making trade-offs helps adolescents and parents think about treatment
- To examine preferences for hypodontia care and determine whether preferences differ between respondent based on pre-defined hypotheses
- To examine characteristics associated with choosing no treatment
- To identify which groups of people might benefit from trade-off tasks and who finds them challenging
- To evaluate DCE method in terms of reliability and validity for adolescent and joint adolescent-parent preference elicitation
- To synthesise the findings to identify the key implications for clinical practice and future research

5.3 Design and Methods

The design was a cross-sectional survey using a preference elicitation instrument to collect data with two different approaches:

- **Online:** Online survey to elicit population level preferences for hypodontia care from adolescents with hypodontia and parents of adolescents with hypodontia, individually and together
- **Observed:** Face-to-face individual and joint survey, completed by adolescent-parent dyads under observation to explore decision-making experience

5.3.1 Sample

The target population included 1) Adolescents (aged 12-16 years) with hypodontia of any severity at any stage of treatment and 2) Parents of adolescents with hypodontia.

Online survey: The sample provided three respondent groups; 1) Adolescent alone 2) Parent alone 3) Adolescent-parent dyad. More than one respondent from a family was able to participate, for example a parent alone then an adolescent alone, or a mother-adolescent together then the father separately. In this case respondents were requested to create a code (using initials and date of birth of the adolescent with hypodontia) to allow matching of family members.

Observed: Adolescent-parent dyads completing the survey individually and together.

5.3.2 Sample size

5.3.2.1 Preference estimates

The number of observations, calculated from both the number of participants and the number of choice tasks, constitutes the sample size for DCEs. As previously outlined, calculating the ideal sample size for quantitative analysis of stated preference experiment data is complex (Orme, 2010, de Bekker-Grob et al., 2015) but for this research, sample size was limited by the relatively low prevalence of hypodontia and consequently, the limited number of potential participants. ‘Rules of thumb’ for sample size for DCE include 300 respondents in total or between 50-200 per group where subgroup analysis is planned (Orme, 2010), or 20 respondents per questionnaire version (Lancsar and Louviere, 2008).

Sawtooth Software’s CBC manual (2016) recommends the minimum sample size for aggregate-level full-profile is:

$$\mathbf{n \geq 500c / (t\ a)}$$

where n = number of respondents
t = number of tasks
a = alternatives per task
c = number of analysis cells

In this research, two-way interactions may be used so *c* is the largest product of the levels of any two attributes (3x3). Hence, the minimum sample size would be (500x9)/56 =80. Based on these recommendations, a pragmatic approach was taken, aiming to recruit as many participants as possible in a 6-month period, with a target of around 200 participants.

5.3.3.2 Observation of decision-making

Guidance for exploratory qualitative methods using a descriptive approach recommends a preliminary sample size of 15-30 (Moser and Korstjens, 2018). An initial recruitment target of 15 dyads was set with purposive sampling based on age, gender, severity of hypodontia and stage of treatment. Data analysis was undertaken contemporaneously to assess data saturation in terms of new observations about behaviour across dyads to inform ongoing recruitment.

5.3.3 Setting

Online Survey: To increase recruitment numbers and diversity in the sample, sites across England and Wales were invited to act as Participant Identification Centres (PICs). An email was sent via the British Orthodontic Society Consultant Orthodontist Group requesting expressions of interest. Consultant Orthodontists from 22 sites in 20 NHS Trusts replied. Consultants from Newcastle and Southend volunteered to participate but failure to finalise R&D approval in time for recruitment prevented their inclusion. In total, twenty NHS hospitals providing hypodontia care acted as PICs (Appendix Figure 7).

Observed: Three hospitals in Yorkshire (LDI, SLH, York). Further information about these settings is provided in Section 1.7.

5.3.4 Materials

Development and testing of the preference elicitation instrument is described in Chapter 4. A copy of the survey is given in the Appendix (Figure 8). The preference elicitation instrument was programmed and administered by the survey company Accent (http://www.accent-mr.com) following design instructions provided by the researcher. Responses were automatically collected from the survey, collated and formatted by Accent. The survey was accessible from mobile telephones, tablets, laptops or desktop computers. Two versions of the survey were created, adolescent and parent, to enable appropriate wording for questions for the respondent type. For the online survey, a section with study information and consent was included at the start of the study. This provided written information about the study alongside a study information video where the researcher explained the purpose and details of the survey. All additional materials were co-developed with PPI contributors specifically for use in the study.

Online Survey: Respondents selected who was completing the survey at the start and this directed them to the appropriate version. Providing individual links for access would have required the direct clinical care team to collect and share contact details or alternatively, respondents would have been required to apply for a survey link. Both approaches were judged to be a potential barrier to participation based on PPI contributor feedback. Instead the survey was openly accessible to the public but the survey was not evident on internet search engines and required a specific web address that was provided on a postcard to eligible people. Measures taken to minimise the likelihood of ineligible or repeat respondents included screening questions and a request for an email address for the £5 incentive voucher.

Additional materials used in the online survey included:

- Information for Participant Identification Centres: Members of the direct clinical care team at each site were given summary information about the study including the purpose of the study, a brief outline of the methodology and the eligibility criteria
- Recruitment poster for waiting room
- Recruitment postcards for distribution to all potential survey respondents fulfilling the eligibility criteria. The postcard contained brief information about the study and directed participants to the DCE survey website, which contained further details and the participant information video.

Observed: A trio of links was used by the researcher for each dyad to create linked adolescent, parent and joint responses. The individual adolescent and parent versions contained demographic and background questions alongside the choice tasks to allow direct comparison of self-reported data between dyads. The joint version contained only the eight choice tasks. Within the dyad, each survey contained the same choice tasks to allow direct comparison, but the tasks were randomly ordered.

Additional materials used in the observed study included:

- Participant information sheets (adolescent and parent versions)
- Consent and assent forms
- Standardised data caption sheet to allow clinical care team to anonymously report actual treatment options and chosen treatment
- Topic guide to assess respondents experience of completing choice tasks and their perception of the decision-making process (Table 5.1). Questions were modified iteratively throughout data collection.

Table 5.1: Preliminary questions in the topic guide for the post-survey interview

Questions for the adolescent first, then the parent

How did you choose between options?
Was there anything that was most important to you?
Was there anything that was not at all important to you?
Was there anything that would make you (definitely) pick a treatment?
Was there anything that would make you (definitely) **not** pick a treatment?
Has answering these questions made you think about your dental treatment differently?

Question to both respondents together

How did you pick together?
Was there anything you did not agree on?
If you disagreed, how did you decide?

Did answering these questions make you talk more about what is important to you both?

5.3.5 Online survey methods

Identification: The direct clinical care team at each site identified potential participants during routine dental appointments. For feasibility reasons identification was opportunistic and no additional measures, such as database searching, were used to identify suitable patients. All those fulfilling the eligibility criteria were given brief verbal information by the clinical care team and invited to participate through provision of the recruitment postcard. The direct clinical care team was asked to record the number of people approached and any stated reasons for refusal to participate. No personal or clinical data about potential participants was requested from the clinical care team.

Enrolment and consent: Participants were able to access the survey website at their own convenience. Written information was provided at the start of the survey alongside a short video of the researcher explaining the research and survey. A small incentive (£5 voucher) was offered to encourage enrolment and completion of the survey. Screening questions were included at the start of the survey to confirm eligibility to participate. Explicit consent was obtained from the respondent and for those under 16 years old completing the survey alone, confirmation of parental consent was requested.

Respondents were asked separately for consent to be included for selecting a random sample for retesting.

Data collection: Response data was immediately transferred to Accent. Email addresses were removed from the research data immediately and stored separately to ensure anonymity. Data was accessible to the researcher through a password protected portal. The study identifier was recorded for all respondents who indicated that they were willing to complete a retest survey. Every 2-4 weeks a 20% sample was randomly selected using a random number generator (https://www.random.org/). The anonymous identifier was sent to Accent who matched it to the email address and sent a unique link to the respondent via email. The 2-4-week period between initial and repeat survey completion was judged to be sufficient time to minimise recall of previous answers but not long enough to allow significant changes in preferences. The retest survey aimed to test intra-respondent reliability and contained only the DCE choice tasks along with brief information and further consent. The retest took less than 10 minutes to complete.

5.3.6 Observed survey methods

Recruitment: Potential participants were identified by the clinical care team from clinic lists during routine appointments. Verbal and written study information was provided by the clinician, or researcher if present. All those fulfilling the eligibility criteria were invited to participate and the number of people approached and any stated reasons for refusal to participate were recorded. Permission was sought for the researcher to contact the potential participant via telephone or email. If agreed, contact details were recorded.

Enrolment: Potential participants were given at least one week to decide if they wished to participate. The researcher provided further information if requested before enrolling participants. Those who agreed were provided with a date and location for the survey. Reimbursement was provided for travel if participants attended at a separate time to their dental appointment. An incentive for participation (£10 voucher) was provided for the adolescent only. Consent for participation was obtained on the day of data collection by the researcher. This provided additional time to consider participation and an opportunity for the researcher to answer any further questions.

Data collection: Survey completion was undertaken in a private room with the researcher present to observe and audio-record the process. Field notes were taken and the audio-recording was used to check the field notes during analysis; however, audio was not transcribed in full. The adolescent and parent first completed individual surveys separately using tablet devices. After this the adolescent-parent dyad then completed the joint survey, which only included the choice tasks, together on one tablet device. Dyads were encouraged to discuss the negotiation process and their choices aloud. The order of choice tasks differed between the adolescent, parent and joint survey but same choices were given. To assess the validity of the DCE, clinical information about treatment options and choices was required. To obtain this information the researcher provided the name of the participant to the direct clinical care team who extracted the necessary information using a standardised data collection sheet. No personal or clinical data was collected that enabled identification of the patient.

5.3.7 Data analysis plan

Anonymised data was collated in Microsoft® Excel v16.23 (2016). Respondent data was analysed using IBM SPSS® Statistics 22 (2017). Preference data analysis was performed using Stata/IC v13.1 (StataCorp, Texas, USA) with support from a supervisor (David Meads) and an expert advisor (Joachim Marti). The Stata coding file is available on request.

5.3.7.1 Characteristics of sample

Respondent characteristics and information about the decision-making context were reported descriptively to provide an understanding of the sample. Logistic regression was used to examine the respondent characteristics associated with dental anxiety, hypodontia-related quality of life impacts and decisional uncertainty.

5.3.7.2 Qualitative data analysis

Qualitative data was analysed initially using a descriptive approach to understand the experience of those completing the preference elicitation instrument. Field notes were used to create a framework for analysing the data to address the objectives. This included categories relating to: 1) individual (adolescent / parent) respondent behaviour; 2) approach to joint decision-making including discussion and negotiation; 3) perceived usefulness of completing choice tasks; 4) self-reported preferences; 5) other. Themes developed about the way individuals and dyads make decisions, and the role of trade-off tasks in changing adolescent and parent decision-making.

Following this, typologies were developed to explain how adolescent-parent preference selection occurred in the context of a joint stated preference experiment (Starks and Trinidad, 2007, Noble and Mitchell, 2016). Typical cases were examined to see if the typologies reflected the actual selection of joint choices compared to individual choices.

5.3.7.3 Preference data analysis

Quality of responses

The quality of responses was examined based on survey completion time and task non-attendance. Time taken to complete the survey was reportedly descriptively. ANOVA was used to identify differences in mean survey completion time between different respondent groups. No existing criteria for determining valid response time limits were identified in the literature, so the minimum valid time was determined based on time taken for survey completion under observation to assess the minimum time required to read instructions, consider attribute-levels and complete choice tasks and other survey questions, and by examination of survey completion time data to identify outliers.

Task non-attendance was suggested by 'straight-lining' (Kim et al., 2019), which for this study task was defined as selection of Treatment A or Treatment B in all eight choice tasks. The number of respondents selecting Task A was reported for the sample to look for evidence of straight-lining; however, it was recognised selection of A or B in every task could represent genuine preference, so respondents demonstrating this behaviour were examined further to determine whether they should be excluded from the analysis. This included examination of time taken on the survey, straight-lining in contextual questions and confirmation attribute descriptions were accessed.

Model specification

Different models (conditional logit main effects, alternative specific logit (ASC) conditional logit, mixed logit) were tested to identify the most appropriate model for preference data analysis. Preference data was analysed using best practice guidance (Hauber et al., 2016) to estimate preferences for attributes of hypodontia care and examine differences in preferences based on respondent characteristics. The conditional log (CL) relates choices to the attribute-levels defining the alternatives based on the assumption that error terms are independent and identically distributed across observations and there is no preference heterogeneity between respondents (Adams et al., 2015). Preference heterogeneity in the sampled population can be assessed by data segmentation, which uses a different model for each attribute level range, or attribute segmentation, which generates separate betas for different attribute level ranges. The challenge for this approach is selecting valid segmentation criteria and attribute-level range cut-offs.

Alternatively, the mixed logit yields a mean effect and standard deviation of effects across the sample. Mixed logit models are better able to take into account individuals' inter-related systematic and random components of alternatives in the choice set, but the ability to capture more true behavioural variability in choices means the mixed logit requires better quality data than the CL (Hensher and Greene, 2011). The latent class (LC) models assumes attributes of the alternatives can have heterogeneous effects across different classes of respondents and by segmenting the sample into classes, it is possible to obtain preference weights for different classes. Hence, LC can be used to test the influence of individual characteristics on probability of class membership and ratios of change in preference weights can be used to overcome scale differences to compare across classes.

Goodness-of-fit statistics informed model selection (Hauber et al., 2016). The likelihood ratio chi-square test was used to determine whether including attribute-level variables significantly improved the model fit compared with a model without any attribute-level variables. Minimising chi-square is the equivalent of maximising log likelihood, hence a lower chi-square value is preferable. The Akaike information criterion (AIC) and Bayesian information criterion (BIC) were used to evaluate the plausibility of models, with lower numbers suggesting a better fit (Burnham and Anderson, 2004).

Dummy coding was selected over effects coding for ease of interpretation (Daly et al., 2016). The level expected to be most preferred a priori was assigned as the omitted (reference) level. Attribute-levels were initially coded as categorical to allow examination of functional form to determine whether waiting time and treatment time should be recoded to continuous (Hauber et al., 2016).

Estimation of average preferences for hypodontia care

Following selection of the most appropriate model, preference weights (regression β-coefficients) for different attribute levels were calculated. Estimates were reported relative to the reference level, with 95% confidence interval. Distribution curves for each of the attribute-levels was calculated using the post-estimation command "mixlbeta", which uses the method proposed by Revelt and Train (2000).

Willingness to Wait (WTW) (Marshall et al., 2018) calculations were planned where time attributes demonstrated a linear functional form. Where attributes demonstrated a non-linear functional form, preference weights for specific levels were directly compared as ratios. Confidence intervals were calculated using the post-estimation command "wtp", developed by Arne Hole (2007), which uses the Krinsky and Robb method.

Examination of heterogeneity in preferences

Expected differences in preferences between respondents are discussed in the introduction to this chapter based on evidence from the literature and the pilot study (Section 5.1). Specific hypotheses were developed about expected differences in preferences between certain respondents (Table 5.2). To test these hypotheses the respondent characteristic was interacted with the specified attribute in a mixed logit model (Sivey et al., 2012). The coefficient on the interaction represented the additional degree to which the characteristic affected the preferences relative to the main effect. To create interactions, categories were used:

- **Respondent type:** Adolescent; parent; dyad
- **Age of person with hypodontia**: Younger (12-14 years); Older (15-16 years)
- **Severity of hypodontia:** Mild-moderate; severe
- **Stage of treatment:** Treatment-naïve (pre-treatment); treatment-experienced (mid or end-treatment)
- **Adolescent dental anxiety**: Dentally anxious (Score agree-strongly agree to either question about adolescent's dental anxiety); Not dentally anxious *(*Score ambivalent-disagree-strongly disagree) to both questions about adolescent's dental anxiety.
- **Hypodontia-related quality of life impact**: High; moderate; low (based on summary score of ratings for the eight OHRQoL questions)

The latent class was used as an alternative approach to examine the contribution of respondent characteristics to preference estimates. The number of classes was selected based on ability of classes to explain decision-making behaviour and goodness-of-fit statistics. Preference weights derived from the latent class model were not compared across classes due to potential differences in scale parameter, hence, preference weights were used only to describe the characteristics of preferences within the class (Hauber et al., 2016). The class-probability function was used to examine which respondent characteristics contributed to class membership relative to the omitted class.

Table 5.2: Pre-defined hypotheses based on expectations about differences in preferences between respondents (for rationale see Section 5.1)
Hypothesis 1: Adolescents are less tolerant of a long waiting time and more tolerant of severe discomfort than parents
Hypothesis 2: Parents will have a stronger preference to avoid severe problems than adolescents
Hypothesis 3: Older adolescents have a stronger preference for improved appearance, while younger adolescents are more avoidant of severe discomfort
Hypothesis 4: People in treatment have a stronger preference for improvements in appearance and bite, while treatment-naïve people are more avoidant of discomfort
Hypothesis 5: Respondents with severe hypodontia have stronger preference for improved function than those with mild-moderate hypodontia
Hypothesis 6: Dentally anxious respondents are more willing to accept a long wait for treatment, but more avoidant of severe problems and severe discomfort
Hypothesis 7: People reporting a high quality of life impact from hypodontia would be more willing to accept severe discomfort, severe problems and a long treatment time but less willing to wait for treatment

<u>Examination of choice behaviour</u>

Choice behaviour was examined in relation to opt-out (selection of No Treatment) to help identify factors that may prevent adolescents accepting treatment for hypodontia. Choice task difficulty ratings were examined to identify whether certain respondent factors were associated with difficulties with the choice tasks.

Opt-out responses were analysed using a mixed approach:

- Descriptive reporting of the use of the opt-out by respondents
- Logistic regression to examine whether characteristics (respondent type, data collection mode, characteristics of the adolescent with hypodontia (age, gender, severity of hypodontia, dental anxiety and quality of life impact of hypodontia)) were associated with selection of No Treatment
- Grouping of respondents into those always opted-out (non-consumers) and those

who never opt-out, and inclusion of the groups as a predictor of latent class membership
- Creation of a third alternative with clinically appropriate levels (Zero months waiting time and treatment time; No discomfort and problems during treatment; Appearance and bite unchanged) then use of the ASC logit to examine whether the preference for the opt-out was significant
- Creation of categories of respondents based on those who always, sometimes and never opt out to allow interaction with attribute-levels in a mixed logit model to test the effect of opt-out behaviour on preference estimates

Choice task difficulty ratings were analysed using a mixed approach:

- Descriptive reporting of ratings of choice task difficulty
- Logistic regression to examine whether characteristics (respondent type, data collection mode and characteristics of the adolescent with hypodontia (age, gender, severity of hypodontia, dental anxiety and quality of life impact of hypodontia)) were associated with a rating of Difficult
- Creation of a category for respondents rating ≥6 choice tasks as Difficult and inclusion of this as a predictor of latent class membership. Categorisation was based on examination of the spread and distribution of difficulty ratings.
- Calculation of utility difference between alternatives in each choice task using preference weights from the mixed logit model to provide an objective estimate of task difficulty. This was compared to utility balance and self-reported task difficulty using Pearson correlation

5.3.3.4 Evaluation of DCE as a preference elicitation tool

The instrument and data collection method were evaluated using combined qualitative and statistical methods to assess validity and reliability.

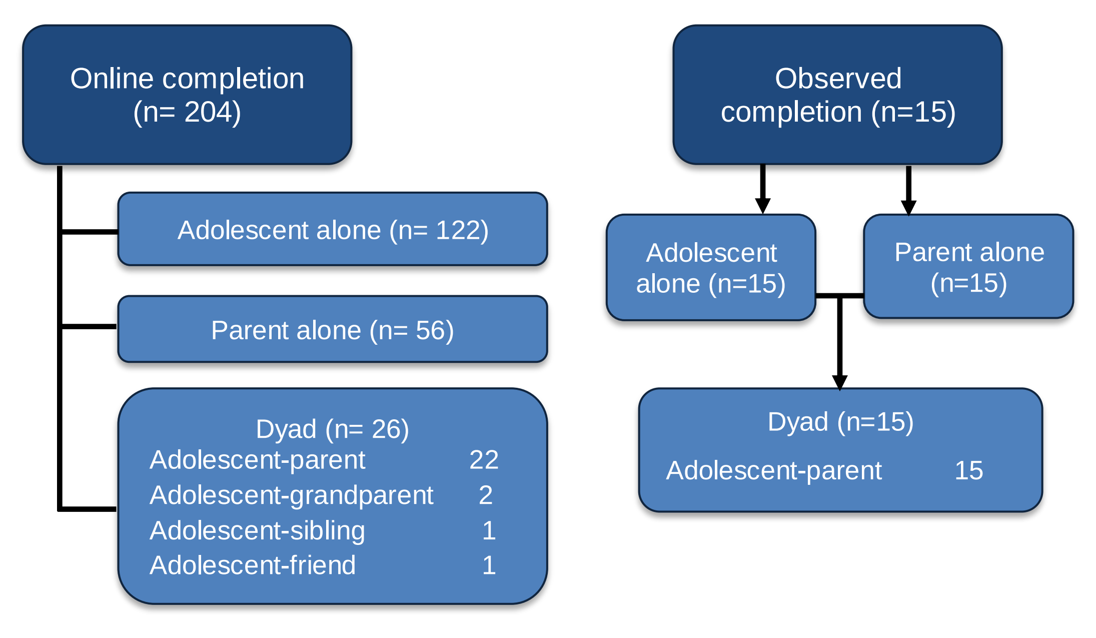
1. Examination of DCE choice task validity based on concordance between preference estimates and those expected a priori and respondent feedback in the post-survey interview (de Bekker-Grob et al., 2010, Hofman et al., 2014)
2. Inclusion of data collection mode as an interaction to examine the effect on preference estimates
3. Sensitivity analysis to determine the effect of excluding poor quality respondents
4. Examination of repeat choice task in all surveys
    - Description of number of tasks in agreement

  - Intra-examiner agreement tested with intra-class correlation coefficient (Two-way mixed, absolute agreement) (Koo and Li, 2016)
  - Relationship between mean intra-examiner agreement and difference in utility across alternatives in the repeat choice task (calculated from preference estimates), tested with Pearson correlation
  - Descriptive examination of intra-examiner concordance between initial and repeat rating of difficulty and selection of No Treatment
5. Examination of repeat online survey
   - Description of the number of tasks in agreement per respondent
   - Intra-examiner agreement tested with intra-class correlation across all observations (Two-way mixed, absolute agreement) (Koo and Li, 2016)
   - Relationship between time taken to complete repeat survey and number of tasks in agreement, tested with Pearson correlation
   - Relationship between mean intra-examiner agreement and difference in utility across alternatives in the repeat choice task (calculated from preference estimates), tested with Pearson correlation
   - Descriptive examination of intra-examiner concordance between initial and repeat rating of difficulty and selection of No Treatment
6. Examination of actual treatment choice compared to stated preference findings as an indication of external validity

5.4 Results

Data collection for the online survey ran from May-December 2018. In total 204 people completed the survey including 122 adolescents alone, 56 parents alone and 26 joint completions by dyads. The observed survey was undertaken between June 2018-January 2019. In total 15 adolescent-parent dyads were included (Figure 5.1). The majority of parent respondents, completing the survey alone or with their child, were mothers.

Figure 5.1: Respondents for the preference elicitation survey



Matching codes provided for the online survey indicated more than one completion from within the same family for 11 sets of respondents. For nine of these cases, the survey was completed by an adolescent alone and a parent alone separately. For two cases, the survey was completed by mother, father, adolescent and sibling who also had hypodontia.

5.4.1 Response rate

Participants were recruited for the online survey from across multiple sites in the England and Wales. The direct clinical care teams were asked to record the number of people approached to participate; however, this was not recorded by all units due to numerous staff members being involved in recruitment. It was therefore not possible to calculate response rate; however, 1000 recruitment postcards were distributed to the recruitment sites suggesting approximately 20% response rate. No online surveys were abandoned incomplete.

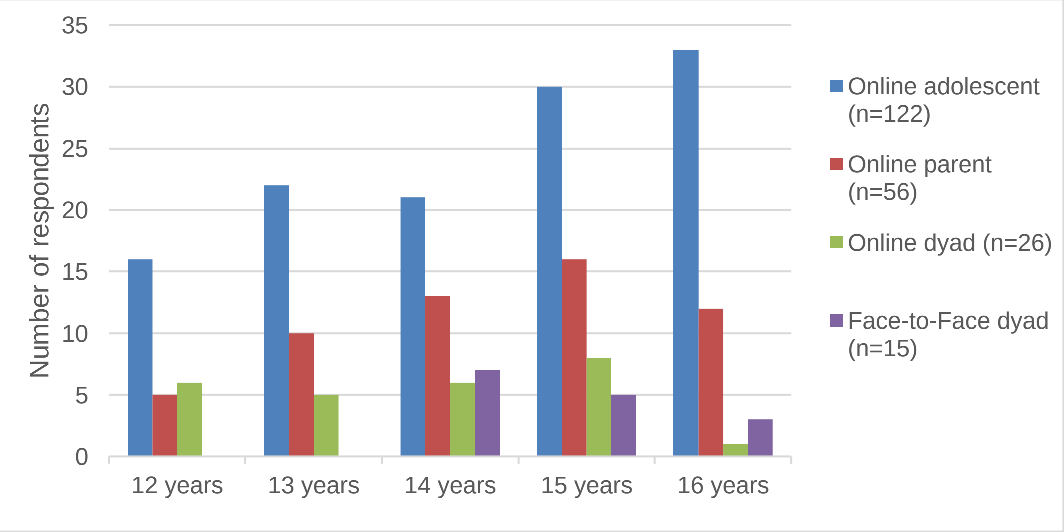
Similarly, multiple clinicians were involved in identifying and inviting dyads to participate in the observed survey so accurately estimating response rate is challenging, but of those who initially agreed to be contacted about the research, approximately half agreed to participate. The most common cited reasons for not wanting to participate by parents was the time required to attend an additional or longer hospital visit. For the adolescent group, the most common reason not to participate was being ‘shy’, as reported by the parent. No-one who attended to complete the survey requested to withdraw during or after data collection.

5.4.2 Sample characteristics

The age of the adolescent with hypodontia, as reported by the adolescent or parent, is given in Figure 5.2. Adolescents completing the survey alone were generally older (15-16 years), while completion of the online survey by dyads included proportionately more adolescents. This may reflect the complexity of the DCE choice tasks and the requirement for a parent to support a younger respondent.

All respondents in the observed survey were 14-16 years despite attempts to sample across the age range. This may reflect the most common age for dental treatment and increasing willingness to participate in the research with age.

Figure 5.2: Age of adolescent with hypodontia for respondents of the preference survey

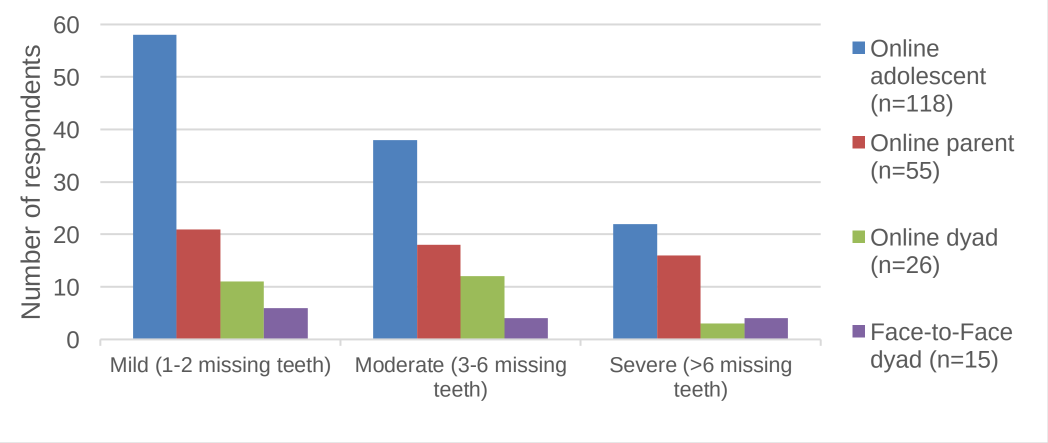


The gender of the adolescent with hypodontia, as reported by the adolescent or parent, was 64% female and 36% male. This approximately reflects the expected female to male proportions given the reported 3:2 female predominance. For both methods of data collection, the sample was principally white (96%) and the majority of respondents lived in England (95%). This homogeneity in the sample populations has implications for the generalisability of results. One recruitment site for the observed survey serves a high minority ethnic population so it was hoped this would promote greater inclusion of non-White participants; however, there were difficulties in getting a more diverse sample. Fewer minority ethnic adolescents were identified as eligible and for some who were, participation was prevented by parents being unable to read English to an adequate standard, or the adolescent did not want to participate.

Adolescent self-reported proposed education plans suggest the majority of adolescents (66%) plan to continue education to A-level or beyond (Appendix Figure 9). The proportion expecting to attend university (28%) is lower than the current UK national average of around 33%, although this increases to 31% if those who indicated uncertainty about future plans are removed. The number of parents (n=24) reporting a higher degree was higher than the UK national average.

The severity of hypodontia in the sample is given in Figure 5.3. Four adolescent and one parent respondent in the online survey answered ‘Not sure’. The overall proportions of mild (45%), moderate (34%) and severe (21%) hypodontia are not reflective of the usual distribution. The observed survey approach allowed respondent answers to be checked against clinical data provided by the dental team to assess the validity of the question. Three adolescents were unsure how many teeth they were missing but in all cases the parent provided the correct answer. There was disagreement between one adolescent-parent dyad about the number of missing teeth, with the parent answering correctly. This suggests there might be some errors in self-reported data regarding the severity of hypodontia by adolescents.

Figure 5.3: Survey respondents’ reported severity of hypodontia (based on self-reported data for the online survey and corrected data for the observed group)

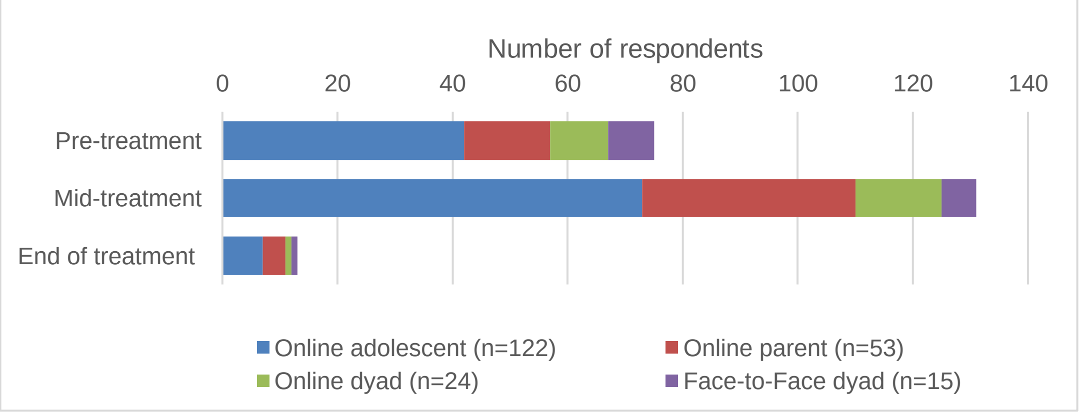


Twenty-one respondents from the online survey (12 adolescents, 4 parents and 5 dyads) and 6 respondents from the observed survey (4 adolescents, 2 parents) were not sure of the location of their own / their child's missing teeth. Observation of the respondents completing the survey indicated confusion about the location of missing teeth, suggesting this data may be inaccurate. The proportion of respondents reporting missing teeth in each area of the mouth was 61% in the anterior maxilla, 39% in the posterior maxilla, 40% in the anterior mandible and 43% in the posterior mandible. The high proportion of missing teeth reported in the anterior maxilla is likely to reflect the relative frequency of lateral incisor agenesis and increased awareness of missing anterior teeth. The proportion reporting absent posterior mandibular teeth is lower than expected based on frequency of second premolar agenesis; however, it is possible respondents were unaware of missing teeth if primary molars were present.

5.4.3 Decision-making context

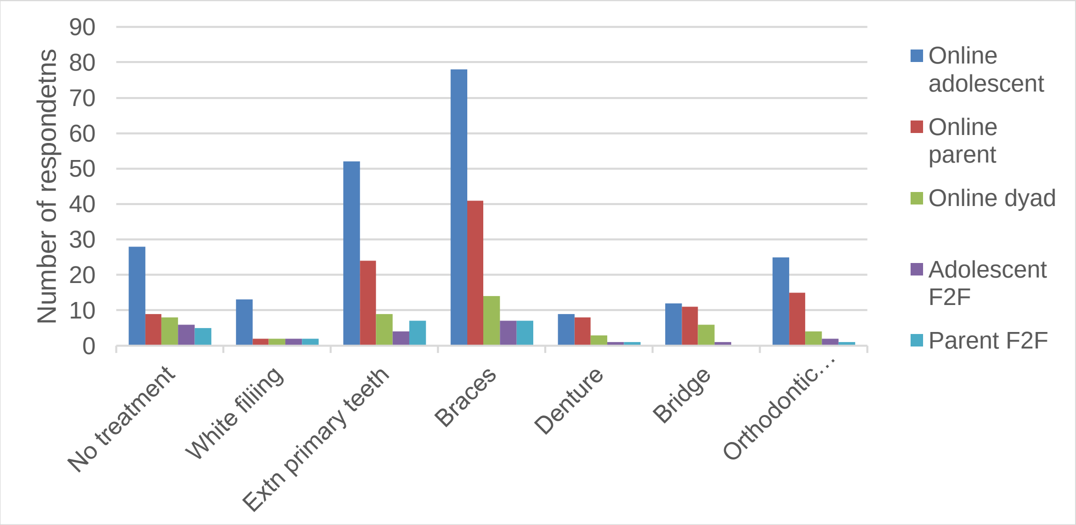
The majority of respondents (60%) were in active treatment for hypodontia (Figure 5.4). Only 13 (6%) adolescents had completed dental treatment; this reflects the extended duration of hypodontia treatment and it is also possible respondents might perceive orthodontic retention to be active treatment. Age correlated with stage of treatment; younger respondents were more commonly at the start to middle of treatment and those at the end of treatment were commonly 15-16 years.

Figure 5.4: Survey respondents' reported stage of treatment (based on self-reported data for the online survey and corrected data for the observed group)



Treatment experience varied across the respondents (Figure 5.5). Orthodontic treatment was the most commonly reported treatment, followed by extraction of primary teeth. The number of respondents who had undergone restorative camouflage (white fillings) to alter the size and shape of teeth was lower than expected. Comparison between the treatment reported by the adolescent-parent dyads in the observed survey found agreement in 10 dyads and discordance in 5 dyads.

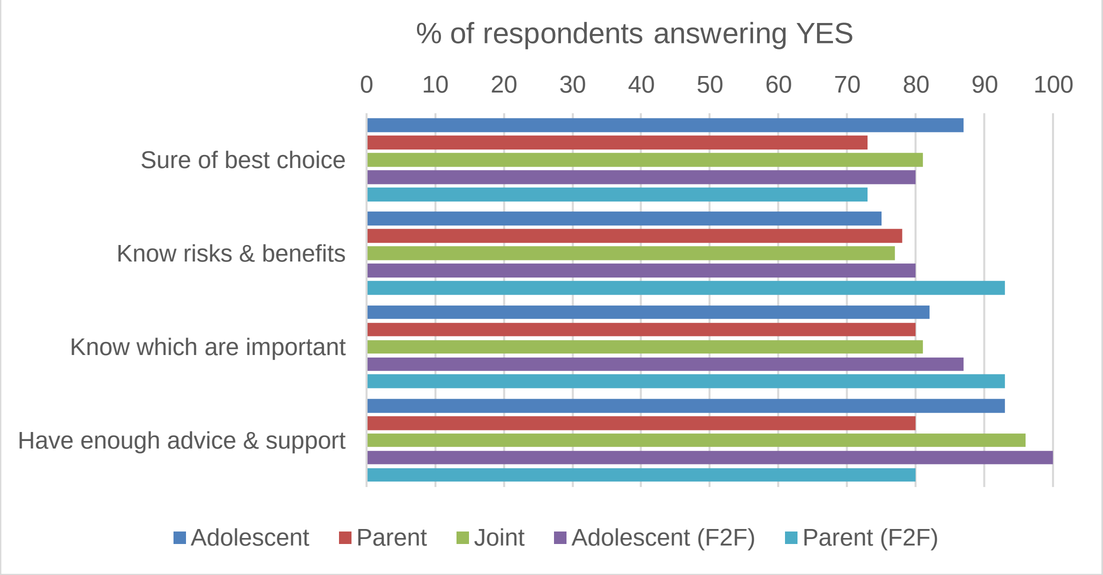
Figure 5.5: Survey respondents' self-reported experience of dental treatment for hypodontia



The majority of respondents indicated that dentists (97%) and parents (93%) frequently help with decisions about dental treatment (Appendix Figure 10). Fewer adolescents reported that friends were important in decision-making than expected; 14 (10%) adolescents reported a role of friends compared to 19 (27%) parents. Observation of survey completion suggested 'Other people with hypodontia' was ambiguous; some respondents interpreted it as parents or friends with missing teeth, rather than it being a formal or informal peer-support network as intended.

The SURE tool, which includes four questions to measure self-reported certainty in decision-making, suggested people were most confident about having enough advice and support. The areas with least certainty were knowing the risks and benefits and which are of these are important to the adolescent with hypodontia. Generally, parents expressed greater uncertainty than adolescents (Figure 5.6). Respondents were grouped into two based on those who answered 'Yes' to all four questions ('certain') and those who answered 'No' to one or more questions ('uncertain'). Logistic regression found being 'uncertain' was more likely in younger adolescents and parents of younger adolescents, females, those with moderate hypodontia, and those completing the survey online (Appendix Table 22).

Figure 5.6: Survey respondents' beliefs about decision-making



Adolescent and parent anxiety about dental treatment are displayed in Appendix Figure 11. The majority of adolescents (54%) disagreed or strongly disagreed with the statement '*I feel anxious about having any dental treatment with my normal dentist'*, indicating no anxiety about normal dental treatment, while 21% and 9% agreed and strongly agreed, suggesting 30% have some degree of dental anxiety. Generally, adolescent anxiety about dental treatment for hypodontia was higher than for routine dental care. In total, 111 respondents (47%) indicated some degree of anxiety and fewer (42%) suggested no anxiety by disagreeing or strongly disagreeing with the statement. Parental anxiety about dental treatment was similar, with 39% reporting some anxiety, 42% indicating no anxiety and 19% demonstrating ambivalence.

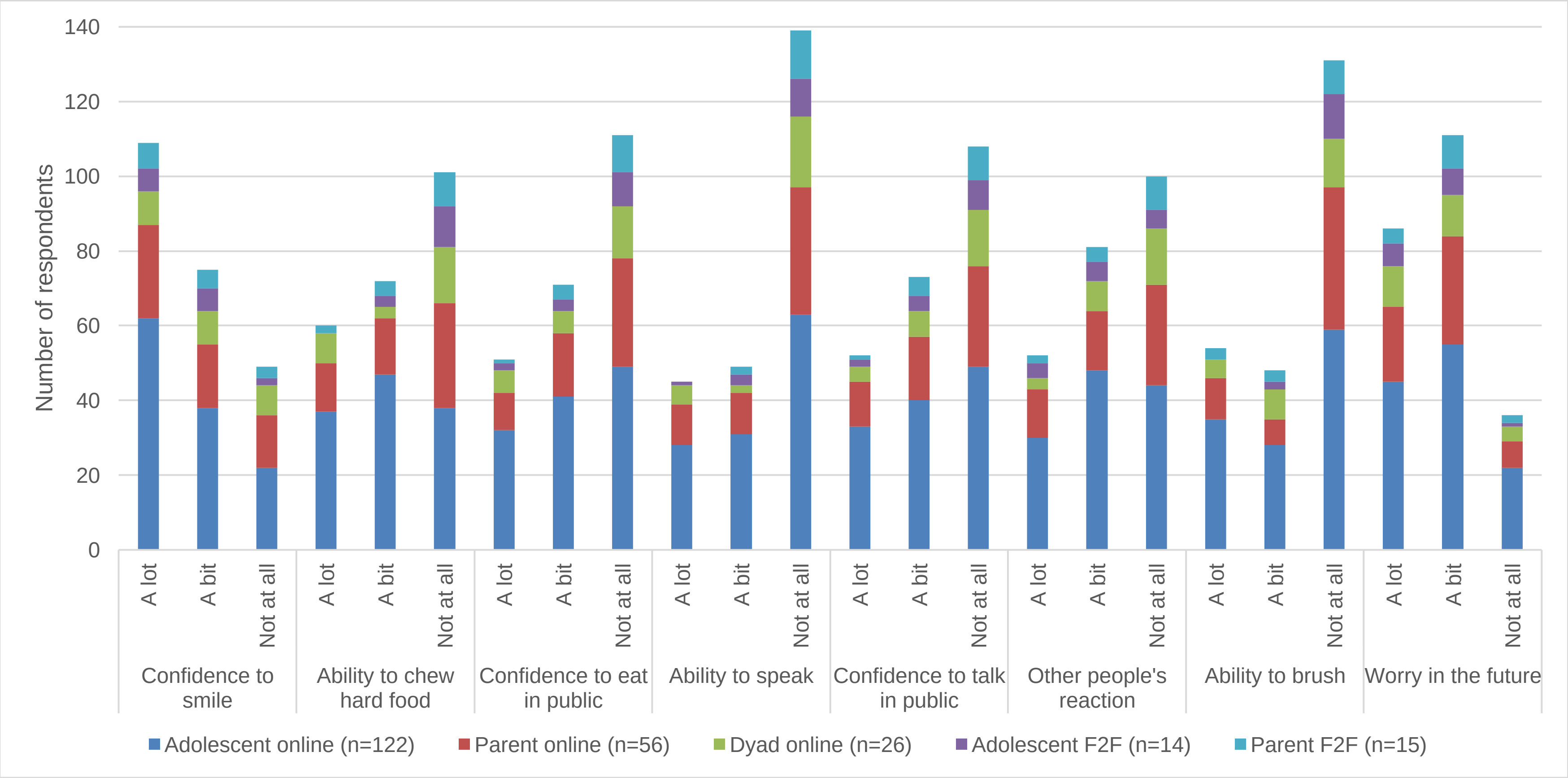
For fourteen dyads, it was possible to compare adolescent-reported anxiety to the anxiety reported by their parent for normal dental treatment and dental treatment for hypodontia. This showed that for normal dental treatment, 43% of adolescents rated their anxiety higher than their parents rated their anxiety. For hypodontia treatment the discordance was lower (29%), suggesting parents recognised this anxiety more.

Adolescents with moderate and severe hypodontia were found to be 2.3 and 2.6 times more likely to be anxious than those with mild hypodontia. Other characteristics associated with dental anxiety were younger adolescents and males (Appendix Table 23).

The reported impact of hypodontia on oral health-related quality of life (OHRQoL) domains is given in Figure 5.7. Greatest effect on everyday life was reported for confidence to smile (47% a lot, 32% a bit) and worry in the future (37% a lot, 47% a bit). Functional issues with eating were much less frequent, with ability to eat causing problems for 56% (25% a lot, 31% a bit) and confidence to eat in public causing problems for 52% (22% a lot, 30% a bit). The least impact was reported for ability to speak (no impact in 60%) and ability to brush (no impact in 56%).

A high quality of life impact from hypodontia was strongly associated with severity of hypodontia; those with severe hypodontia were 4 times as likely to have high quality of life impact compared to mild hypodontia. Other factors associated with higher quality of life impact was people who were pre-treatment compared to mid- or end-treatment, older adolescents, and adolescent respondents compared to parents and dyads (Appendix Table 24).

Figure 5.7: Adolescent and parent-reported impact of hypodontia on adolescent’s oral health-related quality of life



5.4.4 Examination of the joint decision-making processes

Observation of the dyadic interaction for negotiation of joint preferences identified two typical approaches; mutual negotiation, in which there was a shared approach to decision-making, and the other where one respondent was dominant (Table 5.3).

Table 5.3: Characteristics of dyadic approaches to joint preference selection

Mutual negotiation	Dominant respondent
Attributes discussed prior to starting tasks Both participants offer a preferred choice Joint preference is deliberated	Little prior discussion One respondent makes choice (variability in whether this is agreed beforehand) Little deliberation

‘Mutual negotiation’ dyads typically approached tasks by discussing attributes first then looking at tasks. The dyads discussed the importance of attributes then used the tasks as a way to identify attributes that required discussion. The approach to choosing tended to involve both respondents in the dyad first stating which treatment they would pick then discussing their choice to explore shared preferences that determine whether they agree. Dyads discussed and negotiated treatment choice, persuading each other, with choices taking longer where there was greater disagreement requiring negotiation of preferences. In some cases, the child was encouraged to choose first then justify choice to the parent, encouraging greater deliberation. Parents were generally more analytic in approach, while adolescents appeared more intuitive in their response. Interestingly, often dyads selected the same option but for different reasons and while further discussion encouraged greater consideration of all attributes it did not necessarily result in a change in choice. Parents were often also more realistic about the compromise required in choices, as typified by this exchange between a dyad

“Easy, easy! that’s the best I’ve seen!” (Adolescent)

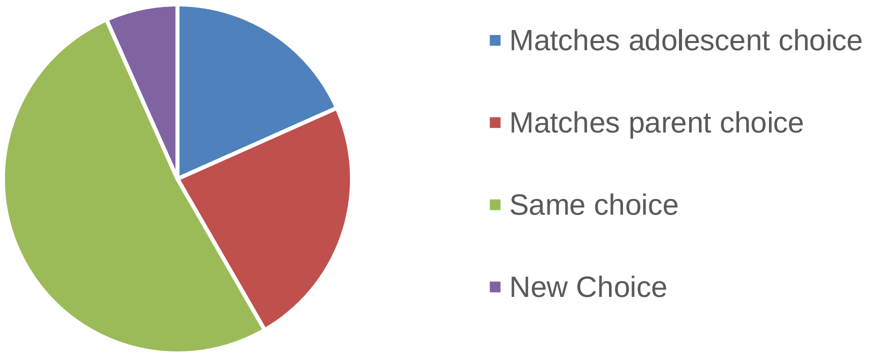
“That’s called a magic wand” (Father, Dyad 2)

Within the negotiation, one person with a strong preference for a specific attribute became an advocate, for example, adolescents were more likely to consider “Bite” while parents were more sensitive to “Problems” and became more persuasive about this particular attribute. Adolescents showed a greater interest in outcomes, while parents were able to consider the risk-benefit balance of treatment. This was reflected in the greater willingness of parents to consider No Treatment. Possibly as a result of the extent of the discussion, dyads tended to agree on the perceived difficulty of tasks.

In the second type of behaviour observed, one respondent tended to be dominant. When approaching tasks and decision-making there was less discussion about attributes beforehand and one respondent took the lead. This approach appeared to correlate the individual's understanding and engagement with the choice tasks and beliefs about who should make decisions about dental treatment. Where one respondent was dominant, decision-making was generally quicker with less discussion around options. The limited discussion made it difficult to gauge how joint decision-making had been approached. These respondents were also more reticent to discuss their experience of the DCE tasks in the follow-up interview.

The choice made in the DCE Choice tasks for individual adolescents, parents and dyads was directly compared. Agreement between the individual and joint choice task responses for the 15 dyads is shown in Figure 5.8. Approximately half the dyads (52%) selected the same alternative when completing the choice tasks individually and together. Where individual choices differed, the joint choice matched the adolescent choice and parent choice in 22 (18%) and 28 (23%) tasks respectively. In 8 (7%) choice tasks, the joint choice represented a new choice for both respondents and did not match either adolescent or parent choices.

Figure 5.8: Agreement between joint adolescent-parent choice and individual choice in choice tasks



Cases who displayed typical behaviour based on the categories described above were selected to test whether choices matched observed behaviour. It was expected that for dyads where one respondent was dominant, the joint preference would reflect those of the dominant individuals, whereas for dyads displaying a more negotiated choice, the joint preferences may reflect individual choices more variably. Examination of typical cases suggests variation in whether actual choices mapped to observed behaviour (Table 5.4). Dyad 1 and 6 showed a high proportion of cases where individual and joint choices were the same, while Dyad 2, 5, 10, 12 and 13 showed a spread in the joint choices. For these cases it is difficult to interpret choices without the additional information gained from the observation. The only case where the joint choice clearly matched the joint choice was Dyad 11.

Table 5.4: Examination of joints choices for typical cases in the observed survey

	Dyad	Joint choice				Survey time (min:sec)
		Reflects adolescent	Reflects parent	Same choice	New choice	
Mutual negotiation	1	0	1	**6**	1	11:49
	2	2	3	3	0	6:38
	10	0	3	3	2	15:48
Dominant respondent (Adolescent)	5	2	3	1	2	3:07
	6	0	0	**7**	1	3:29
	11	**6**	0	2	0	4:46
Dominant respondent (Parent)	12	1	4	3	0	6:18
	13	2	2	4	0	9:12

5.4.5 Perception of the choice tasks

The perception of the usefulness of completing the DCE choice tasks found respondents reacted to the tasks in different ways. The first group of people was those who found it useful and described a change in their approach to thinking about options. This tended to be those who were unsure which treatment to choose prior to completing the survey and wanted to find out more about treatment, and those who were interested in the research method. Approximately half adolescent respondents were not sure whether such a tool was useful, often because they displayed a strong preference for a single attribute, or they felt they were already sure of treatment decision. A minority of respondents did not find it useful and demonstrated difficulty with the requirement to trade-off. The most obvious case like this was Dyad 5, where the adolescent completed the survey quickly with little discussion with the parent and subsequently reported little engagement in the tasks.

Generally, parents were more willing to admit finding the trade-offs challenging at times. One parent in the observed survey commented:

> *"It is like being asked if you would rather be eaten by a lion or a tiger"*
>
> (Father, Dyad 2)

A comment from an online survey respondent commented that completing the tasks emphasised the complexity of decision-making and feeling inadequately prepared:

> *"Completing this survey made me realise how much trust we are putting into the hands of the specialists and how helpless I feel as a parent to advise my daughter whether we are doing the right thing."*
>
> (Mother of 15-year-old female with moderate hypodontia)

Other comments from the online survey related to experience of decision-making, treatment and outcome from treatment (Appendix Table 25). There was evidence of the complexity of dental treatment for hypodontia, a lack of clarity about treatment options and expected outcome, and dissatisfaction with the decision-making process. Satisfaction with outcome was variable, with two respondents emphasising dissatisfaction with the colour of their bridge.

5.4.6 Examination of preference data

In total, 204 respondents completed the online survey and 15 dyads completed the observed survey, providing a total of 234 individual respondents with 7 Choice tasks per respondent, totalling 1,638 observations. The additional repeat task in all surveys was not part of the experimental design and was not included in the main analysis. The repeat online survey and the observed dyads' joint survey were not included in the main analysis, as this data duplicated existing responses.

5.4.6.1 Quality of responses

Time taken to complete the survey by different respondent groups was, on average, approximately 15 minutes. Considerable range was seen in survey completion time (Appendix Figure 12). The online adolescent group showed greatest variability in response time, with some respondents completing the survey in less than 3 minutes. The observed adolescent group spent least time completing the survey. Five outliers in the online survey were identified who took more than 45 minutes. The observed survey respondent who appeared to take over an hour for completion was due to a delay in pressing submit, rather than an extended completion time. There was no significant difference in survey time completion between groups of respondents (p=0.59).

Observation of survey completion suggested a minimum of 6 minutes was required to adequately read the attribute descriptions, consider the choice tasks and answer the remaining survey question. Using a minimum response time of 6 minutes resulted in exclusion of 18 online survey respondents (14 adolescents, 3 parents and 1 dyad) from the main survey analysis. Characteristics of these respondents is given in Appendix Table 26. The effect of removing these respondents from the preference analysis is tested in Section 5.4.9.

Examination of the data to assess task non-attendance identified only two participants who consistently selected Treatment A or Treatment B (Appendix Table 27). The respondents did not display any other behaviour that suggested the survey was not completed correctly; the attribute descriptions were accessed, there was variation in responses to other questions and the survey completion times were 14 and 17 minutes. For this reason, the respondents were not excluded from the analysis, as it was judged the choices could represent the true preferences.

The final sample for analysis included 216 respondents providing a total of 1,512 observations.

5.4.6.2 Model estimation

Three-level time attributes were initially modelled as categorical to assess the functional form (Appendix Figure 13). The attributes were found to be non-linear, hence, all independent variables were coded as categorical to avoid imposing any functional form on preference weights and to provide a validity check on the correct ordering of naturally ordered attribute levels (Bridges et al., 2011).

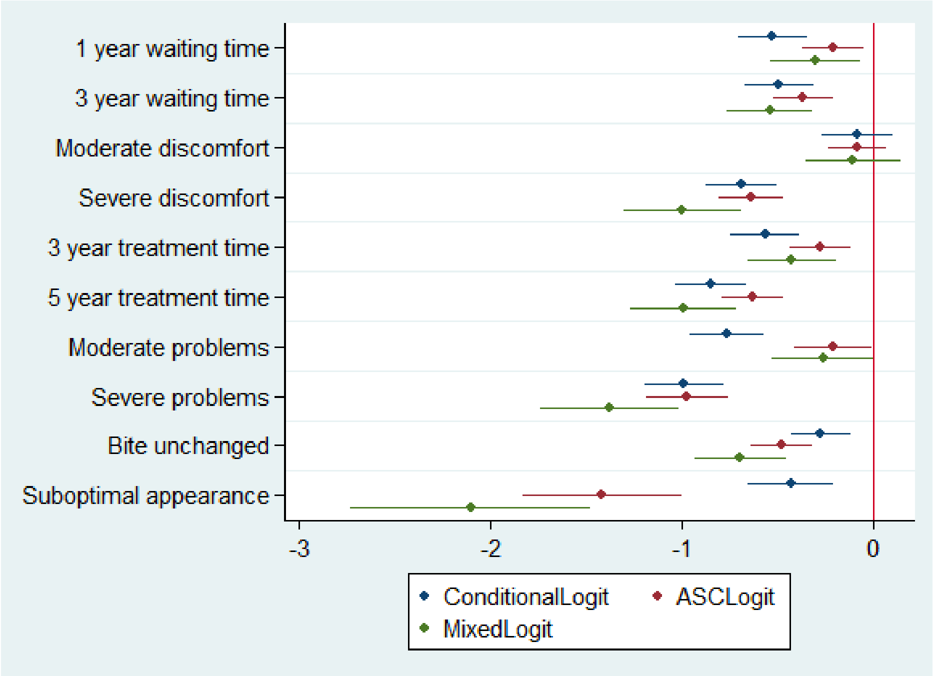
The conditional logit (CL) with main effects, alternative constant specific (ASC) conditional logit and mixed logit model were tested using the log likelihood ratio chi-square, AIC and BIC used to compare goodness of fit (Table 5.5). Mixed logit models were estimated by simulated maximum likelihood using Halton draws with 500 replications and all attributes included as random parameters to account for unobserved variation in respondents' preferences. No constraints were placed on the distribution of the attributes. The model was repeated with and without correlation to test the assumption of correlated normally distributed random coefficients (Chouaid et al., 2019). The mixed logit without correlation provided the best fit to the data.

Table 5.5: Goodness-of-fit for different estimation models

	Conditional logit	ASC Conditional logit	Mixed logit (random effects with correlation)	Mixed logit (random effects without correlation)
AIC	3214.322	1870.823	1695.1	1796
BIC	3274.456	1936.1935	2085.73	1916
Likelihood ratio chi-square test	322.55	194.03	275.39	94.32

The models resulted in considerable differences in preference estimates and different relative preferences between attributes (Appendix Table 28). The mean coefficient estimates and 95% confidence intervals produced by different models are displayed in Figure 5.9. The mixed logit results best reflected the preferences reported by observed survey respondents; that is, that appearance and severe problems are most important. The mixed logit without correlation was selected for estimating preference weights and marginal rates of substitution.

Figure 5.9: Preference weight estimates from different estimation model (mean estimate with 95% CI)



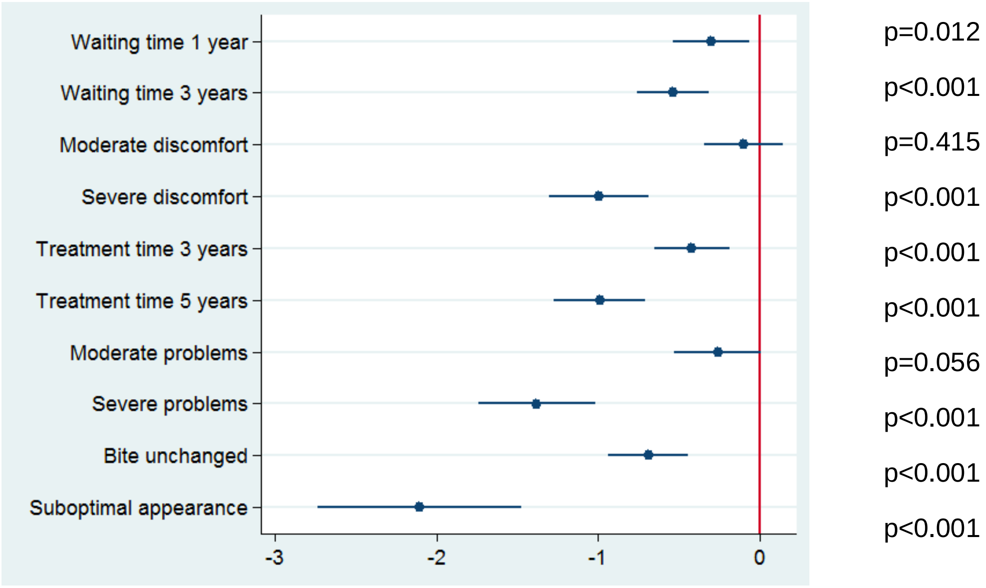
Preference weights for attribute-levels are given relative to the omitted level for that attribute, which has a value of zero (red line). Reference levels are

- Discomfort: none/mild
- Treatment time: 2 months
- Waiting time: 3 months
- Problems: none/mild
- Bite: improved
- Appearance: improved

5.4.7 Preferences for attributes of hypodontia care

Preference estimates for the whole sample (n=1,512) are given in Appendix Table 29 and summarised in Figure 5.10. A statistically significant reduction in utility relative to the reference level was found for all attribute-levels except moderate discomfort and moderate problems. The greatest drivers of preference were suboptimal appearance then severe problems, followed by a 5-year treatment time and severe discomfort. The overlapping confidence intervals for a waiting time of 1 year and 3 years suggests that although increasing waiting time from 3 months to 1 year causes significant disutility, further increasing the length of wait does not have a significant effect. The effect between levels within an attribute was significant for all other attributes.

Figure 5.10: Preference weights estimates reported for attributes of hypodontia care relative to the reference level (mean estimate with 95% CI)



Preference weights for attribute-levels are given relative to the omitted level for that attribute, which has a value of zero (red line). Reference levels are

- Discomfort: none/mild
- Treatment time: 2 months
- Waiting time: 3 months
- Problems: none/mild
- Bite: improved
- Appearance: improved

Significant preference heterogeneity was identified for a number of attribute-levels: Severe discomfort; 5-year treatment time; Severe problems; Unchanged bite; and suboptimal appearance. This preference heterogeneity explains the difference in preference estimates between the conditional logit and mixed logit models. Examination of the distribution curves for the attribute-levels suggested moderate discomfort demonstrated a symmetrical bimodal distribution, while severe discomfort and suboptimal appearance showed an asymmetric bimodal distribution (Appendix Figure 14).

Due to the non-linear form of attributes, ratios between specific attribute levels were calculated rather than marginal rates of substitution (Appendix Table 30). A suboptimal bite was equivalent to 63 months wait, or 167 months total treatment time. An unchanged bite was equivalent to 21 months of waiting, or 54 months total treatment time. Compared to no/mild problems, severe problems was the equivalent to 41 months wait, or 112 months total treatment time. Compared to no/mild discomfort, severe discomfort was equivalent to 30 months of waiting, or 82 months total treatment time. It should be noted the confidence intervals for the ratios were wide due to uncertainty in estimates.

5.4.8 Difference in preferences between respondents

5.4.8.1 Interactions

Interactions were used to test differences in preference based on the specific hypotheses given in Table 5.2. The following findings support the pre-defined hypotheses:

- Parents demonstrated a significantly greater disutility from severe problems, approximately 1.5 times that of adolescent and dyads (p=0.038)
- Older respondents (adolescents aged 15-16 years and parents of older adolescents) have around 1.5 times stronger preference for an improved appearance than younger adolescents (adolescents aged 12-14 years and parents of younger adolescents) (p=0.029)
- Those reporting a high quality of life impact from hypodontia were less concerned about severe discomfort; preference estimate for respondents with severe quality of life impact was 43% that of respondents with low-moderate quality of life impact from hypodontia (p=0.016)
- People in treatment have around a 30% stronger preference for improvement in appearance than those who have not yet started treatment (p=0.011)

The following hypotheses were rejected:

- Adolescents are less tolerant of a long waiting time and more tolerant of severe discomfort than parents
- Younger adolescents are more avoidant of severe discomfort
- People in treatment have a stronger preference for improvement in bite and treatment naïve people are more avoidant of discomfort
- Respondents with severe hypodontia have stronger preference for improved function than those with mild-moderate hypodontia
- Dentally anxious respondents are more willing to accept a long wait for treatment, but more avoidant of severe problems and severe discomfort
- People reporting a high quality of life impact from hypodontia would be more willing to accept severe problems and a long treatment time but less willing to wait for treatment

The full results are given in Appendix Table 31.

5.4.8.2 Latent class

Observation of decision-making in the pilot study and observed study suggested there were three types of decision-makers: those with no pre-determined preference who debated the attributes, those with strong existing preference for one or more attributes, and those who did not engage well in the task. It was possible there would be more latent classes, as there might be differences in what was important to those with strong preferences.

Three, four and five classes were examined to determine which best explained the clinical findings (Appendix Table 32). Four classes was preferred as this captured the group of respondents who did not engage with the task and explained the data consistently with observed behaviour in this stage and the pilot. Five latent classes provided the best model fit (Appendix Table 33) but the groups created were difficult to interpret.

The four classes of respondents were characterised as follows:

Class 1 ‘Non-engagers’ (12%): The magnitude and erratic nature of preference estimates suggest the data is poor quality. This class is likely to represent the respondents who did not understand or engage in the task.

Class 2 ‘Appearance-focussed’ (34%): Disutility for suboptimal appearance is four times that of any other attribute, suggesting appearance is strongly dominant and there is little consideration of other attributes. Clinically this might represent those who express a desire to improve their dental appearance, regardless of what this might involve.

Class 3 ‘Bite-focussed’ (19%): Respondents show a strong disutility for unchanged bite and long treatment time and are willing to trade waiting time. Appearance and severe discomfort have a moderate effect. Clinically this may represent those with functional issues who are keen to improve bite but are willing to wait for a shorter treatment. The utility gain severe problems is difficult to interpret, but it is possible the attribute was not well understood or it was genuinely considered unimportant.

Class 4 ‘Risk avoidant’ (34%): Respondents have the strongest preference for avoidance of severe problems and discomfort. Outcome attributes (appearance and bite) are considerably less important. Clinically this class may represent those who perceive gains in treatment to be moderate and are not willing to undergo ‘high risk’ treatment.

Membership to the classes (Appendix Table 34) suggested relative to the reference class (Class 4):

Class 1 is more likely to be adolescents than dyads, and people in treatment compared to those who have not yet started treatment. The greatest predictor for being in this class was online rather than observed survey respondents. Parent respondents, older adolescents and parents of older adolescent, those with milder hypodontia, anxious respondents and those with a high quality of life impact from hypodontia are less likely to be in this group.

Class 2 is more likely to be parents and adolescents than dyads, people in treatment compared to those who are pre-treatment, younger respondents and parents of younger respondents, and those with low quality of life impact from hypodontia.

Class 3 is more likely to be adolescents and less likely to be parents compared to dyads. Those who are not dentally anxious, those with a more severe hypodontia and those with a high quality of life impact from hypodontia are more likely to be in this class.

5.4.9 Choice behaviour

5.4.9.1 Selection of *No Treatment*

The selection of '*No Treatment'* was examined for the online survey respondents (n= 186) and both individual and dyadic responses for the observed survey (n=45) for the seven choice tasks, providing a total of 1,617 observations. The majority of respondents (58%) did not select '*No Treatment'* in any tasks, while 12 respondents (5%) selected No Treatment in all seven choice tasks (Appendix Figure 15).

In total, '*No Treatment'* was selected in 287 (17.7%) tasks. Logistic regression found a number of respondent characteristics were significantly associated with selection of No Treatment (Appendix Table 35). This found that dental anxiety influenced the selection of *'No treatment'*, with people reporting dental anxiety being more than twice as likely to choose no treatment. Parents were 1.7 times as likely to select No Treatment as adolescents, but dyads were 80% less likely to opt out. Online data collection reduced the likelihood of opting out by 34%. Females were 41% less likely than males to select No Treatment. Compared to mild hypodontia, those with moderate and severe hypodontia were 46% and 69% less likely to opt out. The only characteristic that did not have the expected impact on selection of No Treatment was quality of life impact from hypodontia; those with moderate and low quality of life impact from hypodontia were 70% and 28% less likely to opt out than those reporting a high quality of life impact.

The twelve respondents that selected No Treatment in all tasks were considered to be non-consumers. Examination of latent class membership found this group (non-consumers) were more likely to be in Class 1 ‘Non-engagers’. This suggests the opt-out might have been being used by respondents who found the tasks difficult. Those who never selected No Treatment were more likely to be in Class 3 ‘Bite-focussed’ suggesting those with a high motivation for treatment want an improvement in function (Appendix Table 36).

A third alternative was created to mimic no treatment (Zero months waiting time and treatment time; No discomfort and problems during treatment; Appearance and bite unchanged). Those that choose no treatment after the forced choice were assigned to the third alternative. The alternative-specific constant was not significant (-0.02, 95% CI -0.33 to 0.26, p=0.853) suggesting there was no preference for no treatment at a population level.

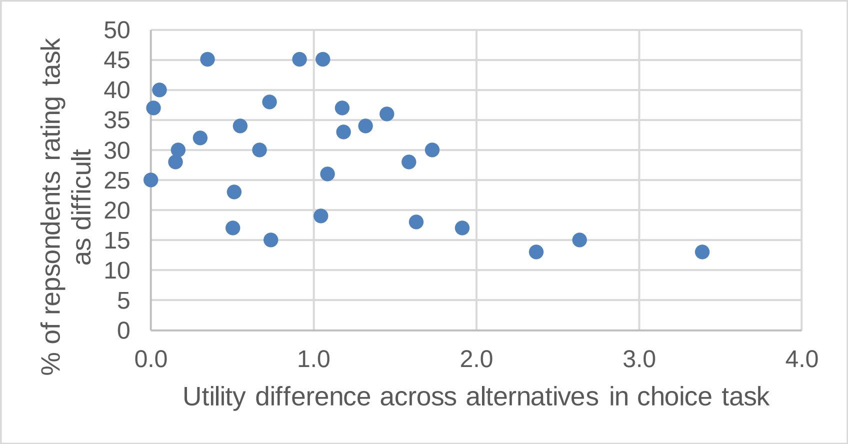
Respondents were categorised based on those who always, sometimes and never opt out and included as an interaction with the least desirable levels for each attribute in the mixed logit. The non-significant results indicates those who always and never opt out do not have significantly different preferences to those who occasionally opt-out (Appendix Table 37).

5.4.10 Choice task difficulty

Of the 1,512 tasks completed, 452 (30%) were rated ‘Difficult’, 743 (49%) as ‘Quite Easy’ and 317 (21%) as ‘Easy’. Logistic regression was used to examine factors that may contribute to a rating of Difficult (Appendix Table 38). Parents were 1.2 times more likely to rate a task as difficult than adolescents and respondents classified as dentally anxious were 1.8 times as likely to rate a task as difficult. Gender, age, severity of hypodontia, quality of life impact of hypodontia and mode of survey administration were non-significant. Respondents who rated 6 or 7 tasks as difficult (n=12) were more likely to be in Class 1 ‘Non-engagers’ suggesting difficulty with the task might have contributed to lower engagement and unclear preferences (Appendix Table 39).

Correlation between utility balance and self-reported task difficulty suggested a significant moderate negative correlation (Pearson correlation -0.505, p=0.006) (Figure 5.11). This indicates tasks with greater utility difference between alternatives were generally perceived to be less difficult than tasks with more equal alternatives.

Figure 5.11: Correlation between objective task difficulty (utility difference in choice tasks) and self-reported task difficulty (Pearson correlation = -0.505)



5.4.11 Evaluation of DCE Choice tasks

5.4.11.1 DCE choice task validity

Preference weights met prior expectations, that is, a longer wait, longer treatment time, greater discomfort, more severe problems and compromised outcomes were less favourable. Appearance resulted in the greatest effect and this supports the pilot results and verbal feedback that this is most important. Severe problems were predominantly highlighted as important by parents in the joint negotiation, but adolescents agreed.

In support of the pilot, there was a general consensus that outcome was more important than process and people were willing to trade-off undesirable process attribute, mostly waiting time and treatment time, for a gain in appearance, and to a lesser extent bite. This is reflected in a comment provided in the feedback box by a survey respondent:

> *"Whilst the treatment process is important the most important factor for me was the outcome at the end"*
>
> (Mother of 15-year-old female with moderate hypodontia).

5.4.11.2 Effect of data collection mode on preferences

The mode of data collection was tested as an interaction in the mixed logit model. The mode of data collection had a significant effect on preferences for severe problems and unchanged bite (Appendix Table 40). Severe problems were significant for the observed group and although still significant for the online survey respondents, utility loss was approximately 38% less. Unchanged bite was not significant for the observed group but significant for the online group, with four times the preference weight. The preference estimates for the remaining attribute-levels did not differ significantly between the two data collection methods.

5.4.11.3 Effect of time taken to complete the survey

A sensitivity analysis was performed to examine the effect of excluding respondents who completing the survey in a time that was judged to be incompatible with valid completion of the tasks. Inclusion of the rapid respondents had minimal effect on preference estimates (Appendix Figure 16).

5.4.11.4 Repeat choice task (test-retest)

The repeat tasks were examined for the online survey, the repeat online survey and individual and joint observed surveys providing a total of 272 observations. Of these, 200 (74%) were in agreement giving an overall ICC of 0.67 (95% CI 0.58 to 0.74) (Table 5.6). Variation was noted by respondent group, with higher agreement for parents and dyads than adolescents, and higher agreement for the observed survey than online. Variation was also found by block and survey completion time.

A strong correlation (0.85) was found between the repeat choice task difficulty and mean intra-examiner agreement (Appendix Figure 17). This suggests increasing difference in utility between alternatives in the task, which indicates choice tasks are easier, is related to higher intra-examiner agreement. Examination of the difference in self-reported difficulty of tasks between initial and repeat completion found the majority of respondents (63%) reported the same perceived difficulty of task, while 59 respondents (22%) found the repeat task easier and 16% found it more difficult. Examination of the change in selection of treatment or No Treatment between initial and repeat completion found the majority of respondents (87%) made the same choice in the initial and repeat task. Nineteen respondents (7%) changed from a treatment to No Treatment, while sixteen respondents (6%) changed from No Treatment to treatment.

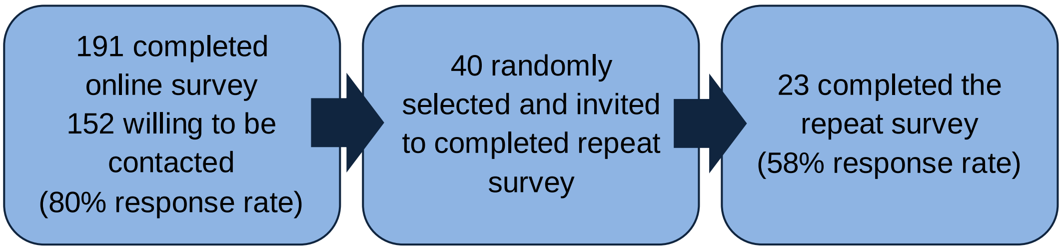
Table 5.6: Intra-respondent agreement in the choice task test-retest

		Number of Observations	Number in agreement	Intra-respondent agreement	
				ICC	95% CI
Online survey	**All**	204	142	0.60	0.48 to 0.70
	Adolescent	122	81	0.50	0.28 to 0.65
	Parent	56	41	0.76	0.58 to 0.86
	Dyad	26	20	0.70	0.34 to 0.87
Online repeat	**All**	23	18	0.71	0.30 to 0.88
Observed survey	**All**	45	40	0.88	0.78 to 0.93
	Adolescent	15	13	0.85	0.57 to 0.95
	Parent	15	14	0.93	0.81 to 0.98
	Dyad	15	13	0.86	0.59 to 0.95
Block	**1**	69	53	0.81	0.70 to 0.88
	2	92	72	0.68	0.51 to 0.79
	3	56	37	0.51	0.18 to 0.71
	4	55	38	0.55	0.23 to 0.74

5.4.11.5 Survey test-retest method

The sampling and response rate for the repeat survey containing the DCE Choice tasks is given in Figure 5.12. Twenty-three respondents completed the repeat survey providing 161 observations (repeat tasks were excluded). The tasks were completed 2-5 weeks after initial completion. Characteristics of the repeat sample are given in Appendix Table 41. Compared to the baseline survey, fewer adolescent respondents and more parents completed the repeat survey; however, differences between the repeat and baseline sample characteristics were not statistically significant.

Figure 5.12: Sampling and response rate for the repeat online survey



The majority of respondents showed agreement in 4-6 out of the seven tasks (Appendix Figure 18). Four respondents made the same choice in fewer than 4 tasks. The total agreement across tasks, measured with the ICC, was 0.46 (95% CI 0.26 to 0.60). The median completion time for the repeat survey was 5 minutes 40 seconds (range 2.5-17 minutes). A weak non-significant correlation (0.17, p=0.453) was found between time to complete tasks and number of tasks in agreement per respondent. This suggests more time spent on tasks does not improve agreement in choices.

Examination of difficulty ratings for tasks found 59% of tasks were given the same rating, 19% were rated as more difficult and 22% as easier. Examination of the change in selection of treatment or no treatment between initial and repeat survey completion found that in 142 tasks (88%) made the same choice. In twelve tasks (7%) respondents changed from a treatment to No Treatment, while in seven (4%) respondents changed from No Treatment to treatment.

5.4.11.6 External validity

Self-reported planned treatment for the online survey respondents (n=186) is summarised in Table 5.7. The responses demonstrate considerable uncertainty about future treatment for all methods except orthodontic treatment.

Table 5.7: Actual planned treatment, as reported by online survey respondents (n=186)

	Treatment planned	Not sure
Restorative camouflage	43 (26%)	64 (34%)
Orthodontic treatment	139 (75%)	16 (9%)
Tooth replacement with bridge	81 (44%)	47 (25%)
Tooth replacement with implant	25 (13%)	77 (41%)

Planned treatment, as reported by the adolescent, parent and clinical notes, for the 15 dyads is given in Appendix Table 42. Agreement was found in 8 of the 15 dyads, although there was evidence that the simple choice options given in the survey to self-report planned treatment were not able to accurately capture the complexity of the actual clinical decision-making, causing an inaccurate perception of respondent uncertainty. For example, in Dyad 3 the parent reports uncertainty about tooth replacement with a resin-bonded bridge (RBB), but further examination of information from the clinical notes confirms RBB treatment is not planned at this time but may be required in the future if the primary teeth are lost. Similar ambiguity is seen in Dyads 4, 8, 9, 10,11, 12, 13; in these cases, initial treatment is planned but, in the future, further decisions may be required based on the outcome of initial treatment and physical changes, such as loss of primary teeth.

Two cases suggested respondents were unsure or misunderstood the proposed treatment. In Dyad 1 the decision had been made that no treatment was desired at this time, yet the adolescent reports planned tooth replacement treatment. In Dyad 13, the clinical notes suggest orthodontic treatment followed by tooth replacement has been agreed, yet both adolescent and parent report uncertainty about all options. Other cases also demonstrate misunderstanding by one or both dyad respondents (Dyad 4 parent, Dyad 6 adolescent, Dyad 11 parent, Dyad 15 parent). Dyad 4 and 6 are undergoing or nearing the end of treatment, so apparent uncertainty about orthodontic treatment may be a reflection that this treatment has already been delivered. This would suggest a problem with the question and response format rather than genuine uncertainty.

At the outset, testing external validity by comparing predicted choice, calculated from stated preference data, to actual treatment choice was planned. This was not possible for two reasons. Firstly, even with the clinical notes it was difficult to determine the choices given to the adolescent and parent during the decision-making process. Secondly, it was difficult to characterise treatment outcomes to specific treatment, because outcome will depend on clinical and technical factors so only process attributes could be assigned to treatment options. Using preference weight estimates, the utility of the process attributes of the four treatment options was calculated (Table 5.8). This suggests implants would need to provide a significantly improved outcome to counter the increased burden of the treatment; the utility loss of implants compared to restorative camouflage only (-3.904) is approximately twice the utility loss of a suboptimal appearance (-2.103), so the appearance outcome from implants would need to be ‘twice as good’. This is challenging to translate into a meaningful clinical value, as appearance is subjective and multifactorial.

Table 5.8: Combined utility of process attributes for treatment options (calculated using preference weight estimates)

	Discomfort during tx	Wait for treatment	Risk during tx	Treatment time	Estimated utility
Restorative camouflage	Low	3m	Low	2 months	0
Orthodontics only	Moderate	12m	Moderate	3 years	-1.019
Orthodontics and RBB	Moderate	12m	Moderate	3 years	-1.019
Orthodontics and implant	High	3 years	High	5 years	-3.904

5.5 Discussion

5.5.1 Understanding of hypodontia & the decision-making context

The preference elicitation survey produced further evidence the adolescent and parent understanding about hypodontia is variable, which was demonstrated in the lack of clarity about the location of missing teeth and previous and planned treatment. Examination of dyadic reporting of previous dental treatment found discordance in a third of cases. This may be a result of recall bias in treatment received, misunderstanding about whether previous treatment was related to hypodontia or not, or a lack of clarity in the question and/or answer options. The observation method also raised a question about whether respondents were unaware of the attributes of their own treatment. Use of a ‘report card’ (Bridges et al., 2011) to capture respondents’ perceived attributes of real-life treatment at the start of the DCE may have been useful to assess this.

Information gained about decision-making beliefs and the environment in which decisions are being made is interesting information for shared decision-making. A small minority (3%) reported that dentists do not help with decision-making, which raises questions about where personalised information about the treatment options for an individual would come from. The role of friends was lower than would be expected from general literature about peer influence, which suggests teenagers are more prone to influence by peers and social norms (Trulsson et al., 2002). The discrepancy might be attributed to the more subtle, and possibly subconscious, effect of social pressure, rather than friends being seen as having an explicit role in decision-making.

Responses from the SURE tool suggested on average, around 70-80% felt confident in the four domains, which reflects findings from a previous study using the SURE tool (Legare et al., 2010). The SURE tool has not been validated as a screening tool for the dental setting so it may not be appropriate for identifying clinically significant decisional conflict without formal validation of the its predictive validity in these contexts (Decary et al., 2017). Despite this, these results highlight that for a notable proportion of adolescent and parents there is a degree of uncertainty around decision-making and the ‘best’ treatment. Validation of the tool to allow its application in routine care would be valuable for determining those where there is a decision support need.

Around 85% of adolescent online survey respondents reported feeling sure of the best choice, yet around 10% fewer reported to know the risks and benefits. This may indicate adolescents do not feel the need to know risks and benefits of treatment to make a choice, particularly if they feel their parent or the dentist are responsible for this. Online parent respondents showed greatest uncertainty generally; this might reflect a greater honesty from those completing the survey remotely, which gives a greater sense of anonymity, or it might reflect that parents choosing to access the online survey are those who have uncertainty and are looking for further decision-making support. Dyadic responses from the online survey were generally an average between adolescent and parent responses, except for the question about adequate advice and support, where 95% respond positively. This might be due to an unwillingness to admit a support need to the other person completing the survey. Similarly, all adolescents in the observed survey reported having enough support and advice, and parents reported high levels of certainty about risks and benefits. This may be due to a reluctance to criticise the care team when under observation.

Generally, some degree of dental anxiety was reported for around a third of adolescents for routine care, and nearly half for hypodontia care. This may reflect uncertainty about the treatment involved in hypodontia care and the effect of an unfamiliar environment and dental team. The developmental nature of hypodontia means it is unaffected by previous good dental health, so those receiving treatment for hypodontia may have had little treatment need previously. Levels of parental anxiety about dental treatment was also notable, at around 40%. Parental anxiety has been found to be a contributor to child anxiety (Seligman et al., 2017) but the effect of anxiety on decision-making for another person is not clear . For the dyads it was found adolescents’ self-reported anxiety was higher than reported by parents for normal dental treatment and hypodontia treatment for 6 (40%) and 4 (27%) respondents respectively. This discrepancy is important for clinicians to recognise, particularly if a parent is the dominant voice in treatment selection, as it may affect treatment acceptance, adherence and experience.

The impact of hypodontia on quality of life reflects previous research, that is, dental appearance and worry in the future causes considerable quality of life effect (Akram et al., 2011, Meaney et al., 2012, Hvaring et al., 2014a). Interestingly, severe hypodontia was found to significantly increase the likelihood of having a high quality of life impact, where previous research has been equivocal. Functional issues were identified for approximately half the respondents but of these, less than half reported ‘a lot’ of effect day to day. This contrasts with other research where functional issues were found to be higher (Locker et al., 2010, Wong et al., 2006b).

5.5.2 Decision-making behaviour

Observation of adolescent-parent dyads resulted in formulation of a typologies that describe two types of approaches to joint decision-making. Mutual negotiation appeared to be a more analytic approach, characterised by reasoned choice requiring a conscious engagement with the attributes and choice tasks. On the other hand, when joint choices were dictated by one dominant respondent decision-making appeared to be less reasoned and more heuristic or in some cases, more random. This suggests that dyads displaying mutual negotiation behaviour are more consistent with a prescriptive model of ‘good thinking’, hence these dyads should make better choices (Bekker, 2009). In reality, this is was difficult to test within this study design as there was no correct answer against which to judge the decision-making. It was evident that dyads displaying mutual negotiation were more interested in the task and the dominant responder behaviour may, in some cases, have arisen from difficulties with the task.

Examination of typical cases found the selection of joint tasks compared to individual tasks did not always reflect what was observed; however, this may be because in the majority of cases the individual choice by the adolescent and parent and the joint choice were all the same, but importantly, the underlying rationale was different. The selection of a new choice by dyads could be interpreted as a shared choice by the dyad following discussion. In the mutual negotiation cases this appeared to be the case; however, in Dyad 5, the selection of two completely different choices in the joint completion reflected a random approach to task selection arising from low engagement. These examples highlight the added value gained from observing the discussion around choices rather than relying on data in isolation, which may have led to inaccurate assumptions about choice behaviour (Ryan et al., 2009).

The preliminary theory developed about types of joint decision-making, if valid in the clinical context, has implications for clinical practice that warrant further investigation. Those who display traits of mutual negotiation may benefit from decision support that provide a framework for reasoned deliberation, such as tasks requiring a trade-off between attributes like the DCE choice tasks. This is supported by the feedback gained from the follow-up interview, which loosely correlated to the observed experience task completion; dyads who deliberated more reported that completion of the tasks was useful and helped them think about treatment options in a different way.

In contrast, the dominance of one party in clinical decision-making may require further investigation to understand the cause of limited discussion between decision-makers. If little discussion of treatment options is required because there is already good understanding of values and preferences, then there may be little to be gained from additional decision support; however, this may raise concerns clinically if the focus on one aspect of treatment, often outcome, prevents willingness to consider other attributes, such as treatment risks. If limited discussion is a result of one person not understanding the information or not having the ability to balance different options, further alternative approaches to information provision and decision support are required. This was demonstrated by one dyad where the parent visibly struggled with the choice tasks, resulting in the child taking control of the joint task selection without any contribution from the parent. The choice tasks, rather than promoting a reasoned approach were causing vacillation, and in one case a degree of frustration. In such cases, a decision aid employing similar trade-off tasks may be detrimental. It is possible that the five online survey outliers, who took more than 45 minutes to complete the survey, similarly struggled with the tasks.

The observed study also provided further information to examine individual decision-making. The pilot suggested that respondents were divided into those who weighed up individual attributes in each alternative to reach a decision, those who tended towards selection based mostly on one or two attributes and those who found the task difficult and picked without a clear reasoning process. This grouping appeared valid in the observed study. For dyads where attributes were discussed, the parent often took a supportive role by helping the adolescent with interpretation of information, wider consideration of attributes and verbalisation of reasoning, and encouraging their child to acknowledge where decision-making was difficult. Parents tended to be more reasoned, while the adolescents were more heuristic or instinctive; however, when challenged the adolescents were able to justify their choices to explain seemingly irrational choices.

Cognitive processes and the ability to make ‘good’ decisions is pertinent in discussions about the ability of adolescents to provide informed consent in healthcare. Legal limitations on the autonomy of adolescents is based on assumptions of adolescent immaturity, attributed to cognitive and psychosocial differences compared to adults. Informed consent requires knowledge and competence but this overlooks the importance of non-cognitive psychosocial variable, namely judgment maturity, which requires responsibility, temperance and perspective (Steinberg and Cauffman, 1996). There is a tendency to assume adolescents are not rational decision-makers, but the present research suggests providing an opportunity for explanation of behaviour may provide useful insight into underlying reasoning and judgement maturity.

The relative proportion of different groups of respondents in the online survey (60% adolescents, 27% parents and 13% dyads) may reflect that parents are keen to give autonomy to their child. This contrasts with observations from the clinical consultations (Section 3.5), where clinicians predominantly talked to the parents and the adolescent made little contribution, but supports the interviews (Section 3.6), where parents often expressed a desire to let their child choose. The tendency by clinicians to focus on parental involvement may be driven by informed consent, which is more familiar to clinicians than shared decision-making and tends to emphasise the medico-legal requirements of decision-making. The observed study also suggested a tendency for parents to see their role reduce with adolescent transition into independence.

5.5.2.1 Selection of ‘*No Treatment’*

Examination of characteristics and behaviours of respondents selecting no treatment is important clinically, as it may help to identify certain groups who would not seek treatment or may not fully adhere to a treatment that they do not judge to be worthwhile. The majority of respondents did not opt for no treatment in any of the tasks, which suggests a strong motivation for treatment; however, in some cases this seemed to come with a disregard for the risk-benefit analysis. The low selection of no treatment might be due to the sampling method, which recruited people who had sought care, or the hypothetical nature of the method and alternatives. A minority (5%) selected no treatment in all tasks, which could be attributed to little desire for treatment or inaccurate characterising of their own treatment making all the options seemed unattractive. Those selecting no treatment in all tasks were more likely to be in the class of respondents classified as ‘Non-engagers’, which suggests the opt-out was used due to poor understanding of the task or opt-out, or preference for the consequence of inaction rather than the wrong action (Campbell and Erdem, 2019).

Selection of no treatment was associated with increased dental anxiety and parent respondents. This is concordant with expectations, that is, anxious people will require greater treatment benefit to make treatment worthwhile, and parents are better able to process risk. For clinical decision-making, it is important clinicians consider whether a reluctance to undergo treatment is associated with underlying dental anxiety. Examination of task difficulty ratings found parents and anxious respondents were more likely to rate tasks as difficult, and it would seem logical that if these respondents are considering the risk-benefit of alternatives more closely, rather than selecting based on a single attribute, this would increase perceived task difficulty.

Females were less likely to opt out, suggesting females are more motivated towards treatment. Respondents with more severe hypodontia were less likely to pick an opt-out. This might be attributed to the association found between severe hypodontia and a greater impact on quality of life in this sample; however, this is not supported by other literature (Locker et al., 2010, Kotecha et al., 2013, Anweigi et al., 2013a). Alternatively, it may be attributed to severe hypodontia being associated with an earlier diagnosis resulting in unintentional priming of the patient towards uptake of treatment. Respondents who never chose the opt-out were more likely to be in the ‘Bite-focused’ group, suggesting a desire for functional improvement increased treatment uptake. Observation of dyads supported the preference findings; parents were more willing to consider no treatment and took a role in helping adolescents consider the risk-benefit balance of the alternatives versus no treatment.

5.5.3 Preferences for hypodontia care

Analysis of preference data found appearance was highly important, causing a disutility of approximately twice that of the next most important attribute, severe problems. The wide confidence interval for appearance reflects uncertainty in the estimate as a result of the small sample and heterogeneity in preferences. The bimodal distribution curve for suboptimal appearance suggests there are two groups of respondents; those with a moderate preference to avoid a compromised appearance, and those with a much stronger preference. This is supported by the latent class analysis, which found one class of respondents labelled as ‘appearance-focussed’, where appearance was highly dominant, but in other classes appearance had only a moderate effect. The findings also reflect initial preference estimates from the pilot, where appearance was found to be the most important attribute.

The importance of dental appearance is a common finding throughout this research and more widely in hypodontia literature. Dental appearance is recognised as a major contributor to oral health-related quality of life and psychosocial wellbeing (Appendix Table 1). The hypothesis that older adolescents and those undergoing treatment have a stronger preference for improved appearance was supported. The reason for this is may be increasing awareness of dental appearance with age or increasing expectations as treatment progresses (Meaney et al., 2012).

For clinical decision-making it is critical that appearance is considered a key outcome for patients and not disregarded as a primarily ‘cosmetic’ concern that is outside the scope of NHS treatment. The value placed on dental appearance is important in decision-making for two reasons; firstly, to allow adequate information provision about options and what they are expected to deliver, and secondly, to establish and manage expectations. Currently the methods for accurately forecasting appearance after treatment are limited in the NHS, but in other healthcare systems such as the USA there is greater emphasis placed on digitally-manipulated images to support decision-making. With advances in 3D technology, such as is used for orthognathic treatment planning, there is scope for greater use of simulation technology (Motohashi and Kuroda, 1999).

The second most important attribute was severe problems, which was significantly more important than mild and moderate problems. Severe problems had a narrower confidence interval and a more normal distribution curve, suggesting less heterogeneity between respondents. The observed study suggested it was often parents who helped adolescents recognise the longer-term implications of problems, and this was supported by the 1.5 times increase in disutility seen for severe problems for parents compared to adolescents; however, in some cases, parents reduced the perceived risk, for example stating:

> *‘That is not likely to happen….. You only ‘might have’ problems”* (Mother, Dyad 9)

The difference in reaction to the problem attribute may be ascribed to risk tolerance, previous experience of dental treatment and problems, or parents’ perceived role in helping their child with decision-making. For example, it was hypothesised that parents of children who are ‘worriers’ may wish to reassure, while those who feel the child is not taking risk seriously may emphasise the risk. The different reactions to describing risk is an area that has been highlighted for further research in previous chapters.

Similar preference estimates were found for severe discomfort, a long (5-year) treatment time and unchanged bite. Although the confidence intervals were generally narrower, for severe discomfort there was an asymmetric bimodal distribution. The two groups may represent a larger group of people who are willing to accept discomfort for other perceived gains, and a smaller group who are much more avoidant of discomfort. This is supported by the latent class analysis, which identified a group of around a third of respondents who have a strong preference to avoid discomfort, then a slightly smaller group where discomfort has a moderate effect. The preference estimates for discomfort also reflect findings from the observed survey, where there was often a willingness to accept discomfort to avoid problems or suboptimal appearance.

"I don't care as long as it looks OK" (Adolescent, Dyad 11)

Adolescents were generally accepting of some discomfort and parents realised their child was more able to cope than they initially realised, as highlighted in comments from respondents:

"I didn't realise she's such a tough little thing" (Parent, Dyad 4)

"It's all painful anyway" (Adolescent, Dyad 9)

It was expected a priori that a long wait would be important, as this is commonly reported as a concern both in research (Akram et al., 2011) and clinically. It is interesting that a long treatment and severe discomfort were valued comparably, as both represent an impact on everyday quality of life during the treatment period. The majority of respondents were undergoing treatment at the time of answering the survey and the effect of this on preference for timing is unclear, but it is possible for some respondents it may seem less important because they have already experienced and coped with treatment, and for others it may be very important, if they are finding treatment challenging and are keen to finish. Some observed respondents added waiting time and treatment time together to give a cumulative time and made trade-offs directly between the two attributes. This is relevant clinically, as in some cases delaying the start of treatment may reduce the overall treatment time minimising intervals between treatment stages, so explaining this as a trade-off may help acceptance.

The second outcome attribute, bite, appeared to be much less important than appearance; however, the latent class analysis identified a group of approximately 20% of respondents who prioritised bite over all other attributes. Adolescents and those with a high quality of life impact from hypodontia were more likely to be in this group. This supports both the pilot study and feedback from the observed study, where parents did not feel bite was a key concern pre-treatment but some adolescents reported it was important in the choice tasks. It was expected that those with more severe forms of hypodontia would place more value on bite due to functional issues, but this was not supported in this sample. Orthodontists often talk about bite as a simplified explanation of ideal occlusion and include improving inter-arch relationship as a treatment goal to gain an aesthetic and stable outcome, rather than for function per se. A parent in the pilot suggested that the importance of bite becomes 'learned' during orthodontic treatment because it is discussed extensively by the dental team. This, and the subjective nature of the bite, may explain the difference in preferences for bite between adolescents and parents.

For both problems and discomfort, the moderate level had little effect relative to the reference levels of little or none. A waiting time of 3 years caused less disutility than was expected. Like extended treatment times, waiting time is a common concern for patients clinically. Apparent willingness to trade-off waiting time for other attributes was also found in the pilot. Willingness to wait might be due to acceptance of waiting as a result of their own treatment experience, and growing awareness of NHS pressures and waiting lists reported in mainstream media. It was hypothesised that dentally anxious respondents would be more willing to wait for treatment while those with a high quality of life impact from hypodontia would be less willing to wait. Examination of the preference data did not support either hypothesis.

Heterogeneity in the preferences in this study was marked and this is important for information provision and decision-making in practice. The current approach to consultation and decision-making is standardised, with little scope for tailoring information and no attempts to elicit preferences from adolescents and parents. The assumption that the important components of treatment are the same for everyone is a potential barrier to value-based decision-making. This research suggests there are three types of patients alongside the group who did not understand or engage in the task. The first large group is those who prioritise appearance, possibly without giving much consideration to other important treatment issues, such as the risk and side-effects of treatment. This was more likely to be individuals than dyads, which may suggest joint completion of the survey encouraged greater discussion of other attributes and reduced the tendency to consider only appearance. The second large group is those who prioritise avoidance of severe problems and discomfort over outcome attributes. This is less likely to be adolescents and observed survey respondents, supporting the previous suggestion that parents are more sensitive to side-effects. The smaller group was those who prioritised bite, which appeared to be those adolescents and those with more severe hypodontia and a high quality of life impact from hypodontia.

5.5.4 Validity of DCE

Overall, the survey findings fitted with expectations and data could be interpreted relatively easily with observations from the study and wider clinical experience. Although not all hypotheses about differences in preferences were supported, a number were found to be significant and in addition, latent class model was found to explain decision-making behaviour well. The overall effect of respondent characteristics aligns with clinical observations. This supports the overall view that despite some preliminary reservations about the use of a relatively complex DCE with adolescents, the approach worked well and produced good quality data to support recommendations for decision-making in practice.

Prior expectations about the effect of the attribute-levels on preferences were met and the relative preference for attributes supported feedback from the observed survey and the pilot. A significant moderate correlation was found between the utility balance between alternatives in tasks and self-reported task difficulty. This meets expectations that more equal alternatives will be harder to choose between. Clinically, these findings suggest that when the risks and benefits of options are more balanced, the decision is likely to be more difficult and patients are likely to require greater decision support and deliberation.

The effect of face-to-face versus online data collection was examined to explore the effect on preferences. Observed survey respondents showed significantly greater disutility for severe problems, which may indicate this method encouraged more thorough reading and provided an opportunity to clarify the meaning of more complex attributes. This is supported by experience in both the pilot and observed survey, where respondents asked for clarification about problems more than any other attribute. Online respondents showed a significant disutility for unchanged bite, whereas this was not significant for the observed group. The reason for this discrepancy is less clear, but may be due to a greater proportion of adolescents to parents in the online survey causing a skew in preferences. The effect of online administration is of interest clinically, as developing a decision support tool to use remotely would potentially reduce clinical time and allow preparation for decision-making in a familiar environment; however, if remote completion does not promote engagement and validate preference elicitation the benefit would be lost. While online methods provided a larger and potentially more diverse sample, online respondents were more likely to be in the ‘non-engaged’ class of respondents. There appears to be a balance to be made between the number and quality of responses, and for future studies face-to-face methods may be preferable if there are likely to be a high number of respondents who may not initially understand or engage with the task.

Moderate agreement was found in the repeat choice tasks, with the same alternative being selected in around three quarters of tasks. Agreement was higher for parents, then dyads, then adolescents, suggesting parental presence may promote a more consistent approach to choice. The observed survey agreement was higher than online. This may be interpreted as observed survey respondents were more engaged in the tasks and making more rational choices, or alternatively, the observed survey respondents were less willing to select a different option even if their preference changed over the course of the survey due to the researcher being present. The latter explanation seems less likely as the researcher was not able to see the choices being made by individuals.

An important finding from the repeat choice task was the strong correlation between ease of task and intra-examiner agreement. Experimental design of DCEs aims to maximise the statistical power by making choices as similar as possible, but if this results in greater variability in data the benefit is negated. A reasonable proportion of respondents (19%) found the repeat task more difficult than the initial completion. This might be the respondents who struggled to understand the tasks and those who fluctuated throughout the tasks and did progress to identification of their preferences.

Assessment of external validity for online survey respondents was complicated by respondents’ uncertainty about treatment choice. A quarter were unsure if restorative camouflage was planned, a third were unsure about tooth replacement with bridges, and 40% were unsure about future implant treatment. Only 75% reported planned orthodontic treatment, yet the majority of recruitment occurred in orthodontic departments and in most cases, management of hypodontia does involve orthodontic treatment. Uncertainty might arise from ambiguity in the question and answer format used but it might also reflect the complexity of hypodontia decision-making and the need for ongoing re-evaluation as the clinical features and treatment options change.

For the observed respondents it was possible to extract clinical information from the patient notes, yet there were still barriers to assessing external validity. The full list of possible treatment options were recorded, but often only the one or two judged ‘best’ by the clinical team were presented to the patient. It was difficult to judge the outcome of treatment without extensive clinical information, for example, post-treatment appearance will depend on numerous factors such as tooth morphology and colour, gingival architecture, operator skill (Savarrio and McIntyre, 2005).

These challenges prevent prediction of treatment choice from the stated preference data, but it is possible to broadly consider the inferences from the DCE on treatment choice. The stated preference method predicts appearance will drive treatment choice for around a third of respondents, avoidance of discomfort and problems will drive another third, and around a fifth will select based on bite at the end of treatment. Restorative camouflage is a low risk treatment that primarily enhances appearance with little functional change. This would suggest uptake would be high in the appearance-focused and risk-avoidant groups. The number reporting this treatment is lower than might be expected, but this is likely to be due to the treatment being unsuitable. Implant treatment carries the highest risk in terms of severity of problems and discomfort during treatment, but it is often presented as delivering a superior outcome. This would suggest uptake in the appearance-focused and function-focused groups, but again the number selecting this treatment option is lower than expected. This is likely to be due to clinical factors contraindicating this treatment, uncertainty about the suitability of this option at this time because further patient maturation is required and limited funding for implant treatment.

5.5.6 Strengths and limitations

5.5.6.1 Sample

Examination of the sample characteristics suggests some limitations for generalisability. The sample was predominantly White despite attempts to sample from different ethnic populations, and it is possible that other communities may be different in their general health beliefs and expectations, understanding and ability to navigate healthcare systems and social structures, which may influence the impact of treatment. The severity of hypodontia was skewed towards the more severe end of the spectrum, and the majority of respondents were in treatment. This is likely to be a reflection on the recruitment from secondary care, and it is important to recognise decision-making and preferences might differ for people with hypodontia receiving care in other settings. Similarly, the results do not represent those who have not accessed care, most importantly, those who may be highly anxious or have additional barriers to dental access.

It is likely that there was bias in those who chose to participate in the study. As is often found with research, it is possible that respondents tended to be those with an interest in research, or who had a particular desire to share positive or negative experience. The research information alluded to the purpose of the study being to understand decision-making, and this might have appealed to people looking for extra decision-making support. It is challenging to predict how selection bias will have altered the research findings, as apart from the specific hypotheses about difference in preferences there were no expectations about what the research would find. It is possible that more diversity in the sample would lead to more ‘types’ of respondents, for example in approach to joint decision-making or in the latent class analysis of preferences.

To reduce the burden associated with recruitment and optimise the number of people approached, clinicians were not asked to record details of recruitment. This limited the scope for ascertaining response rate, which is important for establishing whether the method was acceptable and for estimating the generalisability of the sample. For the observed survey, approximately half the people approached who expressed initial interest did not wish to participate, often due to the time commitment or the young respondent being unwilling to participate. This might reflect the high burden of hypodontia care, the sensitive nature of the condition, or a perception that the research was either asking for opinions or testing respondents. On the other hand, the majority who participated were engaged and interested in both the research and the opportunity to talk about their experiences to improve care, with a number offering to help with future research.

5.5.6.2 Observational method

The observed survey was an exploratory study with relatively small numbers and limited representation. All adolescent respondents were 14-16 years old, so observations about parent-child relationships in decision-making may not pertain to younger children. Further work is warranted to test whether the findings are consistent in other contexts; however, the findings provided valuable insight into decision-making that would not have come from preferences alone. The approach aligns with a descriptive theory of decision-making, that is, by examining cognitive processes it is possible to describe a mechanism for decision-making (Bekker, 2009).

The main limitation was in assessing the cognitive processes of dyads who did not discuss choices as extensively. For example, some dyads pointed at the screen rather than verbalising which attribute they were discussing. The choice to observe only without prompting was taken to gain a true representation of how deliberation occurred. In hindsight, as the tasks were already artificial, it may have been useful to prompt the participants to verbalise their discussion to gain more contemporaneous feedback, as occasionally participants were unable to recall or articulate what they had been discussing when asked in the post-survey interview. An alternative method would have been to video record respondents as they completed the tasks, as has been used with eye-tracking methods (Vass et al., 2018a). This was outside the scope of this research but would have value in future similar studies.

The impact of having the researcher present during task completion is debatable. On one hand, the opportunity to clarify instructions and ask for further explanation of the attributes and choice tasks is likely to increase the validity of results. On the other hand, it is possible that respondents would alter their behaviour to meet perceived expectations, for example, discussing the options more than they naturally would, or maintaining choices even if preferences have changed over the course of the choice tasks. The latter concern was reduced by the voluntary admission by independent respondents (two adolescents and two parents) that they had misunderstood the attributes or tasks initially and as a result, their later preferences or the joint preferences were more reflective of their true preferences.

5.5.6.3 Preference elicitation method

The number of participants for DCE surveys varies considerably (Clark et al., 2014, de Bekker-Grob et al., 2012, Soekhai et al., 2019), but given the limited population the sample size was acceptable. Guidance for identifying the sample size for DCE is limited, although a more robust method to calculate sample size has been recommended which uses the significance level, statistical power level, the intended model, prior estimates of the parameter values and the DCE design (de Bekker-Grob et al., 2015). This method was considered for this research but as recruitment was restricted by the limited population it was not judged to be necessary.

The absence of abandoned online surveys suggests either the survey was acceptable or alternatively, that the £5 incentive was sufficient to encourage people to complete the survey. Approximately 9% of the online survey respondents (18/204) were excluded for completing the survey in a time that was judged to be incompatible with valid survey completion. This is likely to represent those who did not engage in the survey but wanted the £5 voucher. Unsurprisingly, the majority were adolescents, but three reported to be parents and one reported to be a mother-daughter dyad. It is possible these were adolescents who were answering the survey again to get another voucher and provided different details to avoid detection.

Examination of preference data using the latent class identified a second group of poor-quality respondents, who appeared to be unable or unwilling to engage in the task. This represented around 12% of respondents. A proportion of poor-quality responses might be expected with DCE but if this approaches 20% there is potential to impact on sample size. Reassuringly, removal of rapid respondents did not significantly alter preference estimates so it is expected that if others who did not completely engage in the task were included in the analysis, this is unlikely to have significantly altered the results.

The survey completion time was on average 15 minutes, which is acceptable for a DCE survey but also shorter than expected after piloting. In the pilot the average time was nearer to 30 minutes, but this might have been due to interruptions caused by verbal probing. The lack of significant difference in survey completion time across groups suggests those completing the online survey spent an adequate time considering options.

This study was predominantly an applied use of DCE and much of the approach was derived from the intended purpose of the research, that is, to obtain evidence about decision-making processes and preferences that could inform future changes to clinical decision-making practice and development of decision support tools. Different models were tested to identify the most suitable for analysis but extensive complex statistics was outside the scope of the research. The difference in results between the mixed logit and conditional logit emphasises the effect of preference heterogeneity on mean coefficients and justified the selection of the mixed logit. The latent class provided additional information about preference heterogeneity to explain the data in a clinically relevant way.

Methods to evaluate the DCE provided additional useful information without significantly adding to the burden or complexity of the tasks. Test-retest methods provided some indication of intra-examiner reliability, but it is accepted that there will be a degree of learning with task completion which may result in a genuine change in choice. It was evident that obtaining real-time feedback from respondents through think aloud feedback or observation of joint decision-making provided the most valuable information to aid interpretation of preference data and assess validity of the DCE. As discussed, external validity presented a number of challenges related to both the real decision-making context and the proposed method for identifying actual treatment choices. The aim of the research was not to predict treatment choice but to gain a better understanding of decision-making behaviour and preferences, so the inability to verify the external validity of the method does not detract from the research findings.

5.5.7 Implications for clinical practice

Understanding of hypodontia and individual's experience of hypodontia and decision-making varies

The survey provides further evidence that understanding of hypodontia, particularly the number and location of missing teeth, and previous and planned treatment for hypodontia may be lower than assumed by the dental team. It may be necessary to recap information at important decision points in treatment to ensure patients and families are able to participate in conversations with the dental team about treatment. Individual's experience of hypodontia is important and varied, but previous research methods (Chapter 3) indicate this information may be overlooked in favour of clinical information. It is important that teams establish the impact of hypodontia, any anxiety about routine or hypodontia-specific dental treatment, important people to include in decision-making, and any uncertainty around the decision. This may help build rapport and foster a patient-centred approach, as well as providing important information for decision-making.

There is variation in adolescent and parent approaches to individual and joint decision-making

Adolescent-parent approaches to information assimilation and joint decision-making varies. Parents have a valuable role in interpreting and rationalising information, but equally, it is apparent that in many case adolescents have valid reasoning underpinning decisions. Different approaches may be required in to support clinical decision-making, for example, helping those focussed on single outcome to consider risks of treatment, and identifying those who are seeking more information and who might require additional support to think about options. Completing trade-off tasks was helpful for some respondents but others showed indecision that was not improved by this type of exercise. It appears that some adolescent-parent dyads work collaboratively to reach consensus discussing their own understanding of information and reasons for their choices; however, other dyads may require support to ensure all relevant parties have adequate information and can contribute to the decision process. The findings from the pilot were supported in that there was further evidence that some people benefitted from the choice tasks, while others found the trade-offs challenging and unhelpful. This reinforces that there is not one single approach for all patients and a degree of insight and flexibility is required.

Appearance at the end of treatment, severe problems during treatment and high levels of quality of life impact are important but there is significant variation in preferences across individuals

For the sample as a whole, appearance was found to be a very important driver of treatment choice. This has implications for discussing treatment options and providing a realistic prediction of the outcomes that are achievable with different methods. Currently, patient-centred information about dental appearance is lacking and this requires development. Severe problems were the next most important factor in decision making. Parents appeared more able to understand the risks and implications, suggesting they may have a supporting role in presenting this information and ensuring understanding. A long treatment time, severe discomfort during treatment and improvement in bite appeared to have a similar level of importance, but it is likely individuals will assign different values to each.

A significant amount of heterogeneity was identified in the preferences of this sample. The respondents could be broadly grouped into those who prioritise appearance, those who prioritise improvement in bite, and those who wish to avoid severe side-effects associated with treatment. This grouping requires validation, but it is apparent that clinicians need to be aware of the extent and importance of variation in preferences across individuals. It is also important to recognise there may be differences in preference between adolescents and parents, those with severe hypodontia, greater treatment experience, a high impact from hypodontia and dental anxiety. Different preferences need to be elicited and incorporated into decisions, particularly those where treatment choice is primarily a preference-based decision.

5.5.8 Implications for future research

Research to further examine decision-making processes and preferences improve decision-making practice:

- The exploratory study using DCE to examine decision-making has allowed a theory about types of dyadic decision-making to be developed. Future research to test the proposed model with a more diverse sample in different clinical environments would provide useful information about the processes in adolescent-parent decision-making.
- Information about individual and joint decision-making gained from this study can be used to inform development and testing of decision support tools. A robust approach to development and testing is required and it might be necessary to make different tools to support different needs.
- Respondent characteristics appear to influence decision-making and preferences. Further work to map characteristics to decision-making behaviour and preferences is required to support clinical tools (e.g. screening tools) and changes in practice that will promote personalised and patient-centred decision-making.

Research to expand preference elicitation methods for adolescents and dyads:

- DCE has not been used extensively with adolescents but the results from this study are promising. Further DCE studies with adolescents are needed but methods to evaluate the DCE should be incorporated. In particular, the suitability of an online data collection method requires further testing, alongside other design choices outlined in Chapter 4.

Chapter Six:

Discussion and Conclusions

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The final thesis chapter synthesises the key findings from the different stages of the research. The research findings are used to provide recommendations to improve clinical care for adolescents with hypodontia through improved communication and decision-making. The broad implications for health service research are discussed alongside recommendations for future research arising from this work. A critique of the research approach is provided with reflection on the research experience. The chapter closes with the final thesis conclusions.

6.1 Decision-making and preferences in hypodontia

This research has provided valuable information about adolescent and parent decision-making and preferences in hypodontia. While a number of recommendations have arisen from the research to improve clinical practice it is important to emphasise the general positivity from adolescents and parents about their dental care, and the commitment and enthusiasm of the dental teams providing care. Improving the quality of care requires examination and critique of current practice, but with recognition that the findings are not a criticism of any individual or service. Many of the issues identified are a result of the training and care delivery structures and change will require a fundamental shift in the approach to thinking about and delivering care.

The first key finding from the research is that hypodontia and its treatment is complicated, and this presents challenges for adolescents, parents and the dental team in engaging in shared decision-making. Adolescents and parents demonstrate uncertainty about hypodontia and its treatment, and current communication strategies do not appear to be adequately improving knowledge. It is important that the complexity of hypodontia is recognised by dental professionals to allow the knowledge gap created by professionalisation to be addressed and meaningful discussion about treatment to happen (Legare et al., 2011, Street et al., 2005). Fostering greater awareness of the complexity of decision-making and the fundamental role of patients in decision-making is suggested as a means to empower adolescents and parents to ask questions and be involved (Shepherd et al., 2011, Elwyn et al., 2017). Presenting the options as comparable options, defined by their core characteristics, appeared to help some adolescents and parents who were seeking further information to think about treatment in a different way, and provided a language and framework around which treatment options could be discussed (Elwyn et al., 2013b).

The second key finding is that information used to prepare and support adolescents and parents in decision-making is inadequate. It is apparent there is a lack of high-quality research about hypodontia treatment methods and different approaches to care delivery, which presents challenges for evidence-based practice. Communicating uncertainty, particularly around risks and possible immediate and long-term outcomes, may contribute to the sense of indecision around treatment described by some research participants. It is possible that explaining the lack of strong evidence to support the selection of one treatment over another would in itself promote the required shift to preference-based care. Even if healthcare practitioners are unable to reduce uncertainty inherent in difficult care decisions, they can target other subdomains of the decisional conflict construct by providing knowledge, offering support, and eliciting patient values in an attempt to reduce decisional conflict among surrogates (Pecanac et al., 2018).

The third finding is that hypodontia care decisions require a collaborative approach, but currently this is not happening. SDM requires explicit statement of choice, an invitation to participate, appropriate information exchange about important issues and integration of patient and family preferences. Observation of consultations found choice was not explicitly stated and patients were not able to engage. This might be in part due to barriers in understanding the information about treatment options and a lack of awareness about their role in decision-making. Social media users and survey respondents alluded to dissatisfaction with aspects of the treatment process and outcome, but also with the decision-making process itself. To date the patient perspective has been lacking in much of the literature around hypodontia, and the clinical emphasis remains on treatment planning from a clinician's perspective. It is evident from this research that individual's prior experience and circumstances may influence preferences, yet this information is often overlooked in favour of clinical signs and symptoms. Anxiety in particular emerged as potentially having a significant influence on treatment selection, yet clinical experience indicates questions to assess dental anxiety are not part of routine assessment. Similarly, this study and numerous others suggest that the impact of hypodontia on quality of life varies between individuals; however, often adolescents are not often given the opportunity to discuss this adequately.

The SURE tool, though not validated for the dental setting, suggested that around a quarter of survey respondents were uncertain about one or more aspects of decision-making. While this is comparable to other research (Legare et al., 2010), it is important to identify those in need of additional decision support. The use and benefit of decision support tools is an emerging field and to date there is not one known superior approach to shared decision-making (Stacey et al., 2017, Fagerlin et al., 2013). It is probable that the variability in clinical circumstances will prevent agreement on one approach that works, but as with other areas of evidence-based practice, existing knowledge and future research should drive implementation of change in practice. The question of how to more effectively elicit and integrate patient preferences into routine care in hypodontia, and more widely across dentistry, is a research priority. It is likely that this will improve the quality of care as much, if not more, than many technical advances in dental care.

The fourth key finding is that there are strong preferences for particular components of hypodontia care, and understanding these preferences is fundamental for patient-centred decision-making. The preference elicitation survey supported the clinical experience and previous research (Hobkirk et al., 1994), which suggest an improvement in dental appearance is a key motivator for treatment for many people with hypodontia. Parents recognised their child’s drive for a normal appearance and commonly cited that they just wanted their child to be confident to smile. It is conceivable that within the limited resources of NHS treatment, appearance is often dismissed as a cosmetic concern; however, there is strong evidence that appearance is the strongest contributor to oral health-related quality of life (Akram et al., 2011). Appearance is a complex and multifaceted phenomenon, as highlighted by the challenges faced in framing appearance as an attribute and determining appropriate levels. Current methods for predicting treatment outcome are inadequate and discussion with clinicians and patients throughout this research highlights this is a challenge for decision-making.

The risk of severe problems was the second most important attribute, yet clinical observation found the discussion of risk in clinical consultations was minimised and often this information was provided through information leaflets. Although the risks of treatment are routinely discussed within informed consent, the ability to compare risks and side-effects across different treatments should form part of the decision-making process leading up to the selection of a particular treatment and obtaining consent. The quality of life impact of treatment was not discussed extensively, possibly due to professional’s reluctance to cause undue worry. In addition, it seems there may be a substantial proportion of adolescents and parents who are heavily orientated towards treatment and who may choose to minimise the negative aspects of treatment. This was demonstrated by the survey respondents who never selected No Treatment, even where the alternatives conferred little benefit.

Preference elicitation methods in this research suggest treatment outcomes are generally considered more important than the undesirable aspects of treatment delivery, such as a long waiting time. It may be that severe problems were considered both a feature of process and long-term outcome, as some complications have long-term effects. This research did not examine long-term success of treatment and cost because the attribute covering this concept that was developed and tested in the pilot was not judged to be valid. The lack of long-term outcome in the preference elicitation survey is an important consideration, as it means it is not possible to estimate the relative importance of future treatment burden and dental health; however, the process of developing and testing the future cost attribute provided important preliminary information about adolescents' ability to consider the long-term implications of choices and this is an area that requires further examination.

The fifth key finding is that there was considerable heterogeneity in the preferences of this sample. The respondents fitted into four classes: those who prioritise appearance; those who prioritise bite at the end of treatment; those who are keen to minimise severe problems and discomfort; and a fourth group who did not produce any clear preferences, most likely as a failure to understand or engage with the choice tasks. Differences were associated with characteristics including respondent type, age, gender, severity of hypodontia, stage of treatment, dental anxiety and quality of life impact form hypodontia. This demonstrates the need for clinicians to establish the individual's particular background and preferences, rather than using a generic approach to communication and decision-making. Differences were found between adolescent and parent preferences, but joint preference elicitation suggested in many cases adolescents and parents negotiated effectively to explain and justify their choices.

The sixth key finding related to parent's role in decision-making with their child and dyadic approaches to collaborative decision-making. The DCE survey suggested parents predominantly see their role as one of support, to help their child to make a choice that they feel comfortable with; however, there were comments in the survey and in social media posts that indicated parents felt unable to fulfil this role due to their own uncertainty and lack of knowledge. Adolescents, parent and joint approaches to decision-making varied and it is important to consider the process underlying clinical decisions. In some cases, dyadic decision-making was not one of mutual negotiation and one person dominated. Although this does not necessarily equate to poor decision-making, in the clinical context this may give rise to conflict and uncertainty. The effect might be attributed to some degree to the artificial environment and it is recognised the preliminary theory developed about different types of decision-making requires development and testing.

The final key finding was that DCE methods or similar trade-off tasks have potential to be a useful clinical tool. The findings suggest trade-off tasks might help adolescents and parents i) think about and discuss treatment in a different way ii) identify their own preferences iii) understand their own willingness to accept trade-offs between process and outcome. These changes may, in turn, improve understanding of treatment, increase satisfaction with choice and increase adherence to treatment. Research is being conducted in other areas of healthcare currently (Ryan, 2017, Dowsey et al., 2016). It is important to note there were respondents who found the tasks challenging and appeared to fluctuate between alternatives and attributes without assimilation of knowledge and identification of their own preferences. This might be a reflection of the tool rather than the trade-off method per se, but it is important any future decision support tool is tested with a representative sample from the target group. Furthermore, the use of qualitative methods alongside quantitative analysis provided rich data that greatly improved the interpretation of findings and this approach is recommended for future evaluation of interventions.

Recommendations for improving understanding about hypodontia and its treatment and promoting a shared decision-making approach in dentistry are given in Tables 6.1 and 6.2 respectively.

Table 6 1: Recommendations for improving patient and family understanding of hypodontia and its treatment to support greater engagement in care decisions
The complexity of hypodontia and decision-making about care should be explicitly acknowledged, alongside an explanation of shared decision-making and the patient's and family's role.
Information exchange needs to establish existing knowledge and target information to address information deficiencies and misinformation. Areas of uncertainty and risk need to be discussed and understanding and interpretation assessed. Individuals may have different previous experiences and priorities in treatment that require a flexible and personalised approach to information provision.
Comprehensive, accessible and engaging patient information resources are required and co-development with target users is essential to optimise the presentation and relevance of information.
The hypodontia care pathway requires revision to provide more opportunities for adolescents and families to learn more about their condition and treatment options prior to decision-making.

Table 6 2: Recommendations promoting a collaborative approach to decision-making
Evidence-based training is warranted to raise professional awareness and skills in SDM. SDM should also be embedded throughout undergraduate and postgraduate training.
Rephrasing consultations and treatment planning as opportunities for shared decision-making may promote a more collaborative and patient-centred approach.
Decision-making ideally involves explanation and discussion of all options, not just those judged 'best' by the dental team, particularly for care where the evidence-base is equivocal.
Evidence-based interventions and process to promote and support SDM are required. These need to be developed and tested with the target population from a wide geographical area.
Trade-off tasks may be a useful method for helping patients think about treatment options and understand their own preferences; however, further testing is required prior to clinical application.

6.2 Implications for health service research

The research indicates there is considerable scope for further health service research within hypodontia but also more broadly across dentistry. Dental research has historically included a high number of observational studies examining treatment effect, and more recently the hierarchy of evidence underpinning evidence-based practice has promoted prioritisation of randomised controlled trials. While randomised controlled trials are the gold standard for testing the effectiveness of interventions, arguably this focus has undervalued other methods, such as qualitative approaches, which can provide rich and important data to understand complex phenomena. A broader approach to research in dentistry is required, that incorporates patient experience to improve care delivery and also draws on the expertise from other disciplines outside dentistry. Cross-discipline research teams can promote effective transfer of knowledge and skills to enhance the quality of research. Furthermore, greater involvement of patient stakeholders in research conception, design and dissemination is warranted to ensure the accessibility and relevance of research and effective use of research resources.

Improvement in professional-patient communication is a research priority and shared decision-making sits within this field. SDM research is required to provide evidence about the best way to integrate collaborative decision-making into dental care systems to gain the reported benefits for patients, professionals and the healthcare system. This includes integration of SDM care into routine practice, clinical guidelines and care pathways. It is important the value of decision-making science and behavioural psychology is recognised and applied in development and testing of clinical interventions. Integration of SDM has implications for the quality of service delivery and optimisation of health outcomes in hypodontia but also more broadly across dentistry, both in other interdisciplinary conditions and in routine dental care. Recommendations for future SDM research are given in Table 6.3.

In hypodontia specifically, evidence relating to treatment options and service delivery with relevant patient-centred outcomes is required. This might include development of a Core Outcome Set for hypodontia, examination of the feasibility of using of NHS data sets to examine components of service delivery such as waiting time and treatment time, and national audits to complement health service delivery research and to provide a more cohesive view of standards of care.

DCE is gaining traction as a key method for establishing patient preferences for inclusion in healthcare decisions; however, if it is to be used with adolescents, further examination of the method is required. This includes 1) establishing the validity of DCE compared to other stated preference methods, such as multi-profile BWS; 2) examining aspects of design that might impact on validity, such as attribute selection and framing, task construction (number of alternatives, effect of opt-out, preference elicitation format), explanation of the attributes and tasks; 3) examining the effect of different data collection methods, such as online versus face-to-face; 4) assessing the external validity of DCE methods; and 5) incorporating DCE or other preference data into the SDM process and undertaking longitudinal assessment to measure its impact on decisional conflict.

Table 6.3: Recommendations for further research to improve communication and shared decision-making in hypodontia and more broadly across dentistry
Examination of professional awareness and attitude to SDM and patient experience of SDM in routine clinical encounters to identify barriers and facilitators, and inform integration of SDM into practice.
Examination of effective approaches for communicating complex information, such as risk and uncertainty.
Evidence-based development and testing of decision support interventions to promote greater patient involvement in care decisions. This might include tools to foster choice awareness and participation at the desired level, screening tools to identify uncertainty and decision support needs, methods for eliciting values and preferences, methods for evaluating decision making processes.

6.3 Critique of methods

The research question underpinning this research arose directly as a result of clinical observation that decision-making in hypodontia was difficult for patients, families and the dental team. The research has provided high quality evidence in an area where information to guide patient-centered decision-making is lacking, which has real value and integrity for improving care delivery both in hypodontia and more widely across dentistry. The subsequent section thoroughly appraises the approach and methods to identify areas for improvements for future research.

The population in the research was determined by the research question and clinical appropriateness, but choice of an adolescent sample did present challenges, mostly in terms of recruitment. Hypodontia can cause significant quality of life impacts and some adolescents approached indicated they were not confident enough to participate. Hypodontia carries a high treatment burden and for some parents and adolescents, additional or longer visits to the hospital were often not possible. Despite these challenges, the adolescents who did participate were largely interested and engaged in the research and they provided a valuable contribution. The benefits of focusing on an adolescent population were numerous: gaining a different perspective on clinical care; acquiring insight into adolescent decision-making processes; examining the adolescent-parent relationship in a clinical context; giving a voice to an often under-represented group in dental research; developing further skills for engagement with adolescents in research. Similarly, including parents in the research widened the research perspective and provided a more accurate representation of the clinical problem under investigation.

This research provided the opportunity to use a number of different methods to look at decision-making, including some innovative methods, such as the use of social media to examine non-professional experience of hypodontia and observation of DCE as a decision-making tool. The social media study arose following presentation of the method at a conference (Noll et al., 2017), which sparked an interest in the use of social media as a research tool. At the time of this study the approach was relatively novel, and the study was initially undertaken as an exploratory study to see if it would work in hypodontia. The results proved a valuable source of information about patient and family experience that was not captured by other methods. Similarly, the examination of patient information resources was not included in the original research plan but was included after the systematic literature review to add complementary information. Analysis of information resources, alongside observation of consultations, provided a new perspective on clinical care, which to me as a dental professional had become somewhat familiar and routine.

The combined methods allowed triangulation of findings and supported the development of some recommendations to improve clinical care in hypodontia and to drive future research; however, it is acknowledged that more research is needed in some areas to provide detail and increase transferability into different contexts. The examination of patient information did not capture patient opinion on current resources, and the numbers for the clinical consultations were low. The interviews were conducted early in the research when I was inexperienced in qualitative methods and the details of the research questions were not fully formed; it is likely that further interviews now would produce additional information that may clarify or validate the findings from the research.

Preference elicitation research has surged in the last 20 years in response to the drive to incorporate stakeholder preferences into a broad range of healthcare decisions and DCE is an increasingly popular method. Examination of the relevant literature throughout the research suggests there is a risk that DCE use is being divided between highly theoretical and methodological applications, which are difficult to interpret without extensive choice modelling knowledge, and use in applied health research, such as this, but without much evaluation of method and tool. In this research considerable effort was made to apply best practice guidance where possible and critique each stage of DCE development; however, understanding of DCE developed throughout the research and in hindsight there are changes to development and design that would be considered.

Firstly, much time was spent on attribute identification through the mixed methods used to understand hypodontia decision-making practice, but less time was allocated to selection and framing of attributes. Including stakeholders provided a valuable perspective but a larger group of stakeholders and use of group methods to discuss attributes would have been beneficial, although it may ultimately not have changed the attributes selected. Secondly, DCE tasks are clearly challenging for some respondents and dedicating more time to trialling different methods for explaining the attributes and tasks might have improved the quality of responses for this group. Thirdly, it was not clear how respondents would characterise the attributes of their own treatment and having no treatment. Incorporating methods to understand this would be valuable to interpretation of data and understanding patient perception of their care. Finally, the selection of a repeat task to assess reliability provided interesting data about intra-examiner agreement for discussion, but it is debatable whether a dominant choice set might have been more useful to identify poor quality responses.

The DCE methods provided data that was coherent with qualitative feedback and clinically plausible, suggesting the preferences elicited were valid. The DCE choice tasks used were relatively complex and observation suggested there was variability between respondents in their perception of the tasks. This makes assessment of validity of the method generally for all adolescents difficult. While it appeared most participants were able to complete the DCE tasks, examination of the quality of data suggests around 9% were poor quality and there was a class of around 12% where preferences were difficult to interpret. It could be argued that from the outset DCE was likely to be too complex for adolescent respondents and that the approach is reductionist for such a complex decision-making area. These are valid criticisms; however, a major strength of this research was the application of qualitative methods alongside the DCE, which resulted in much more powerful data. The inclusion of a method to observe DCE completion by dyads evolved during the research as an exploratory study, so the scope was limited by time and resource constraints in the PhD. The method proved extremely valuable, providing rich and clinically relevant data and in hindsight it is possible that removing the online survey and focussing on this approach would have been beneficial.

The statistical analysis involved in DCE is complex and there was steep learning curve. It is acknowledged that further statistical analysis could be undertaken but this was outside the scope of the thesis and it is likely further work will be of methodological interest, rather than clinical benefit. A pragmatic approach to analysis was taken given the applied nature of the research and the researcher's limited knowledge and experience with DCE analysis.

Stakeholder involvement in the design and conduct of this research provided a perspective that significantly improved the process and outcome. The inclusion of PPI contributors reflects a change in approach to research that has been somewhat slow to be adopted in dentistry. The benefit of involving patients as collaborators in the research to improve research design and delivery requires further promotion in dentistry. Stakeholder involvement was not without challenges. It was a new area for me requiring training and support particularly around recruiting from the clinical environment. The age of patient contributors meant some were not able to help for the entirety of the 4-year research period due to educational commitments and changes in personal circumstances. This research did not incorporate any methods to evaluate the impact and experience of PPI contributors but this will be built into future research.

Overall, reflection on the population and methods indicates that the approach taken was appropriate for the purpose of the research and the findings have delivered clinically relevant information that can be used to guide clinical change and future research. Cross-discipline supervision and engagement with stakeholders has improved the quality of the research to provide information that will be applicable beyond hypodontia. Although improvements are possible, many of the methods can be transferred to other areas of dentistry with only minor modifications. The research was ambitious and at times challenging; however, the quality and value of the research as a whole reflects the considerable time and effort of the participants, contributors, clinical colleagues and research supervisors.

6.4 Conclusions

1. Patient and family understanding of hypodontia and its treatment is inadequate in many cases, which presents a barrier to meaningful engagement in treatment decisions.

2. Hypodontia care decisions require a collaborative approach, with explicit statement of choice, an invitation to participate, appropriate information exchange about important issues and integration of patient and family preferences.

3. Appearance is highly important in decisions about hypodontia treatment. Other important attributes include severe problems, severe discomfort, a long treatment time and bite after treatment. There was significant heterogeneity in preferences across respondents.

4. Trade-off tasks helped some people think about treatment and identify their preferences and this might be useful for future decision support tools; however, not all people found the tasks helpful and any future tools require thorough testing using appropriate methods to understand underlying cognitive processes.

5. DCE methods appear to elicit valid preferences for the majority of respondents; however, around 20% respondents in this study returned poor quality data. Further work is required to evaluate aspects of DCE design and application for adolescent and joint preference elicitation.

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Appendix

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Appendix Table 1: A summary of studies examining the impact of hypodontia on oral health-related quality of life (OHRQoL)

Study	Population	Method	Key findings
(Akram et al., 2011) UK	11-18 year olds Non-syndromic hypodontia Any severity Any stage of treatment (n=22)	Focus groups	▪ **Treatment:** Length and complexity of treatment, anxiety about treatment, future loss of primary teeth, artificial teeth ▪ **Effect on daily activities:** limitations to eating, effect on speech, playing musical instruments, wearing sports guard, brushing ▪ **Appearance:** Gaps, small teeth, self-conscious smiling, appearance of tooth replacement ▪ **Reaction of other people:** Bullying, over protective family, not being ‘normal’, hiding condition from peers
(Meaney et al., 2012) Ireland	16-25 year olds Non-syndromic hypodontia Any severity Any stage of treatment (n=10)	Interviews	▪ **Transition from childhood to adulthood**: change in understanding, participation and expectations ▪ **Waiting time**: Initial wait & delay between stages frustrating, increased satisfaction once treatment starts ▪ **Importance of appearance**: motivation for treatment, concern increases with age, linked to self-consciousness
(Anweigi et al., 2013a) Ireland	16-34 year olds Non-syndromic hypodontia Any severity Any stage of treatment (n=82)	Oral Health Impact Profile (OHIP)	▪ Most reported $\geq$1 recent OHRQoL ▪ Females reported higher impact ▪ Impact not related to severity of hypodontia ▪ Anterior & posterior missing teeth associated with increased psychological discomfort ▪ Increasing age associated with increasing functional limitation
(Hashem et al., 2013) Ireland	18-45 year olds with moderate-severe hypodontia (n=41) Matched controls (routine dental patients)	OHIP Rosenberg Self Esteem Scale	Hypodontia group compared to control: ▪ Overall mean OHIP score significantly higher ▪ Mean score higher in 5/7 OHIP domains (physical, psychological and social domains) ▪ No differences were found in self-esteem
(Kotecha, 2011) UK	11-14 year olds Mild hypodontia (n=43) Severe hypodontia (n=43) Control (non-hypo., low treatment need) (n=30)	Child Perception Questionnaire (CPQ11-14) Parent-CPQ	Hypodontia group compared to control: ▪ Significantly worse scores in all domains ▪ No significant association with hypodontia severity ▪ No effect from gender, SES, agenesis site, retained primary tooth ▪ Moderate correlation between child and parent scores

(Wong et al., 2006b) Hong Kong	11-15 year olds Moderate-severe hypodontia (n=25)	CPQ	▪ All participants reported: ≥1 OHRQoL impacts, ≥1 oral symptoms, some social impact ▪ 88% reported functional limitations and emotional well-being impacts ▪ Correlation between severity of hypodontia and impact with greater impact when retained primary teeth accounted for
(Locker et al., 2010) Canada	11-14 year olds Non-syndromic hypodontia Any severity (n=36)	CPQ	▪ 77.8% reported ≥1 impact ‘often’ or ‘everyday/almost everyday’ ▪ Functional limitation 61.1% (chewing, eating slowly) ▪ Oral symptoms 27.8% (bad breath, food catching) ▪ Emotional well-being 19.4% (concern about appearance, other people’s reaction) ▪ Social well-being 16.7% (arguing with family, being asked about missing teeth) ▪ No correlation with severity, location of missing teeth, age, gender
(Laing et al., 2010) UK	11-16 year olds Non-syndromic hypodontia Any severity Any stage of treatment (n=62) Control: Non-hypodontia but requires treatment (IOTN 4-5) (n=61)	CPQ VAS to measure chewing function and dental aesthetics	Hypodontia group compared to control: ▪ CPQ: Overall score & all sub-group scores were higher but not significant ▪ No effect on CPQ score from gender, age, severity or location of hypodontia ▪ VAS: Significant relationship between the function score and level of hypodontia ▪ Both methods suggest presence of primary teeth in posterior sextants reduces functional limitations
(Hvaring et al., 2014a) Norway	10-17 years old Non-syndromic hypodontia Any severity (n=62) Control: Non-hypodontia but requires treatment (IOTN 4-5) (n=101)	Generic Oral Impact on Daily Performance (OIDP) Condition-specific OIDP	▪ More people in hypodontia group rated their oral health as poor ▪ More people in hypodontia group were satisfied with their own dental appearance ▪ Perceived poor oral health was related to greater OHRQoL impact ▪ Dissatisfaction with dental appearance was related to OHRQoL impact ▪ The generic OIDP identified a similar level of impacts between the hypodontia and control group ▪ The condition-specific version had significantly higher discriminative ability in four domains; showing teeth, social contact, emotional status and speaking

Appendix Table 2: Study eligibility and search criteria to identify stated preference studies involving adolescents	
Eligibility criteria	
Population	Adolescents (aged 18 years or younger) participating in a stated preference experiment in an area of healthcare
Intervention	Any healthcare decision context
Outcomes	Preferences of adolescents
Study types	Studies using multi-attribute stated preference methods
Search strategy	
Data sources	Ovid: Medline, Embase, PsychINFO, PsychARTICLES, All EBM Reviews; Pubmed; Econlit Grey literature: reference lists of relevant publications and reviews and targeted searching of online resources: NICE and SIGN; www.clinicaltrials.gov; Google Scholar; Conference abstracts
Search terms	Search terms for MASP (see Appendix box X) AND (Adolescen$ OR adolescent OR child OR teenager OR youth OR girl OR boy OR pubertal).

Appendix Table 3: Study eligibility criteria and search strategy to identify stated preference studies for preference elicitation from dyads	
Eligibility criteria	
Population	Related respondents participating in a stated preference experiment together No restrictions based on age, gender, ethnicity, location
Intervention	Any decision context
Outcomes	Preferences of dyads
Study types	Studies using multi-attribute stated preference methods
Search strategy	
Data sources	Ovid: Medline, Embase, PsychINFO, PsychARTICLES, All EBM Reviews; Pubmed; Econlit Grey literature: reference lists of relevant publications and reviews and targeted searching of online resources: NICE and SIGN; www.clinicaltrials.gov; Google Scholar; Conference abstracts
Search terms	Search terms for MASP (see Appendix box X) AND (Joint OR proxy OR parent$ OR mother$ OR father$ OR care$ OR guardian$ OR famil$ OR surrogate$ OR advocate$ OR dyad)

Appendix Table 4: Inclusion and exclusion criteria for study selection for systematic review of hypodontia literature

	Included	Excluded
Population	People with non-syndromic hypodontia No restrictions on age, ethnicity or severity of hypodontia	People with syndromic hypodontia Treatment for tooth loss with any other aetiology
Intervention	Any type of dental treatment undertaken to manage hypodontia: Management of retained primary teeth; Orthodontic treatment to eliminate or redistribute space; Restorative treatment for tooth replacement (fixed prostheses, removable prostheses, implant-supported restoration) or tooth camouflage; Tooth auto-transplantation Any aspect of hypodontia service delivery	Simulated treatment such a treatment prediction using computer software and hypothetical treatments Purely laboratory-based interventions Future treatments still in development e.g. biological methods for growing and replacing missing teeth
Comparator	Any treatment listed above or no treatment Studies without a comparator were also eligible	
Outcomes	Any evaluation of hypodontia care (treatment methods or service delivery)	Aetiology of hypodontia Prevalence of hypodontia
Study design	Systematic reviews with data synthesis that produced primary summary data (e.g. meta-analysis) Experimental designs Observational studies Economic studies Qualitative studies Quality improvement reports (audit, service evaluation)	Non-systematic reviews (Reviews without evidence synthesis, Expert opinion) Case series with less than 10 participants or single case reports Conference abstracts with inadequate information regarding methodology and results
Study characteristics	Studies originating from any country Non-English publications	Studies published prior to 1970 Studies not available in full text in English after reasonable attempts to obtain or translate

Appendix Table 5: Search strategy for identifying empirical research reports for systematic review of hypodontia literature

Electronic databases	1. Pubmed 2. Medline via Ovid 3. EMBASE via Ovid 4. Scopus 5. Web of Science (Science Citation Index and Conference Indexes) 6. Dissertations and Theses database	Search terms for hypodontia AND treatment methods
	7. Cochrane Collaboration (DARE, CDSR) 8. NICE 9. SIGN 10. Trials register ww.clinicaltrials.gov	Search terms for hypodontia only
Grey literature searches	▪ Hand searching reference lists of relevant publications and reviews ▪ Additional citation searching for relevant papers identified in searches ▪ Personal contact via email to the members of two dental specialty societies (British Orthodontic Society BOS and Restorative Dentistry UK RDUK) to identify unpublished academic work ▪ Hand searching of the Clinical Effectiveness Bulletins of Dental Specialty Societies (BOS, BSPD) ▪ Web searches for unpublished literature including conference abstracts, research in progress and institution reports using the top three search engines (Google, Bing and Yahoo), an academic search engine (Google Scholar) and a search engine that does not filter results based on personalised previous searches (GoGoDuck). The first 50 (5 pages) results were checked for additional sources to include in the review.	

Appendix Table 6: Summary of research studies & audits grouped by purpose and reported in terms of intervention, design and key findings

Key: MLIA = missing lateral incisor agenesis
MSPA = Mandibular second premolar agenesis
QoL = Oral health-related Quality of life)
OSO = Orthodontic space opening
OSC = Orthodontic space closure

Intervention	Study design (n)	Key findings (Level of evidence)
Evaluate effectiveness of single intervention		
Maintenance of mandibular primary second molars	Prospective observational without control (1) Retrospective observational without control (4) Cross-sectional without control (2)	**Tooth survival:** Teeth survive 1-20 years in 86-100% (Moderate). **Root resorption:** Extent is variable and difficult to predict (Moderate), may be correlated with infraocclusion & age (Low). **Infra-occlusion:** Variable with mean of approx. 1-2mm (Low). **Occlusion:** Teeth cause variable but generally minimal tipping of adjacent teeth (Moderate). **Coronal health:** Restorations required in 25-50% (Low).
(Bjerklin and Bennett, 2000, Bjerklin et al., 2008, Ith-Hansen and Kjaer, 2000, Sletten et al., 2003, Kurol and Thilander, 1984, Hvaring et al., 2014b, Kjaer et al., 2008)		
Loss of mandibular primary second molar +/- space closure techniques	Clinical controlled trial (1) Retrospective observational with control (3)	**Occlusion:** Extraction may result in acceptable space closure with minimal effect of the surrounding dentition if timed appropriately (Low). Staged extraction (hemisection or slicing) may promote desirable space closure (Low). **Harms:** No difference in harms (TMD symptoms & marginal bone loss) found between planned & unplanned extraction groups (Low). **Treatment duration:** Similar for staged extraction (hemisection) of primary teeth & extraction of premolar teeth (Low).
(Valencia et al., 2004, Lindqvist, 1980, Northway, 2004, Mamopoulou et al., 1996)		
Orthodontic space closure in MLIA and MSPA	Retrospective observational with control (1) Retrospective observational without control (2)	**Occlusion:** OSC for MLIA & MSPA does not result in detrimental occlusal outcome (Low) **Periodontium:** OSC for MLIA does not affect periodontal health (probing pocket depth, bleeding on probing, gingival recession & mobility compared to control) (Moderate). **Harms:** OSC for MLIA does not increase TMD compared to a control (Moderate).
(Rosa et al., 2016, Zimmer et al., 2007, Zimmer and Seifi-Shirvandeh, 2009)		

Resin-bonded bridges (RBB)	Prospective observational with control (1) Prospective observational without control (1) Retrospective observational without control (2)	**QoL:** RBBs cause moderate improvement in QoL in adults compared to control (Moderate). **Survival:** *Conventional RBBs:* Mean/median survival in adolescent & adults with MLIA is 59.3/59 months respectively. Operator experience increase mean/median survival to 72.6 and >100 months respectively. 2-year survival is approx. 97% (Low). *Fibre-reinforced composite RBBs:* Survival in adolescents with MLIA is 94% (Low). **Treatment success:** *Conventional RBBs:* 2-year success 92% (Low). *Fibre-reinforced RBBs:* Success in MLIA is 100% (Low). **Technical complications:** Most common cause of failure was bond failure, often secondary to trauma (Low)
(Anweigi et al., 2013b, Allen et al., 2016, Garnett et al., 2006, Spinas et al., 2013)		
Denture for children with severe hypodontia	Retrospective observational without control (1)	**Success:** Majority required replacement within the first 3.5-4 years (Low) **Technical complications:** Failure rate high due to fracture & wear secondary to degeneration of materials. Early fracture (<6months) due to incorrect design & fabrication (Low).
(Hobkirk et al., 1989)		
Implant-supported tooth replacement in adults	Clinical controlled trial (2) Prospective observational without control (3) Prospective observational with control (2) Retrospective observational with control (2) Retrospective observational without control (3)	**QoL:** Significantly increased 1-month & 2-years post-treatment (Low). **Function:** Significantly reduced functional impairment & increased patient-reported masticatory ability (Low). Increased occlusal tooth contact, maximum bite force and masticatory performance (Low) **Satisfaction with treatment:** Mean overall appreciation rated 8 on a 10-point scale (Low) **Implant appearance:** Single tooth implant-supported restorations rated highly for appearance after 2-3 years (Low). Tooth colour greatest challenge (Low). All-ceramic crowns colour match > metal-ceramic crowns (Low). **Survival:** *Implants:* 100% over 2-3 years and 89.9% over 18 years. Higher in mandible (96%) than maxilla (86%) (Low). *Implant crowns and abutments:* 97% over 3 years in young adults (Low). *Pre-implant block graft:* Graft survival 100%. Implants placed into grafts: 95.2% over 3 years (Low). **Success:** *Implants:* 97% after 3yrs (marginal bone levels & soft tissue conditions) (Low). Bone augmentation frequently needed - provides bone gain approx. 4-6mm horizontally & 0-3mm vertically after 6 months. Soft tissue breakdown in approx. 1/3 patients (Low) **Marginal bone levels:** Accumulated mean marginal bone loss <1mm & probing depth approximately 1-2mm around the implant after 36 months (Low) **Harms:** Bone augmentation may be related to persistent complaints (Low) **Biological complications:** Low in single tooth implants after 3 years - includes increased probing depths, bleeding and marginal bone loss (Moderate). Crown material may affect biological complications, abutment material may affect amount of marginal bone loss but not plaque retention or bleeding (Low). Poor crown marginal adaptation may be associated with fistulas, suppuration and

		bone loss (Low). **Technical complications:** Include chipping of the temporary acrylic crown and loosening of the implant (Low). Implant crown material may affect technical complications (Low).
(Hosseini et al., 2011, Degidi et al., 2009, Allen et al., 2017, King et al., 2016, Zarone et al., 2006, Hosseini et al., 2013) (Creton et al., 2010, Mangano et al., 2014, Hu et al., 2011, Finnema et al., 2005, Nissan et al., 2011)		
Implant-supported tooth replacement in children	Retrospective observational without control (1)	**Survival of implants:** 89% over mean loading time of 11 years. **Success of implants:** 17% based on bleeding, plaque, probing depth and bone loss. **Complications:** Implant failure attributed to bone loss associated with peri-implantitis (Low). Implants move relative to surrounding teeth & may require replacement of the restoration or implants (Low)
(Heuberer et al., 2015)		
Preparatory treatment for implant placement	Retrospective observational with control (4) Retrospective observational without control (2)	**Bone volume:** Movement of teeth increased alveolar bone volume during orthodontic treatment but this reduced during the retention period. Overall bone volume increased compared to pre-treatment (Moderate). Implant placement retained more bone than sites without implant placement (Low). No significant differences in bone deficiency between treatment early (age 11-12 years) or later (age 14-15 years) but with considerable individual variation (Low). **Root position:** Relapse following orthodontic preparation is inconsistent but may prevent implant placement in approximately 10% of cases (Low).
(Novackova et al., 2011, Uribe et al., 2013a, Uribe et al., 2013b, Eliasova et al., 2014, Beyer et al., 2007, Olsen and Kokich, 2010)		
Tooth autotransplantation for MSPA in adolescents	Retrospective observational without control (3)	**Survival:** Moderate-term survival of autotransplanted teeth is >95% (Moderate). **Treatment success:** >91% based on pulp, root and periodontal measures of health (Low). Success rate is reduced to 82% if root formation is complete at the time of transplantation (Low). Prognosis is highest when root development is 50-75% at the time of transplantation (Low). **Pulp health:** Pulp survival is associated with the stage of root development: partly formed roots showing much greater potential for regeneration than those with more developed roots (Low). Pulpal obliteration is a sign of pulp healing (Moderate). **Root development:** Root development continues although final root length is variable & often does not reach normal length (Moderate). Probability that reduction in root length of transplanted premolar >2.3mm (low clinical impact) estimated to be <5% (Low). **Treatment failure:** Common reasons failure were root fracture, root resorption, periapical and periodontal pathology (Moderate).
(Slagsvold and Bjercke, 1974, Josefsson et al., 1999, Jonsson and Sigurdsson, 2004)		

Comparison between treatments		
Orthodontic space closure (OSC) vs orthodontic space opening (OSO) with tooth replacement for MLIA	Cross-sectional (6) Retrospective observational (3)	**Appearance:** Self-reported satisfaction with smile was higher for OSC than OSO+implant placement (Low). Smile attractiveness of OSC was generally judged to be higher than OSO+tooth replacement by laypeople and dental professionals (Moderate). Colour & shape of teeth are most important to dental appearance. Dentists also notice symmetry/midlines while laypeople noticed spacing (Low). Sex is associated with differences in dental attractiveness judgments by dental professionals & laypeople (Low). Outcomes from symmetrical treatment are rated as more attractive than asymmetric treatment regardless of treatment method (Low). Gingival exposure influenced judgements of smile attractiveness (Low). Dental aesthetic parameters suggest OSC with reshaping provides closest outcome to non-hypodontia treatment outcome (Low). **Dental health:** There is no significant difference in periodontal health (plaque index, bleeding on probing, probing depth>3mm, papilla index and gingival recession) between OSC and SO and implant placement. Increased probing depth >3mm was seen in approx. 1% (Low). Pocket depths are higher around anterior teeth with fixed or removable tooth replacement than those with OSC (Low). Gingival index is significantly worse for prostheses than natural teeth (OSC) likely due to plaque accumulation 7 mechanical irritant levels (Low). Signs/symptoms of TMD (measured with the Helkimo Questionnaire) were not different between those with OSC or SO and implant placement (Low). **Function:** Minor differences exist in the occlusal evaluation between OSC and SO with tooth replacement but these are not significant (Low). Premature functional contacts appeared to be correlated to periodontal pocket depth (Low).
(Armbruster et al., 2005, De-Marchi et al., 2014, Pinho et al., 2015, Pini et al., 2013, Qadri et al., 2016, Robertsson et al., 2010, Marchi et al., 2012, Nordquist and McNeill, 1975, Robertsson and Mohlin, 2000)		
Tooth-supported vs. implant-supported tooth replacement	Retrospective observational with control (3)	**OHRQoL:** Scores significantly higher for hypodontia than non-hypodontia controls - greatest impact from functional limitations & physical pain. Treatment significantly improved QoL scores. No difference in scores for different severities of hypodontia. (Low). **Satisfaction:** Hypodontia treatment outcome was high; 98% of patients with implant-supported and 84% with tooth-supported reconstructions were satisfied or very satisfied with the treatment result (Low). **Appearance:** Dental professionals judged restorations to be acceptable in 92% of implant reconstructions and 83% of tooth supported restorations (Low), with mucosal discoloration the greatest concern (57% patients). >95% showed acceptable crown morphology & colour match, but only 6% showed normal papilla level (Low). No significant differences reported by patients for problems with appearance between implant-supported (41%) & tooth-supported (47%) restorations but non-hypodontia control lower (14%) (Low).

		Treatment success: Failure approx. 17% implant-supported crowns, 13% implants and 14% tooth-supported prostheses over 4.6-25 years (Low). **Complications:** 13-41% implant-supported crowns/prostheses, 0-20% tooth-supported crowns/prostheses. Implants show complications significantly earlier (Low). Incidence of root resorption, marginal bone loss, periapical and peri-implant pathology and technical complications was low (Low). Majority of people treated with implants (88%) showed small difference (0-4mm) in marginal bone levels between implants & neighbouring teeth (Low). Correction or replacement of restorations due to poor marginal adaptation required in 20% tooth-supported & 9% of implant-supported restorations (Low). **Cost:** Initial cost of rehabilitation with implant-supported methods £1855 per unit (£775-4250) & tooth-supported methods £623 per unit (£450-725) (Low). Maintenance est. £113/yr for supportive periodontal care (Low). Re-treatment: 20% tooth-supported restorations - cost £3476 & 42% implant-supported restorations - cost £5575 (Low).
(Incici et al., 2009, Krieger et al., 2009, Dueled et al., 2009)		
Evaluation of treatment experience		
Treatment experience	Cross sectional qualitative (2) Cross-sectional survey without control (1)	**Quality of life:** Pre-treatment patients felt self-conscious and uncomfortable smiling. Anterior retained primary teeth were a concern for appearance (Moderate). Orthodontic treatment improved patient happiness & confidence (Moderate). **Patient experience:** Understanding diagnosis and expectations from treatment may change during the treatment period (Moderate). Multidisciplinary clinics provide opportunity for dental professionals to explain reasons for attendance and for patients to engage in treatment planning & decision-making (Moderate). Satisfaction with orthodontic component of clinical care & service is good (Moderate). Transition between clinicians can be a source of concern for patients (Moderate). Travel distance & time off school are ‘costs’ of treatment (Moderate). **Patient satisfaction:** Delays in service provision are a source of frustration (High). Inadequate explanation of treatment duration reduced patient satisfaction (Moderate). Initial consultation through a specific multidisciplinary hypodontia clinic increases patient satisfaction (Low). Good communication & involvement in decision-making improved satisfaction (Moderate). Dental appearance is important determinant of satisfaction with outcome (High). **Service utilisation:** Dental appearance and obtaining information about treatment timing have been cited as an important factor in seeking treatment for hypodontia (High). **Treatment efficiency:** Initial consultation through a specific multidisciplinary hypodontia clinic delivers an efficient service (Low).
(Tams and Ashley, 2013, Meaney et al., 2012, O'Keeffe et al., 2016)		

Audit of service delivery		
Access to services in Scotland	Cross-sectional questionnaire	Access to services varies geographically & in some areas is limited. The majority of patients requiring restorative care for hypodontia had not attended a joint orthodontic-restorative clinic.
(Borrie and Cord, 2014)		
Efficiency of assessment clinic	Retrospective audit of notes	93% of patients had evidence of a treatment plan following attendance at an orthodontic-restorative clinic. 60% of patients had written correspondence to the GDP regarding their role in treatment provision, for 40% the provider was unclear.
(Walker and Dyer, 2009)		
Treatment delivery	Prospective audit using proforma (2) Retrospective audit of notes (3)	**Treatment duration:** Average orthodontic treatment time 21-25mnths (range 19-37). 58% patients completed orthodontic treatment within 24mnths. Average treatment time for restorative treatment was 5mnths (range 2–13). 33.3% completing restorative treatment within 2 months (average time 30.86 weeks). **Waiting time:** 77.5% of patients were seen within 18 weeks, with an average wait of 8.43 weeks. Average time between completion of orthodontics and provision of restorative prosthesis was 6 months (range of 1–15). **Treatment adherence:** Failure to complete treatment estimated to be 15-18.5%. Failure to complete associated with dental health issues (6%), patient did not want treatment (5-19%) and a change in treatment plan (2 patients)
(Crawford et al., 2010, Borrie and McIntyre, 2008, Parvizi et al., 2010, Shah and Gwilliam, 2014, Wazani and Sood, 2016)		
Record keeping	Retrospective & prospective audit of notes (1)	The availability of appropriate clinical records, including radiographs, study models and photographs, was highly variable and generally poor. Quality of record keeping for patients attending a hypodontia clinic is inadequate.
(Shah and McKaig, 2016)		

Appendix Table 7: Method for identifying and selecting participants for the Patient Information Resource survey

Dental care professional group	Method of identification	Sample selection
Lead for 'Hypodontia' MDT clinics across the UK	Ectodermal Dysplasia Society list 2015	All Leads included (n=20)
Specialist Orthodontists across the UK	British Orthodontic Society list of Orthodontic Specialists http://www.bos.org.uk/Find-a-Treatment	List filtered by treatment type to include 'NHS adults, NHS children, private adults, private children' (n=316) Duplicates excluded resulting in final sample (n=145)
Specialists in Paediatric Dentistry across the UK	General Dental Council list of Specialists in Paediatric Dentistry	All included (n=218)
Specialists in Restorative Dentistry across the UK	British Society of Restorative Dentists (n=86) British Society of Prosthodontists (n=6)	All included (n=92)
General Dental Practitioners (GDPs) in West Yorkshire	West Yorkshire Area Team list of dental practices http://www.nhs.uk/Services/Trusts/Dentists/DefaultView.aspx?id=89783	Random selection from list (n=174)

Appendix Table 8: Search strategy for systematic online search		
Search terms	Hypodontia; developmentally or congenitally missing teeth; absent teeth; tooth dental aplasia; tooth agenesis	
Search engines	Google (www.google.co.uk); Bing (www.bing.com); Yahoo (www.uk.yahoo.com)	
Targeted website searches	NHS Choices, Department of Health, Royal College of Surgeons, British Dental Association, British Dental Health Foundation, British Orthodontic Society, British Society of Paediatric Dentistry, British Society of Restorative Dentistry, Restorative Dentistry UK, Association of Dental Implants.	
Resource selection criteria	**Inclusion**	**Exclusion**
	▪ Information relating to hypodontia and its treatment ▪ Websites aimed at patients, families and the public ▪ English language ▪ Adequate length to allow quality assessment (approximately >500 words)	▪ Information about other dental conditions ▪ Websites aimed at healthcare professionals ▪ Marketing material ▪ Inadequate information / length to enable assessment ▪ Not available in English

Appendix Table 9: Search terms for social media developed through scoping searches and consultation with colleagues			
Search concept	**Key words**		
Missing teeth	Hypodontia Anodontia Oligodontia Tooth agenesis	Tooth aplasia Missing teeth Absent teeth	Gappy teeth Spaced teeth Gap between teeth
Treatment methods	Dental Dentist	Braces Orthodontic	Dental bridge Dental implant Dentures

Appendix Table 10: Selection criteria for social media records for content analysis

	Included	Excluded
Author	Individual affected by hypodontia	Dental professional; Academic institute; Individual / group with commercial interest; Government
Type of post	Text post Caption for photograph / video	Photograph / video content; Link to other content; Advertisement / promotion
Theme of post	Sharing experience / opinion Information gathering Information sharing	Advertising
Target audience	Public / peers Patients / parents Dental professionals	N/a
Other	English or able to translate	Non-English and unable to translate

Appendix Table 11: Search results for social media (*number of records reported as available)

Source	Accessible records (search term hypodontia/ oligodontia)	Excluded		Relevant records
		Duplicate	Irrelevant / Language	
Facebook	32 / 7	5	4	30
Twitter	96 / 114	5	37	168
Google+	72 / 9	3	11	65
Instagram	69 (110*) / 125 (181*)	10	16	168
Reddit	14 / 6	1	2	17
Tumblr	16 / 11	4	1	22
Total	**571**	**28**	**71**	**470**

Appendix Table 12: Example of knowledge questions for preference elicitation instrument for piloting

	True	False	I don't know
Consequence: Which of these problems can people with missing teeth have?			
Health of the teeth e.g. decay, gum problems	☐	☑	☐
Gaps between the teeth	☑	☐	☐
Baby teeth do not fall out	☑	☐	☐
Difficulty to eat or talk	☑	☐	☐
Cause: What causes teeth not to develop (naturally missing teeth)?			
Not brushing the teeth	☐	☑	☐
Eating sugary foods	☐	☑	☐
Knocking the teeth	☐	☑	☐
Other people in the family have missing teeth (genetic)	☑	☐	☐
No known reason	☑	☐	☐
Consequence: What will happen if people do not have dental treatment for their missing teeth?			
The teeth will get better themselves	☐	☑	☐
The teeth will stay the same	☑	☐	☐
If the baby teeth fall out, there may be more gaps	☑	☐	☐
New teeth will grow in the future	☐	☑	☐
Control: What do you think people can do to help with their missing teeth?			
Brush the teeth twice a day	☐	☑	☐
Avoid sugary food	☐	☑	☐
Protect the teeth e.g. wear a mouth guard for sport	☑	☐	☐
See a specialist dentist for treatment	☑	☐	☐
Nothing	☐	☑	☐
Cure: What do you think dentists can do to help with missing teeth?			
Baby teeth can be kept	☑	☐	☐
Braces can be used to move the teeth around	☑	☐	☐
Teeth can be replaced with false teeth	☑	☐	☐
Teeth can be replaced with a dental implant	☑	☐	☐
Timeline: How long does missing teeth last for?			
Missing teeth is a life-long condition	☑	☐	☐
People can grow out of missing teeth	☐	☑	☐

Appendix Table 13: Example of questions to establish previous treatment for hypodontia

What type of treatment have you already had <u>for your missing teeth</u>?

Please tick all that apply

☐ Fillings in baby teeth

☐ Extraction of baby teeth

☐ Fillings in permanent teeth

☐ Fixed braces

☐ Removable braces

☐ False teeth that I can take out (a plate or denture)

☐ False teeth glued to other teeth (bridge)

☐ Metal screw placed in the jaw to hold a false tooth (implant)

☐ Retainers (after braces to keep the teeth straight)

☐ Other (please specify)

Appendix Table 14: Comprehensive list of all attributes identified, grouped by concept

Key for sources:
SR = Systematic review of hypodontia literature
PIR = Patient Information Resources
SM = Social Media
PI = Patient-Parent Interview
CC = Clinical Consultation
DCE = Systematic review of DCE studies in Dentistry

Attribute	Descriptors	Source
Time		
Treatment duration	Overall treatment duration	SM
	Accuracy of estimate of treatment duration	SR, SI
	Treatment time for individual treatment methods	SR; PIR; CC
	Time for treatment effect / health improvement	DCE
	Overall time off school/work for treatment	PIR; CC; SI
	Lack of progress and delays	SI
Appointments	Appointment schedule	PIR; CC
	Total number of visits	DCE
	Time taken for interdisciplinary consultation appointment / specific procedure	PIR; SM
	Time off school / work for treatment for patient & family	PIR; SI
	Time taken for travel to appointment	DCE; SI
Waiting time	Waiting time for consultation with interdisciplinary team	SR, DCE
	Waiting time between stages of treatment	SR
	Timing - Delay start of treatment due to dental development	PIR; SI
	Timing – Delay treatment start to minimise transition period between stages of treatment	CC
	Time to wait for treatment due to waiting list	DCE; CC
	Time to wait at each appointment	SI
Service configuration		
Treatment planning	Initial consultation is provided through an interdisciplinary clinic	SR
	Treatment plan provided / supported by a hypodontia clinic	SR
	Treatment provider is explicitly stated	SR
	Opportunity to be involved in treatment planning and decision-making	SR

Adherence to treatment	Failure to complete treatment	SR
	Change in treatment plan	SR
Staff	Consistency in clinician / transition between clinicians	SR: CC; SI
	Treatment provider: Specialist or dentist / Expertise of staff / specialist service	PIR; CC; SI
	Dentist part of wider dental care team	DCE
Facilities	Business hours	DCE
	Equipment available	DCE
Location / access	Hypodontia clinics are held in main sites and peripheral sites refer to these	SR
	Patients have access to a multidisciplinary team clinic	SR
	Centres have access to specific treatment	SR
	Distance to travel / location of visits	DCE
	Travel to unfamiliar care setting	SI
	Difficulties with parking	SI
	Cost of travel	SI
Costs associated with receiving treatment		
Direct costs	Cost of initial treatment	SR; PIR; SM; CC
	Option for payment plans / insurance coverage (non-UK)	PIR; DCE; SM
	Benefits / coverage from insurance plans (non-UK)	DCE; SM
	Eligibility for NHS treatment (UK)	PIR; SM; CC
	Cost as a barrier to care	SM
Indirect 'costs'	Travel distance	SR
	Time off work/ school	SR
Impact of treatment		
Experience of treatment	Experience of treatment process e.g. implant placement, whole journey through treatment	SM
	Satisfaction with overall hypodontia care	SR; SI
	Satisfaction with specific treatment	SR
	Satisfaction with provider / treatment by provider	SM; SI

Psychological impact	Psychological state during preparation for procedures (anxiety, excitement)	SM
	Understanding of procedures	SM
	Psychological state in period after treatment (anxiety)	SM
	Treatment impact on socialising / social interactions – confidence to talk to people	SM; SI
	Comparison to peers	CC
	Impact of explaining treatment to other people	SM
	Impact of waiting for treatment	CC
	Reduction in self-confidence / happiness during treatment	SM; SI
Physical impact	Discomfort from treatment	PIR; DCE; CC; SI
	Ability to eat / need to remove brace during eating	PIR; SI
	Impact on speech e.g. lisp	SI
	Ability to play musical instrument (wind or brass)	PIR
	Damage to teeth from treatment (decay, gum problems, injuries from appliances, devitalisation, TMJ problems, root resorption, staining, chipping from brace, allergy, soft tissue damage, nerve damage)	PIR
	Post-operative complications (soreness, swelling, bleeding, bruising, infection, drooling)	PIR; SM
	Appearance during treatment	PIR
	Temporary effect of treatment procedure (e.g. LA/ GA/ sedation)	PIR
Self-care	Change to oral hygiene – e.g. increased time requirement	PIR; SI; CC
	Dental health – ensure disease free prior to treatment, regular attender with dentist	PIR; CC
	Diet modification during treatment	PIR; SI; CC
	Management of pain	PIR
	Attendance for treatment appointments and emergency appointments	PIR; CC
	Compliance with instructions / post-operative care	PIR; SI
	Commitment to long term maintenance – regular dental check-ups, orthodontic retainer wear	PIR
	Change in behaviour to manage risk (e.g. mouthguard for trauma, smoking /alcohol reduction)	PIR, CC
	Strategies for coping with treatment	SM
Impact on family	Commitment to accompany person to appointment	SI
	Support during treatment	SI; SM

Risk during treatment		
Risk	Side-effects during treatment	DCE
	Adverse reaction	DCE
	Severity of side-effects	DCE
	Complexity of procedure/ risk	DCE
Psychosocial effect of treatment outcome		
Well being	Daily happiness / wellbeing	SR; SM; SIS
	Confidence	SR; PIR
	Self-esteem / sense of self / self-perception	PIR; SM
	Self-consciousness / insecurity around other people	SM; SI
	Normalisation compared to peers	SM
Own behaviour	Smiling – confidence to smile, willingness to smile / show teeth when smiling	CC; SM; SI; SR
	Embarrassment in conversations while smiling or laughing	PILs
	Confidence talking to people	SI
	Self-consciousness	SR; PIR
	Willingness to go out or meet family and friends	PIR
	Ability to adapt to condition	SM
	Impact on social interactions – dating, sleepovers, eating in public, missing social events, worried about going to school	SM; CC
Perceived behaviour of others	Bullying	SM
	Stigma related to missing teeth	SM
	Assumptions by others about self-care / dental health	SM; SI
	Treatment by others – being seen as different, effect on socialising/dating	SM: SI
OHRQoL	Quality of life related to OHIP domains (functional limitation, discomfort, disability from oral condition)	SR
	Quality of life based on appearance, function and health	PIR
Perception of tooth replacement	Natural appearance and feel	PIR; SI
	Feel of permanence	PIR; SM; SI
	Perceived characteristics of fixed or removable tooth replacement	PIR; DCE; CC

Effect of treatment outcome on appearance		
Smile attractiveness	“Smile attractiveness” / “smile”	SR; PIR; SM
	Appearance when smiling	CC
Dental appearance	“Dental appearance”	SR
	Tooth shape, colour, space distribution around upper anterior teeth, position symmetry of teeth/midline	SR
	Dental aesthetic parameters - width/height ratio, gingival zenith, apparent contact point dimension	SR
	Gaps / gappy teeth / spaces	SM; SI; CC
	Straightness of teeth	SM
	Symmetry	CC
	Size of teeth	CC
Appearance of specific treatment	White Esthetic Score – tooth form, volume, colour, surface texture and translucency	SR
	Pink Esthetic Score (soft tissues) – presence of papilla, curvature and level of mucosa, soft tissue colour and texture	SR
	Appearance of specific treatment – mucosal discolouration, crown morphology, crown colour match, symmetry/harmony, papilla level	SR; CC; SI; DCE
Effect of treatment outcome on function		
Eating	Eat with confidence / enjoyment	PIR; SI
	Ability to eat / ability to chew/grind / ability to bite	PIR; DCE
	Limitations in what can be eaten	SM; SI
Speech	Lisp	SM
	Improvement in speech – ability to be understood	PIR: SI
Mastication	Mastication OHIP domains - chewing difficulties in general, chewing of different types of food, selection of food, and difficulties in finishing meals due to dental problems)	SR
	Mastication index – normal / able to eat anything but it takes long / able to eat a soft diet only / difficult to eat even a soft diet and it takes long / only liquid diet	SR
	Patient-reported functional impairment (difficulty experienced performing mandibular tasks e.g. chewing, laughing)	SR

	Clinical measures of mastication - occlusal tooth contact, maximum bite force, masticatory performance	SR
Occlusion	Occlusal function (canine or group function, centric relation contact, cross-arch and cross-tooth balancing prematurities, premature protrusive contacts)	SR
	Teeth ‘fit right’	SI
	Change in bite – stop overbite	SI
Limited function	Patient-reported functional impairment	SR
Impact on dental health		
Tooth health	Change in primary teeth (pain; mobility; future restorations required; root resorption; infraocclusion)	SR; SM; CC
	Transplanted teeth (pulp survival; root length)	SR
	Damage to other teeth	CC
Periodontal health	Plaque index (plaque accumulation)	SR
	Periodontal health (probing pocket depth, bleeding on probing, gingival recession, papilla index and mobility)	SR
	Gingival health (Inflammation, colour change, oedema, bleeding on probing, tissue contour and ulceration)	SR
	Irritant Index (presence and severity of irritants other than plaque)	SR
Bone health	Marginal bone levels	SR
	Bone maintenance (ability to place future implant)	PIR
Occlusion	The effect of treatment on position of adjacent teeth	SR
	Tooth position (tooth position, stability, tooth fit, bite)	PIR; CC
	Distribution of biting force – bite more evenly and reduce strain to reduce jaw and joint problems and headaches	PIR
Dental health state	Increase ability to clean teeth / look after teeth	PIR; SI
	Reduce risk of trauma	PIR
	Reduce risk of dental problems (decay, enamel wear, gum problems)	PIR; SI
	Overall dental health	CC
	Health of specific tooth / gum	DCE

Harms		
Generic harms	Complications associated with tooth replacement – gum infected and receding	SM
	Damage to other structures (periodontal, TMJ, nerve)	PIR
	TMD signs and symptoms (Helmiko questionnaire)	SR
	TMD (occlusal stability, speech difficulties, pain on opening, mouth opening, muscle stiffness/fatigue, TMJ crepitus/clicking/locking/pain, pain in jaw muscles, headache, tooth grinding/clenching	SR
Treatment specific harms	Non-permanence (risk of orthodontic relapse)	PIR; CC
	Implant (marginal bone loss around implant; soft tissue breakdown following bone augmentation; increased probing depths; bleeding; gum infection; soreness; loss of implant)	SR; PIR
	Biological complications associated with implant restoration (fistulas, suppuration, bone loss)	SR
	Technical complications associated with implant-supported restorations (chipping of the temporary acrylic crown, loosening of the implant)	SR
	Technical complications associated with implant failure in growing children (relative movement of implant leading to replacement of the restoration or implants)	SR
Long-term		
Psychosocial effect	Worry related to condition or treatment e.g. false tooth falling out while public speaking, forgetting false teeth and being seen publicly without teeth	SM
	Worry related to future implications of treatment e.g. future loss of primary tooth	SM
	Worry related to expectation of breakage / loss of tooth replacement in the future	SI
Survival of treatment	Survival of specific treatment modalities	SR; PIR; CC
Success of treatment	Success of specific treatment - RBBs (restoration unchanged since placement; stability, peripheral seal and morphology preservation; - implants (bleeding, plaque, probing depth, bone loss, marginal bone levels, soft tissue conditions, stability, bone gain from augmentation) - autotransplanted teeth in adolescents (pulp, root and periodontal measures of health)	SR; CC
	Predictability of tooth replacement	CC
	Risk of future breakages / problems	SI

	Failure of transplanted tooth (root fracture, root resorption, periapical and periodontal pathology)	SR
	Failure of tooth replacement / components	SR; PIR; CC
	Proxy markers of success - Bone creation and stability by orthodontic tooth movement for future implant placement - Ability of implant to retain bone in premolar sites - Relapse in root position following orthodontic treatment that may prevent implant placement	SR
Longevity of treatment	Longevity of tooth replacement / treatment components / treatment effect	SM; CC; SI; PIR; DCE
	Replacement of treatment components	PIR
	Longevity of retained primary teeth	PIR; CC
Implications for future treatment	Impact on future treatment need	SM
	Impact on future treatment options	PIR; CC
	Need for maintenance	PIR; CC
	Adaptability of treatment	CC
Long term costs	Cost of maintenance	SR; CC
	Impact of long-term costs on other parts of life e.g. university fees	SI

Appendix Table 15: Summary of decision log for attribute reduction (A=Adolescent; P=Parent; C=Clinician; M=Methodologist)	
Service delivery attributes	
Treatment duration	Total treatment time is most important [A,P,C] Treatment time for individual components of care too similar attributes – only need one attribute 'length of appointment' [A,M] Delays in treatment & accuracy of predicted treatment time important [A,P] but difficult to assign to specific treatment [C,M]
	Include: Overall treatment time, Length of appointment
Appointment schedule	Number & schedule of appointments represent treatment burden [P] Both are similar - disagreement which is easiest to understand & most important [A,P] Appointment schedule incorporates the indirect cost of treatment and the impact of treatment on family [A,P,C,M] Impact of treatment time on school and/or work is a consequence of appointments so use for framing but not as a separate attribute [A,P,C,M]
	Include: Timing of appointment, Frequency of appointment
Time	Waiting time is very important [A,P,C,M], particularly time to wait for initial consultation and treatment start Reason for delay (waiting lists, need to wait for dental development) can help framing [A,P,C] Time to wait at individual appointments is an inconvenience of all types of treatment [C]
	Include: Waiting time to start treatment
Service configuration	Travel time represents familiarity & convenience of services [A,P] Travel distance & parking are similar concept [P,C,M]. Treatment planning is difficult to understand [A] Adherence to care is a result of patient response to care [C] Care setting is a consequence of treatment choice / pathway [C]
	Include: Travel time
Cost	Costs of dental treatment usually within NHS [A,P,C,M] Indirect costs difficult to quantify and related to treatment duration and appointment schedule [P,C,M]
	Include: Indirect costs of attending appointment
Impact of treatment	Impact of treatment on quality of life (psychosocial/ physical/ behaviour) important [A,P] Experience of treatment important but subjective [A,P,C]
	Include: Discomfort during treatment, Effect on eating, Effect on speaking, Effect on hobbies, Effect on social interactions
Risk during treatment	Risk important, particularly type of problem, likelihood, consequence and duration [A,P,C,M] Evidence about adverse reactions to specific treatment is weak [C] Complexity of treatments difficult to understood & not relevant [A]
	Include: Type of problem during treatment, Likelihood of problems, Timescale of problems, Effect of problems

Treatment outcome	
Psycho-social	Effect of treatment outcome on own behaviour or interaction with other people important [A,P,C]. Wellbeing subjective & personal [C,M] Perception of tooth replacement (artificial/natural & temporary/permanent) important [A]
	Include: Self-consciousness, Confidence, Other people's reaction, Feel of teeth
Appearance	Appearance very important [A,P,C] Desire for teeth to look normal important [A,P,C] Straightness, gaps and tooth colour/shape important [A,P] Symmetry and gums less important [A,P]
	Include: Normalisation compared to peers, Gaps and straightness, Colour and shape
Function	Eating and "how the bite feels" most important [A] Bite not important but 'learnt' during treatment [P] Occlusion complex and clinically focused measure [A,P,C] Patient-friendly terms for mastication overlap with eating [A,P,C] Lack of evidence about effect of dental treatment on speech [C]
	Include: Ability to eat, Bite
Dental health	Majority of health descriptors are long-term consequences of treatment - include as long-term risks [A,P,C,M] Change in the risk of dental health problems as a result of treatment are not supported by good evidence [C] Valuation of dental health state not relevant in this context [M]
	None to include
Harms	Possible harms overlap with 'Side-effects/risks of treatment' [C,M] Future harms important but include in 'Long-term risks' [A,P,C,M]
	None to include
Long-term	Treatment success, survival & longevity important but overlap [A,P] Easier to understand in terms of need for repair (success) or replacement (survival) [A,P] The impact of treatment on future treatment need important [A,P] Future cost relating to treatment important [A,P] Long-term risk of temporomandibular disorder (TMD) not supported by evidence [C]. Proxy markers of success complex and irrelevant [A,P] Long-term psychosocial impact of treatment subjective [A,P,C]
	Include: Future treatment need, Future treatment cost

Appendix Table 16: Summary of decision log for attribute selection for pilot using survey scores
(A=Adolescent; P=Parent; C=Clinician; M=Methodologist)

Service delivery attributes	
Appointment schedule & treatment time	Appointment frequency includes inconvenience of treatment, indirect costs and impact of treatment on school and work [A,P] Treatment duration very important [A] Use one attribute to cover both frequency of appointment and duration of treatment for piloting – represents burden [M]
	Include: Treatment burden (Frequency of appointment & overall treatment time)
Access	Waiting time most important - time scales easiest to understand in years or months (not school years) [A,P] Travel time & cost are something ‘you put up with’ [A,P]
	Include: Waiting time
Risk	All risk attribute important [A,P,C,M] Type of problem most important and easiest to understand [A] Likelihood of risk complicated by numeracy issues [A,P] Generic dental examples help understanding and include some indication of effect and timescale of the problem [A,P]
	Include: Types of potential problems during treatment
PREMs	Effect on speech and discomfort most important [A,P]
	Include: Discomfort during treatment
Treatment outcome	
PROMs	Patient-reported outcome measures important [A,P,C] Difficult to include due to subjectivity [A,P,C,M]
	None to include
Appearance	Appearance most important [A,P,C] ‘Teeth look normal’ difficult to define and interpret [A,P] Using the main components of a dental appearance (straightness of teeth, gaps, colour and shape) more objective [A,P]
	Include: Appearance
Function	Bite most important [A] Ability to bite also important but similar concept [A]
	Include: Bite
Long-term	Normal to see dentist so treatment need often misinterpreted [A,P] Easier to think about long-term costs – frame using need for treatment repair [A] and to maintain dental health [P]
	Include: Cost of future dental treatment

Appendix Table 17: Estimate of long-term costs for treatment strategies used in hypodontia

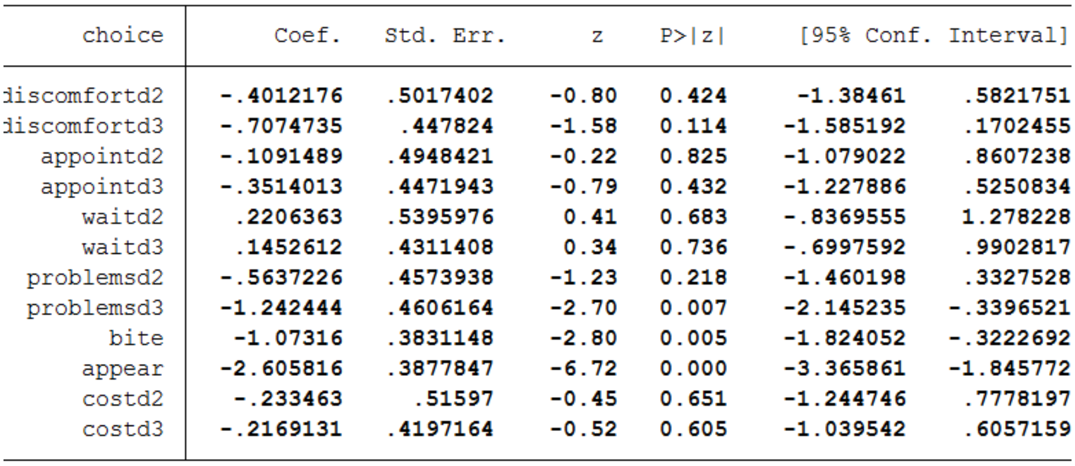
Treatment	Cost to repair / replace	Estimated burden
Orthodontic retainers	Approx. £150	Replace every 3-5 years
Restorative camouflage (white fillings)	NHS charge £59.10	Replace every 5 years (per tooth)
Resin-bonded bridge	NHS charge £256.60	Replace every 8 years
Denture	NHS charge £256.60	Replace every 2-10 years depending on age
Implants	Professional cleaning £50 Crown replacement £500 Implant replacement £2000	2-3 times a year Every 5 years Highly variable

Appendix Table 18: Level balance across the choice tasks in the pilot survey

	Discomfort	Appointment	Wait	Problems	Function	Appearance	Costs
Level 0	24	22	22	23	22	22	23
Level 1	21	21	22	21	22	22	20
Level 2	19	21	20	20	--	--	21

Appendix Table 19: Preference estimates from pilot (conditional logit, dummy coded)

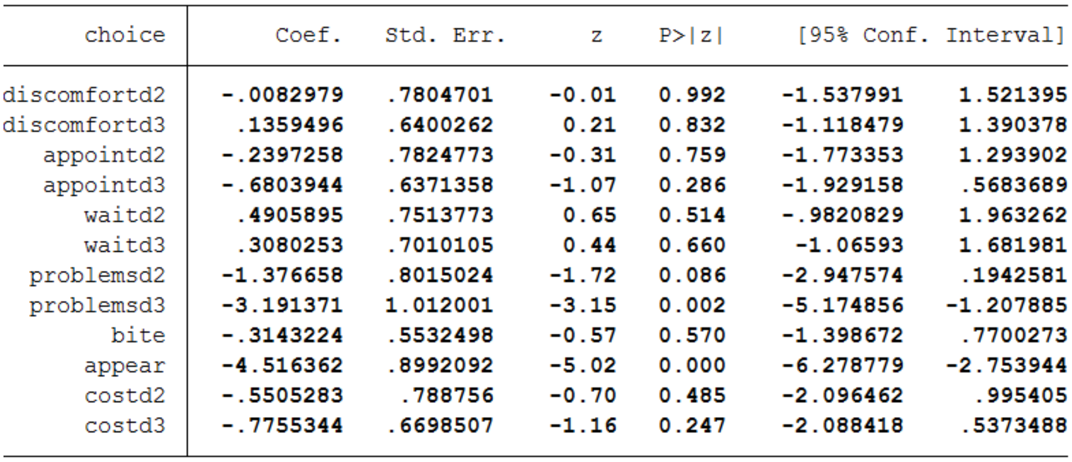
Adolescent only (n=96)



```
      choice       Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]

discomfortd2   -.4012176   .5017402    -0.80   0.424    -1.38461    .5821751
discomfortd3   -.7074735    .447824    -1.58   0.114   -1.585192    .1702455
   appointd2   -.1091489   .4948421    -0.22   0.825   -1.079022    .8607238
   appointd3   -.3514013   .4471943    -0.79   0.432   -1.227886    .5250834
       waitd2    .2206363   .5395976     0.41   0.683   -.8369555    1.278228
       waitd3    .1452612   .4311408     0.34   0.736   -.6997592    .9902817
   problemsd2   -.5637226   .4573938    -1.23   0.218   -1.460198    .3327528
   problemsd3   -1.242444   .4606164    -2.70   0.007   -2.145235   -.3396521
         bite    -1.07316   .3831148    -2.80   0.005   -1.824052   -.3222692
       appear   -2.605816   .3877847    -6.72   0.000   -3.365861   -1.845772
       costd2    -.233463     .51597    -0.45   0.651   -1.244746    .7778197
       costd3   -.2169131   .4197164    -0.52   0.605   -1.039542    .6057159
```

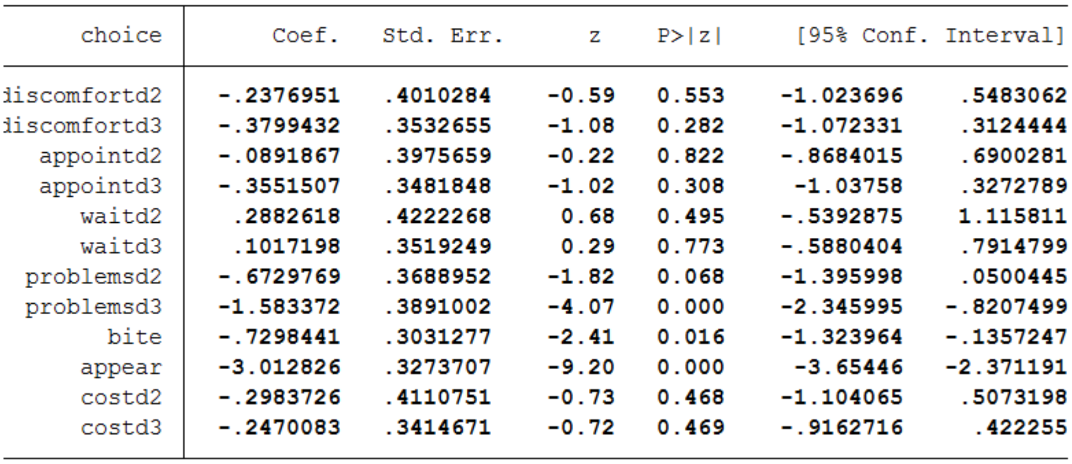
Parent only (n=64)



```
      choice       Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]

discomfortd2   -.0082979   .7804701    -0.01   0.992   -1.537991    1.521395
discomfortd3    .1359496   .6400262     0.21   0.832   -1.118479    1.390378
   appointd2   -.2397258   .7824773    -0.31   0.759   -1.773353    1.293902
   appointd3   -.6803944   .6371358    -1.07   0.286   -1.929158    .5683689
       waitd2    .4905895   .7513773     0.65   0.514   -.9820829    1.963262
       waitd3    .3080253   .7010105     0.44   0.660    -1.06593    1.681981
   problemsd2   -1.376658   .8015024    -1.72   0.086   -2.947574    .1942581
   problemsd3   -3.191371   1.012001    -3.15   0.002   -5.174856   -1.207885
         bite   -.3143224   .5532498    -0.57   0.570   -1.398672    .7700273
       appear   -4.516362   .8992092    -5.02   0.000   -6.278779   -2.753944
       costd2   -.5505283    .788756    -0.70   0.485   -2.096462     .995405
       costd3   -.7755344   .6698507    -1.16   0.247   -2.088418    .5373488
```

Whole sample (n=160)



```
      choice       Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]

discomfortd2   -.2376951   .4010284    -0.59   0.553   -1.023696    .5483062
discomfortd3   -.3799432   .3532655    -1.08   0.282   -1.072331    .3124444
   appointd2   -.0891867   .3975659    -0.22   0.822   -.8684015    .6900281
   appointd3   -.3551507   .3481848    -1.02   0.308    -1.03758    .3272789
       waitd2    .2882618   .4222268     0.68   0.495   -.5392875    1.115811
       waitd3    .1017198   .3519249     0.29   0.773   -.5880404    .7914799
   problemsd2   -.6729769   .3688952    -1.82   0.068   -1.395998    .0500445
   problemsd3   -1.583372   .3891002    -4.07   0.000   -2.345995   -.8207499
         bite   -.7298441   .3031277    -2.41   0.016   -1.323964   -.1357247
       appear   -3.012826   .3273707    -9.20   0.000    -3.65446   -2.371191
       costd2   -.2983726   .4110751    -0.73   0.468   -1.104065    .5073198
       costd3   -.2470083   .3414671    -0.72   0.469   -.9162716     .422255
```

Appendix Table 20: Summary of the features of the pilot and final preference elicitation instrument

	Pilot Preference elicitation instrument	Final Preference elicitation instrument
Decision-making context questions		
Demographics	Age, gender, ethnicity, geographical location, number & location of missing teeth, general & dental health, level of education, household income	Age, gender, ethnicity, geographical location, number and location of missing teeth, level of education
Context	Knowledge, Beliefs about dental treatment, Decision-making, Anxiety, Impact of hypodontia on quality of life, Experience of dental care	Decision-making, Anxiety, Impact of hypodontia on quality of life, Experience of dental care
DCE Choice tasks		
Attributes & levels	7 attributes	6 attributes
	2-3 levels	2-3 levels
Construction of tasks	Full profile with no overlap	Full profile with some overlapping levels for appearance
	Paired alternatives	Paired alternatives
	Inclusion of opt-out after forced choice	Inclusion of opt-out after forced choice
	Generic labelling	Generic labelling
Experimental design	Partial factorial (Full factorial gives 972 profiles)	Partial factorial (Full factorial gives 324 profiles)
	Experimental design – d-optimising efficiency, MNL main effects	Experimental design – d-optimising efficiency, MNL main effects with priors to improve efficiency
	32 tasks as 4 blocks of 8	28 tasks as 4 blocks of 7 (plus one non-experimental task per block to test reliability)
Preference elicitation	Motivation given at start of survey	Motivation given at start of survey and before choice task
	Explanation prior to choice task with practice task	Explanation prior to choice task with non-dental example and annotated instructions for completion
	Choice between pairs	Choice between pairs
	Strength of preference & choice certainty (5-point Likert)	Ease of choice (3-point categorical scale)

Appendix Table 21: Components of the final preference elicitation instrument *(*components that were only used for online survey data collection)*

Introduction	Purpose of the research (motivation for completion) *Information about the survey including information video** Orientation page *Information about eligibility and screening questions** *Consent for participation** *Email address for thank you voucher** *Who is completing survey** *Creation of code to link surveys from within a family**
Respondent demographics	Age, gender, ethnicity, geographical location, number and location of missing teeth, stage of treatment, experience of treatment of person with hypodontia Expected level of education for adolescent/ Parental education
DCE Choice Task	Explanation of the DCE task with non-dental example Description of attributes Annotated example question with instructions for answering Explanation of No Treatment option 7 choice tasks plus one repeat task
Decision-making questions	Decision-making beliefs Anxiety of adolescent (plus parent in parent version) Impact of hypodontia on quality of life of adolescent Actual/planned dental care for hypodontia
Closure	*Consent to be contacted for re-test** Opportunity to provide feedback *Information about completion by other family members** Sources of support and advice

Appendix Table 22: Logistic regression relating respondent characteristics to low decision-making certainty, measured with the SURE tool

	Odds Ratio	95% CI		p value
Parent compared to adolescent	0.97	0.82	1.16	0.766
Dyad compared to adolescent	0.85	0.66	1.1	0.227
Older (15-16 years) compared to younger (12-14 years)	0.41	0.34	0.48	<0.001
Female compared to male	1.18	1.00	1.40	0.05
Moderate hypodontia compared to mild	1.50	1.25	1.79	<0.001
Severe hypodontia compared to mild	1.15	0.93	1.44	0.192
Those in treatment compared to pre-treatment	1.16	0.97	1.38	0.101
Observed survey, compared to online data collection	0.65	0.51	0.83	0.001
Constant	0.60	0.49	0.74	<0.001

Appendix Table 23: Logistic regression relating respondent characteristics to dental anxiety

	Odds Ratio	95% CI		p value
Parent compared to adolescent	1.06	0.89	1.25	0.523
Dyad compared to adolescent	1.34	1.04	1.73	0.025
Older (15-16 years) compared to younger (12-14 years)	0.49	0.42	0.57	<0.001
Female compared to male	0.55	0.47	0.65	<0.001
Moderate hypodontia compared to mild	2.29	1.92	2.73	<0.001
Severe hypodontia compared to mild	2.63	2.12	3.26	<0.001
Those in treatment compared to pre-treatment	0.93	0.78	1.11	0.411
Observed survey, compared to online data collection	0.86	0.69	1.08	0.189
Constant	1.31	1.07	1.60	0.010

Appendix Table 24: Logistic regression relating respondent characteristics to reporting a high quality of life impact from hypodontia

	Odds Ratio	95% CI		p value
Parent compared to adolescent	0.43	0.36	0.55	<0.001
Dyad compared to adolescent	0.55	0.41	0.76	<0.001
Older (15-16 years) compared to younger (12-14 years)	1.58	1.29	1.93	<0.001
Female compared to male	0.47	0.39	0.58	<0.001
Moderate hypodontia compared to mild	1.01	0.80	1.26	0.950
Severe hypodontia compared to mild	3.99	3.15	5.05	<0.001
Those in treatment compared to pre-treatment	0.64	0.52	0.79	<0.001
Observed survey, compared to online data collection	0.23	0.16	0.33	0.001
Constant	0.55	0.44	0.70	<0.001

Appendix Table 25: Comments from online survey relating to experience of hypodontia

Experience of decision-making

It has been a difficult road to navigate the various treatment options, and feel that dentists / orthodontists/max fax surgeons are probably a little unclear about the best treatment options.

Parent of 15-year-old male with moderate hypodontia

My child was first assessed for treatment to sort out a long term solution with regards to her bilateral missing incisors 2 years before returning again to the orthodontist. She was highly encouraged to choose the bridge option which would only come into play once her milk teeth fell out naturally. Neither of us were happy with the option however we took the advice. We however returned after a further referral.

Mother of 13-year-old female with mild hypodontia

It is quite scary entering onto a course of treatment that we have no idea how long it will take to complete, how much discomfort or problems my daughter will experience and what the end result will look like.

Mother of 15-year-old female with moderate hypodontia

Experience of treatment

I have two children with missing teeth. My daughter now 20 years, suffered really badly and never smiled throughout her teenage years. I felt let down by the services offered, we were referred to [redacted] for restoration but they continually cancelled appointments and stated the process would take 3 years or more, offering her false teeth. Her confidence was so low, I had no option to go privately and she had bridges within weeks and it totally changed her life and she hasn't stopped smiling since. There is not enough assistance especially in the South through NHS. I am now worried for my son who has more severe missing teeth. We have been referred to [redacted] and I hope we get a better service from them. It really affects their lives, everyday, dealing with eating difficulties, smiling, chewing, and general confidence in them. It is very upsetting for me as a parent to see my children lack confidence because of this problem which is not their fault.

Mother of 14-year-old male with severe hypodontia

Treatment outcome

My child's treatment was extremely good. Those involved in her care explained the treatment options very well and put my child at ease. The outcome is great and she is very pleased with the outcome.

Mother of 15-year-old female with moderate hypodontia

The treatment of my daughters missing tooth has been very good but the black/grey cement on one of front teeth can be seen and makes her false tooth stand out.

Mother of 14-year-old female with mild hypodontia

Treatment finished. He has a bridge but I'm not totally happy with colour match or the dark colour behind (bridge / which you can see through at the top of new teeth

Mother of 16-year-old male with mild hypodontia

Appendix Table 26: Characteristics of respondents removed from main analysis due to rapid survey completion time suggesting poor quality responses

	Characteristic	Category	Number
Adolescent (n=14)	Age	12 13 14 15 16	2 1 0 2 9
	Gender	Male Female	8 6
	Severity of hypodontia	Mild Moderate Severe	5 5 4
	Stage of treatment	Start Mid	11 3
Parent (n=3)	Relationship	Mother Stepfather	2 1
	Child’s age	15 years 16 years	1 2
	Child’s gender	Female	3
	Child’s severity of hypodontia	Moderate Severe	2 1
	Child’s stage of treatment	Start Mid	1 2
Dyad (n=1)	Mother with 14-year-old daughter Severe hypodontia, at the end of treatment		

Appendix Table 27: Examination of task non attendance

Selection of Treatment A	Number of participants (%)	
Never (always select Treatment B)	1	0.5
1 task	9	4
2 tasks	26	12
3 tasks	61	28
4 tasks	72	33
5 tasks	37	17
6 tasks	9	4
Every task (always select Treatment B)	1	0.5

Appendix Table 28: Preference estimates from the conditional logit (a), ASC conditional logit (b) and mixed logit without correlation (c)

a) Conditional logit

```
-------------+----------------------------------------------------------------
      choice |      Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]
-------------+----------------------------------------------------------------
      Waitd2 |  -.5257993     .09239    -5.69   0.000    -.7068803   -.3447182
      Waitd3 |  -.4896273   .0925479    -5.29   0.000    -.6710179   -.3082366
 Discomfortd2 |  -.0831622    .094437    -0.88   0.379    -.2682554    .1019309
 Discomfortd3 |  -.6893148   .0955192    -7.22   0.000    -.8765289   -.5021006
    TxTimed2 |   -.564205   .0927744    -6.08   0.000    -.7460395   -.3823705
    TxTimed3 |  -.8496339   .0948532    -8.96   0.000    -1.035543    -.663725
  Problemsd2 |  -.7644006   .0980969    -7.79   0.000    -.9566671   -.5721341
  Problemsd3 |  -.9866269   .1064715    -9.27   0.000    -1.195307   -.7779466
        Bite |  -.2715723   .0808188    -3.36   0.001    -.4299742   -.1131704
  Appearance |   -.429944    .112111    -3.83   0.000    -.6496775   -.2102105
------------------------------------------------------------------------------
```

b) ASC Conditional logit

```
-------------+----------------------------------------------------------------
      choice |      Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]
-------------+----------------------------------------------------------------
alt          |
      Waitd2 |  -.2037563   .0816287    -2.50   0.013    -.3637457    -.043767
      Waitd3 |  -.3633635    .078778    -4.61   0.000    -.5177656   -.2089615
 Discomfortd2 |  -.0815636   .0786134    -1.04   0.299     -.235643    .0725158
 Discomfortd3 |  -.6349508   .0871311    -7.29   0.000    -.8057246    -.464177
    TxTimed2 |  -.2760221   .0825548    -3.34   0.001    -.4378265   -.1142176
    TxTimed3 |  -.6279496   .0834614    -7.52   0.000    -.7915309   -.4643682
  Problemsd2 |   -.205376   .1028834    -2.00   0.046    -.4070238   -.0037282
  Problemsd3 |  -.9707654   .1085769    -8.94   0.000    -1.183572   -.7579586
        Bite |  -.4767071   .0798075    -5.97   0.000    -.6331269   -.3202873
  Appearance |  -1.415743   .2133082    -6.64   0.000    -1.833819   -.9976664
-------------+----------------------------------------------------------------
1            |  (base alternative)
-------------+----------------------------------------------------------------
2            |
       _cons |  -.0841734   .0565767    -1.49   0.137    -.1950617     .026715
------------------------------------------------------------------------------
```

c) Mixed logit without correlation

```
-------------+----------------------------------------------------------------
      choice |      Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]
-------------+----------------------------------------------------------------
Mean         |
      Waitd2 |  -.3023937   .1203859    -2.51   0.012    -.5383458   -.0664416
      Waitd3 |  -.5378275   .1134014    -4.74   0.000    -.7600902   -.3155647
 Discomfortd2 |   -.102218    .125376    -0.82   0.415    -.3479505    .1435144
 Discomfortd3 |  -.9959199   .1580501    -6.30   0.000    -1.305692   -.6861474
    TxTimed2 |    -.42286   .1189096    -3.56   0.000    -.6559186   -.1898014
    TxTimed3 |  -.9918055    .142945    -6.94   0.000    -1.271973   -.7116385
  Problemsd2 |  -.2613653   .1369216    -1.91   0.056    -.5297268    .0069961
  Problemsd3 |  -1.378466   .1849352    -7.45   0.000    -1.740933      -1.016
        Bite |  -.6915787   .1242446    -5.57   0.000    -.9350936   -.4480637
  Appearance |  -2.102522   .3197407    -6.58   0.000    -2.729202   -1.475842
-------------+----------------------------------------------------------------
             |
```

Appendix Table 29: Preference weight estimates and heterogeneity (standard deviation) for attribute levels relative to omitted level, estimated using mixed logit

```
─────────────┬────────────────────────────────────────────────────────────────
      choice │      Coef.   Std. Err.      z    P>|z|     [95% Conf. Interval]
─────────────┼────────────────────────────────────────────────────────────────
Mean         │
      Waitd2 │  -.3023937   .1203859    -2.51   0.012    -.5383458   -.0664416
      Waitd3 │  -.5378275   .1134014    -4.74   0.000    -.7600902   -.3155647
Discomfortd2 │   -.102218    .125376    -0.82   0.415    -.3479505    .1435144
Discomfortd3 │  -.9959199   .1580501    -6.30   0.000    -1.305692   -.6861474
    TxTimed2 │    -.42286   .1189096    -3.56   0.000    -.6559186   -.1898014
    TxTimed3 │  -.9918055    .142945    -6.94   0.000    -1.271973   -.7116385
  Problemsd2 │  -.2613653   .1369216    -1.91   0.056    -.5297268    .0069961
  Problemsd3 │  -1.378466   .1849352    -7.45   0.000    -1.740933     -1.016
        Bite │  -.6915787   .1242446    -5.57   0.000    -.9350936   -.4480637
  Appearance │  -2.102522   .3197407    -6.58   0.000    -2.729202   -1.475842
─────────────┼────────────────────────────────────────────────────────────────
SD           │
      Waitd2 │   .1908911   .3678003     0.52   0.604    -.5299842    .9117665
      Waitd3 │   .0456014    .338612     0.13   0.893    -.6180659    .7092686
Discomfortd2 │  -.4850349   .2873249    -1.69   0.091    -1.048181    .0781115
Discomfortd3 │   .9830209   .1817811     5.41   0.000     .6267364    1.339305
    TxTimed2 │  -.2780215   .2384847    -1.17   0.244     -.745443       .1894
    TxTimed3 │   .7443977   .2215531     3.36   0.001     .3101617    1.178634
  Problemsd2 │  -.4732819   .2641025    -1.79   0.073    -.9909133    .0443496
  Problemsd3 │    1.13863   .2085076     5.46   0.000     .7299632    1.547298
        Bite │   .5671572   .1581965     3.59   0.000     .2570977    .8772167
  Appearance │   1.806827   .3671807     4.92   0.000     1.087166    2.526488
─────────────┴────────────────────────────────────────────────────────────────
```

Appendix Table 30: Ratios between specified attribute-levels

		1-year wait (9 months additional wait)		3-year wait (33 months additional wait)		3-year treatment time (34 months additional treatment)		5-year treatment time (58 months additional treatment)	
Suboptimal dental appearance	Ratio (95% CI)	7.0	(1.1 to 12.8)	3.9	(2.0 to 5.8)	4.9	(1.6 to 8.4)	2.1	(1.4 to 2.9)
	Equivalent in months	63	10-115	129	66-191	167	54-286	122	81-168
Unchanged bite	Ratio (95% CI)	2.3	(0.3 to 4.3)	1.3	(0.6 to 2.0)	1.6	(0.5 to 2.3)	0.7	(0.4 to 1.0)
	Equivalent in months	21	3-39	43	20-66	54	17-78	41	23-58
Severe problems	Ratio (95% CI)	4.6	(0.8 to 8.3)	2.6	(1.3 to 3.8)	3.3	(1.2 to 5.3)	1.4	(0.9 to 1.8)
	Equivalent in months	41	7-75	86	43-125	112	41-127	81	52-104
Severe discomfort	Ratio (95% CI)	3.3	(0.4 to 6.1)	1.9	(1.0 to 2.7)	2.4	(0.9 to 3.8)	1.0	(0.6 to 1.4)
	Equivalent in months	30	4-55	63	33-89	82	31-129	58	35-81

Appendix Table 31: Interactions in mixed logit to test pre-specified hypotheses about difference in preferences between groups

		Coefficient	95% CI	p
3-year waiting time	Parent and dyad	-0.72	-1.04 to -0.40	<0.001
	Adolescent (interaction effect)	0.30	-0.09 to 0.68	0.13
	Non-anxious group	-0.55	-0.52 to -0.02	0.034
	Dentally anxious (interaction effect)	-0.07	-0.47 to 0.33	0.736
	Low-moderate quality of life impact from hypodontia	-0.62	-0.90 to -0.35	<0.001
	High quality of life impact from hypodontia (interaction effect)	0.16	-0.33 to 0.66	0.518
Severe discomfort	Parent and dyad	-1.10	-1.50 to -0.70	<0.001
	Adolescent (interaction effect)	0.16	-0.35 to 0.66	0.54
	People aged 15-16 years	-0.90	-1.29 to -0.51	<0.001
	People aged 12-14 years (interaction effect)	-0.24	-0.74 to 0.27	0.361
	People undergoing or completed treatment	-0.93	-0.58 to -1.28	<0.001
	People who have not started treatment (interaction effect)	-0.16	-0.71 to 0.38	0.551
	Non-anxious group	-0.88	-1.31 to -0.46	<0.001
	Dentally anxious (interaction effect)	-0.38	-0.91 to 0.15	0.160
	Low-moderate quality of life impact from hypodontia	-1.29	-1.68 to -0.89	<0.001
	High quality of life impact from hypodontia (interaction effect)	**0.73**	**0.13 to 1.32**	**0.016**
Severe problems	Adolescent and dyad	-1.34	-1.77 to -0.90	<0.001
	Parent (interaction effect)	**-0.63**	**-1.22 to 0.03**	**0.038**
	Non-anxious group	-1.27	-1.72 to -0.83	<0.001
	Dentally anxious (interaction effect)	-0.39	-0.91 to 0.12	0.136
	Low-moderate quality of life impact from hypodontia	-1.55	-1.99 to -1.11	<0.001
	High quality of life impact from hypodontia (interaction effect)	-0.07	-0.79 to 0.65	0.853
5-year treatment time	Low-moderate quality of life impact from hypodontia	-1.01	-1.33 to -0.69	<0.001
	High quality of life impact from hypodontia (interaction effect)	0.09	-0.46 to 0.64	0.755

Suboptimal appearance	People aged 12-14 years	-1.77	-2.50 to -1.06	<0.001
	People aged 15-16 years (interaction effect)	**-0.85**	**-1.60 to -0.09**	**0.029**
	People who have not started treatment	-1.81	-1.01 to -2.60	<0.001
	People undergoing or completed treatment (interaction effect)	**-1.09**	**-0.25 to -1.94**	**0.011**
Bite unchanged	Mild-moderate hypodontia	-0.74	-1.02 to -0.45	<0.001
	Severe hypodontia (interaction effect)	0.14	-0.36 to 0.63	0.587
	People who have not started treatment	-0.75	-0.36 to -1.14	<0.001
	People undergoing or completed treatment (interaction effect)	0.31	-0.39 to 0.45	0.885

Appendix Table 32: Preference estimates and class share for a) three, b) four and c) five latent classes

a)

Variable	Class1	Class2	Class3
Waitd2	-7.540	0.037	-0.259
Waitd3	-5.139	-0.082	-1.115
Discomfortd2	-6.108	0.273	-0.740
Discomfortd3	3.256	-0.177	-2.767
TxTimed2	-6.358	-0.325	-0.910
TxTimed3	-16.017	-0.686	-0.952
Problemsd2	-2.836	0.150	-0.948
Problemsd3	-16.286	0.001	-3.192
Bite	-21.573	-0.428	-0.351
Appearance	-40.483	-1.147	-1.617
Class Share	0.185	0.413	0.401

b)

Variable	Class1	Class2	Class3	Class4
Waitd2	-130.726	-0.461	0.620	-0.168
Waitd3	-70.888	-0.781	0.440	-1.036
Discomfortd2	-46.262	0.114	0.212	-0.572
Discomfortd3	-48.885	-0.679	-0.397	-2.781
TxTimed2	-64.234	-0.154	-0.262	-1.076
TxTimed3	-41.187	-0.420	-0.909	-0.960
Problemsd2	41.219	-0.315	0.043	-1.224
Problemsd3	-84.049	-0.486	0.323	-3.406
Bite	-42.699	-0.246	-1.120	-0.452
Appearance	-92.084	-3.352	-0.376	-1.235
Class Share	0.123	0.341	0.192	0.344

c)

Variable	Class1	Class2	Class3	Class4	Class5
Waitd2	0.021	-0.203	-2.379	-1.936	-0.284
Waitd3	1.288	-0.335	-2.534	-2.006	-1.298
Discomfortd2	-0.227	0.034	3.237	-6.368	-0.705
Discomfortd3	0.795	-0.952	5.015	-1.475	-2.999
TxTimed2	0.592	-0.326	-3.395	-0.970	-1.265
TxTimed3	-1.696	-0.380	-5.527	-6.787	-1.237
Problemsd2	1.089	-0.092	-1.313	-1.053	-1.217
Problemsd3	0.158	-0.899	1.016	-8.509	-3.546
Bite	-0.331	-0.194	-4.392	-14.501	-0.379
Appearance	0.419	-2.775	-6.647	-20.981	-1.080
Class Share	0.091	0.294	0.146	0.134	0.335

Appendix Table 33: Goodness-of-fit statistics for latent class model with different number of classes

Number of classes	**Two**	**Three**	**Four**	**Five**	**Six**
Log likelihood	-860	-829	-818	-802	-803
AIC & BIC	1721	1660	1637	1605	1606

Appendix Table 34: Membership predictors for latent classes

```
Choice model parameters and average class shares

--------------------------------------------------
    Variable |  Class1    Class2    Class3    Class4
-------------+------------------------------------
      Waitd2 |  -1.822    -0.557    -0.016     0.083
      Waitd3 |  -0.914    -1.029     0.041    -1.413
Discomfortd2 |  -1.309    -0.403     0.502    -0.978
Discomfortd3 |  -1.680    -1.577     0.390    -3.442
    TxTimed2 |   0.018    -0.200    -0.324    -1.307
    TxTimed3 |  -0.951    -0.086    -0.938    -1.260
   Problemsd2 |   1.518    -0.705     0.077    -1.194
   Problemsd3 |  -2.358    -1.148     0.348    -3.949
        Bite |  -2.232    -0.113    -0.800    -0.352
  Appearance |  -3.974    -3.498    -1.413    -1.164
-------------+------------------------------------
 Class Share |   0.151     0.235     0.306     0.308
--------------------------------------------------

Class membership model parameters : Class4 = Reference class

--------------------------------------------------
    Variable |  Class1    Class2    Class3    Class4
-------------+------------------------------------
      parent |  -0.783     1.565    -0.579     0.000
  adolescent |   0.535     2.110     0.551     0.000
       older |  -3.918    -0.998    -0.124     0.000
 mildmodhypo |  -1.879    -0.583    -0.302     0.000
     Anxious |  -2.990     0.002    -1.474     0.000
     highQoL |  -0.096    -1.051     0.881     0.000
      Online |   2.602     0.147     2.514     0.000
txexperience |   0.861     1.258     0.097     0.000
       _cons |  -0.417    -2.108    -1.783     0.000
--------------------------------------------------
```

Appendix Table 35: Logistic regression relating respondent characteristics to selection of ‘*No Treatment’*

	Odds Ratio	95% CI		p value
Parent compared to adolescent	1.74	1.40	2.16	<0.001
Dyad compared to adolescent	0.20	0.13	0.33	<0.001
Older (15-16 years) compared to younger (12-14 years)	1.07	0.86	1.32	0.59
Female compared to male	0.59	0.48	0.73	<0.001
Moderate hypodontia compared to mild	0.54	0.43	0.69	<0.001
Severe hypodontia compared to mild	0.31	0.23	0.43	<0.001
Dentally anxious compared to non-dentally anxious	2.26	1.82	2.80	<0.001
Moderate quality of life impact from hypodontia, compared to high impact	0.30	0.22	0.41	<0.001
Low quality of life impact from hypodontia, compared to high impact	0.72	0.56	0.92	0.01
Observed survey, compared to online data collection	0.66	0.47	0.91	0.012
Constant	0.41	0.30	0.55	<0.001

Appendix Table 36: Latent class with those who always opt-out (non-consumers) and never opt-out included as a predictor of class membership

```
Choice model parameters and average class shares

     Variable   Class1    Class2    Class3    Class4

       Waitd2   -0.508    -2.152     0.482    -0.038
       Waitd3   -0.057    -2.834     0.484    -0.998
  Discomfortd2  -0.766     1.730     0.861    -0.696
  Discomfortd3  -0.568    -1.145     0.196    -2.653
      TxTimed2   0.096    -2.000    -0.696    -0.613
      TxTimed3  -0.768    -2.423    -1.143    -0.696
    Problemsd2   0.624    -0.322    -0.285    -0.899
    Problemsd3  -0.593     1.221     0.048    -3.002
          Bite  -1.286     0.987    -1.035    -0.230
    Appearance  -1.499    -4.134    -2.462    -1.295

   Class Share   0.203     0.177     0.227     0.393

Class membership model parameters : Class4 = Reference class

      Variable   Class1    Class2    Class3    Class4

  Never_optout   0.582    -0.199     2.401     0.000
  Always_opt~t   1.168   -15.199   -13.442     0.000
         _cons  -1.074    -0.635    -2.260     0.000
```

Appendix Table 37: Interaction effect to test effect of selecting ‘*No Treatment’* on preferences

		Coeff.	95% CI	p
3-year wait	Treatment selected	-0.54	-0.28 to -0.81	<0.001
	No Treatment selected (interaction effect)	-0.15	-0.66 to 0.36	0.563
Severe discomfort	Treatment selected	-1.13	-0.7 to -1.48	<0.001
	No Treatment selected (interaction effect)	0.25	-0.34 to 0.84	0.406
5-year treatment time	Treatment selected	-1.08	-0.76 to -1.40	<0.001
	No Treatment selected (interaction effect)	0.47	-0.07 to 1.02	0.090
Severe problems	Treatment selected	-1.38	-1.77 to -0.98	<0.001
	No Treatment selected (interaction effect)	-0.52	-1.22 to 0.17	0.141
Unchanged bite	Treatment selected	-0.85	-0.56 to -1.14	<0.001
	No Treatment selected (interaction effect)	0.32	-0.17 to 0.80	0.196
Suboptimal appearance	Treatment selected	-2.48	-1.75 to -3.21	<0.001
	No Treatment selected (interaction effect)	0.81	-0.14 to 1.77	0.093

Appendix Table 38: Logistic regression relating respondent characteristics to rating task as ‘*Difficult*’

	Odds Ratio	95% CI		p value
Parent compared to adolescent	1.20	1.00	1.43	0.048
Dyad compared to adolescent	1.04	0.80	1.36	0.735
Older (15-16 years) compared to younger (12-14 years)	1.07	0.91	1.27	0.402
Female compared to male	1.01	0.85	1.20	0.935
Moderate hypodontia compared to mild	0.95	0.79	1.14	0.582
Severe hypodontia compared to mild	1.09	0.87	1.36	0.455
Dentally anxious compared to non-dentally anxious	1.84	1.55	2.18	<0.001
Moderate quality of life impact from hypodontia, compared to high impact	0.93	0.74	1.18	0.553
Low quality of life impact from hypodontia, compared to high impact	1.11	0.90	1.38	0.323
Observed survey, compared to online data collection	1.22	0.96	1.54	0.098
Constant	0.27	0.20	0.34	<0.001

Appendix Table 39: Latent class with *'Difficult'* tasks included as a predictor of class membership

```
Choice model parameters and average class shares

─────────────┬──────────────────────────────────────────
    Variable │    Class1    Class2    Class3    Class4
─────────────┼──────────────────────────────────────────
      Waitd2 │     2.220    -0.512     1.141    -0.269
      Waitd3 │    -4.194    -0.071    -1.100    -1.050
Discomfortd2 │     0.006     0.041    -1.575    -0.492
Discomfortd3 │    -5.723    -0.040    -1.930    -2.225
     TxTimed2│    -8.496    -0.699     0.944    -0.561
     TxTimed3│    -0.145    -1.490     2.360    -0.930
  Problemsd2 │     0.330     0.099    -1.158    -0.636
  Problemsd3 │    -3.798    -0.346     0.586    -2.860
        Bite │   -10.631    -0.807    -0.434    -0.150
  Appearance │     2.268    -2.947    -0.935    -1.452
─────────────┼──────────────────────────────────────────
 Class Share │     0.086     0.416     0.088     0.410
─────────────┴──────────────────────────────────────────

Class membership model parameters : Class4 = Reference class
─────────────┬──────────────────────────────────────────
    Variable │    Class1    Class2    Class3    Class4
─────────────┼──────────────────────────────────────────
Diff_respond │     2.209   -14.188     0.879     0.000
       _cons │    -1.883     0.063    -1.600     0.000
```

Appendix Table 40: Interaction effect to test effect of data collection mode on preferences

		Coeff.	95% CI	p
3-year wait	Observed survey	-0.80	-0.25 to -01.36	0.005
	Online survey (interaction effect)	-0.23	-0.35 to 0.82	0.425
Severe discomfort	Observed survey	-1.37	-0.60 to -2.13	<0.001
	Online survey (interaction effect)	0.33	-0.44 to 1.11	0.395
5-year treatment time	Observed survey	-0.78	-0.12 to -1.43	0.020
	Online survey (interaction effect)	-0.24	-0.91 to 0.43	0.480
Severe problems	Observed survey	-2.21	-3.04 to -1.38	<0.001
	Online survey (interaction effect)	**0.86**	**0.66 to 1.65**	**0.034**
Unchanged bite	Observed survey	-0.24	-0.72 to 0.23	0.315
	Online survey (interaction effect)	**-0.75**	**-1.27 to -0.24**	**0.004**
Suboptimal appearance	Observed survey	-2.43	-1.22 to -3.63	<0.001
	Online survey (interaction effect)	0.22	-0.89 to 1.33	0.697

Appendix Table 41: Sample characteristics for repeat survey with comparison to baseline sample using Chi-squared

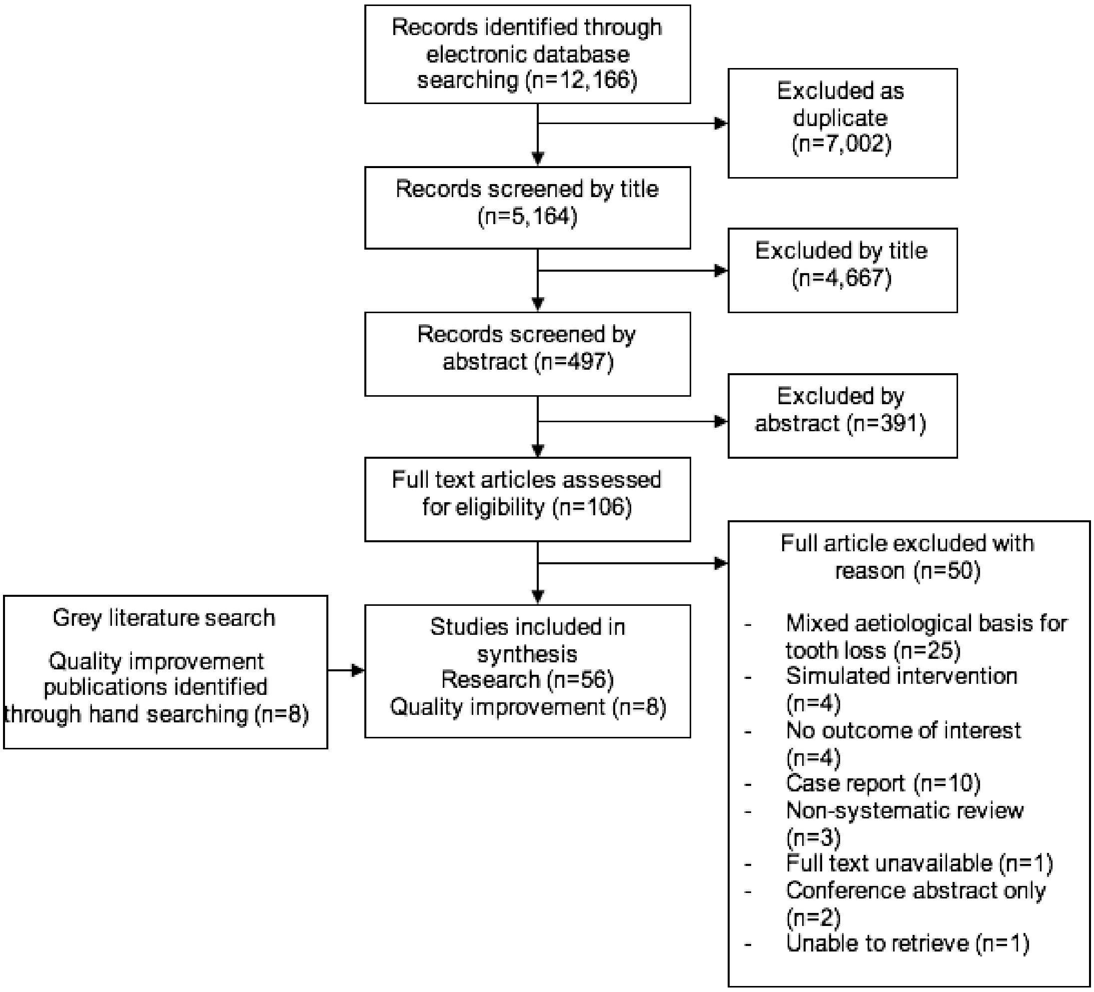
		Repeat sample		Baseline survey		Difference	
		n	%	n	%	%	95% CI
Respondent group	**Adolescent**	11	48	122	60	12	-8 to 32%
	Parent	9	39	56	27	12	-6 to 33%
	Joint	3	13	26	13	0	-19 to 10%
Age of adolescent with hypodontia	**12 years**	2	9	27	13	4	-15 to 12%
	13 years	2	9	37	18	9	-10 to 18%
	14 years	4	17	40	20	3	-17 to 15%
	15 years	9	39	54	26	13	-5 to 34%
	16 years	6	26	46	23	3	-12 to 24%
Gender	**Female**	14	61	128	64	3	-15 to 24%
	Male	8	35	71	35	0	-21 to 17%
	Other	1	4	2	1	3	-1 to 20%
Severity of hypodontia	**Mild**	11	48	90	45	3	-17 to 23%
	Moderate	6	26	68	34	8	-13 to 23%
	Severe	6	26	41	21	5	-10 to 26%
Stage of treatment	**Start**	6	26	67	33	7	-14 to 22%
	Mid	16	70	125	61	9	-12 to 25%
	End	1	4	12	6	2	-15 to 7%

Appendix Table 42: Planned treatment, as reported by the adolescent, parent and clinical notes, for the 15 dyads in the observed survey

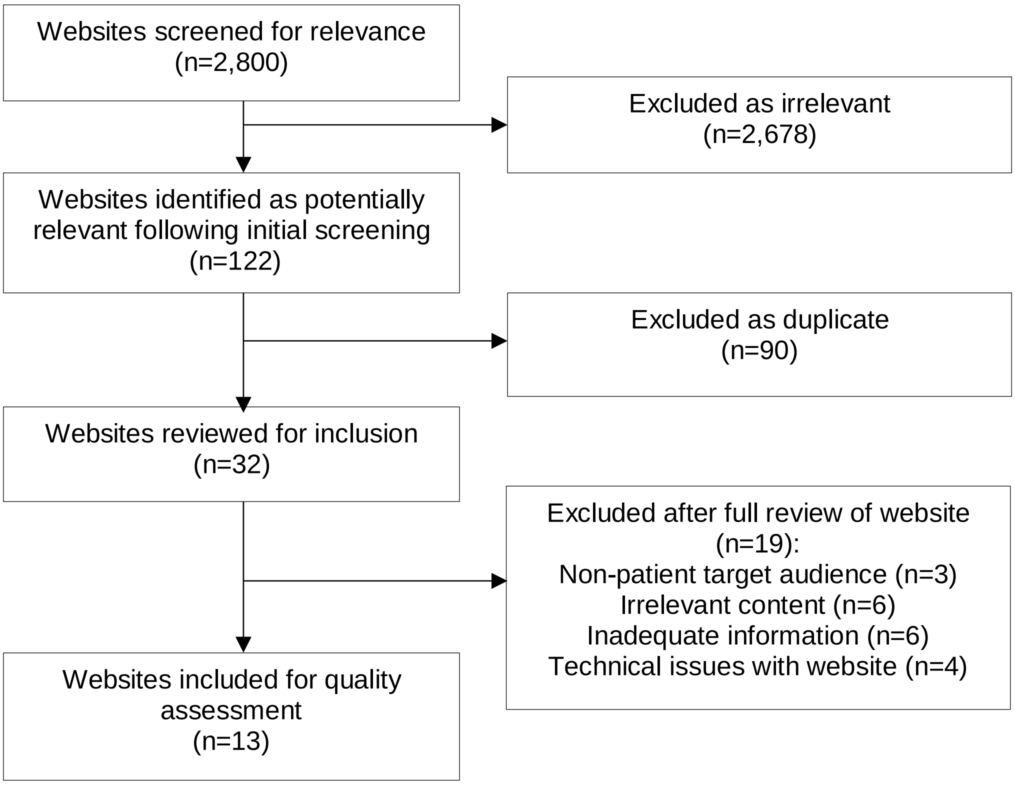
Dyad	Summary of adolescent with hypodontia	Reported by adolescent	Reported by parent	Extracted from clinical notes
1	16-year-old male, severe hypodontia, pre-treatment	RBB & implant	No treatment	No treatment
2	14-year-old female, moderate hypodontia, mid-treatment	Ortho. & RBB	Ortho. & RBB	Ortho. & RBB
3	16-year-old female, mild hypodontia, mid-treatment	Ortho.	Ortho. Unsure about RBB	Ortho. Maintain primary teeth but RBB in future if required
4	14-year-old female, mild hypodontia, mid-treatment	Ortho. Unsure about tooth replacement	Unsure about all options	Ortho. Tooth replacement to be decided
5	14-year-old female, moderate hypodontia, pre-treatment	Ortho.	Ortho.	Ortho.
6	15-year-old male, mild hypodontia, end of treatment	Unsure about all options	RBB	Ortho. (now complete) RBB
7	15-year-old male, severe hypodontia, pre-treatment	Ortho. & RBB	Ortho. & RBB	Ortho & RBB +/- implant
8	14-year-old female, severe hypodontia, mid-treatment	Restorative & ortho.	Restorative & ortho.	Restorative & ortho. +/- tooth replacement in future
9	14-year-old female, moderate hypodontia, mid-treatment	Ortho. & RBB & implant Unsure about restorative	Ortho. Unsure about RBB	Ortho. Preferred tooth replacement to be decided
10	15-year-old female, mild hypodontia, mid-treatment	Ortho. & implant	Unsure about implant	Ortho. Tooth replacement to be decided
11	14-year-old female, severe hypodontia, pre-treatment	Ortho. & RBB & implant Unsure about restorative	Unsure about all options	Ortho. & RBB Maintain primary teeth but RBB/implant in future if required

12	15-year-old female, severe hypodontia, mid-treatment	Ortho. & RBB Unsure about restorative	Ortho. & RBB	Ortho. & RBB Maintain primary teeth but RBB/implant in future if required
13	15-year-old male, severe hypodontia, pre-treatment	Unsure about all options	Unsure about all options	Ortho. Tooth replacement to be decided
14	14-year-old female, moderate hypodontia, pre-treatment	Ortho. & RBB	Ortho.	Ortho. & RBB
15	16-year-old female, mild hypodontia, pre-treatment	Unsure about all options	Restorative Unsure about ortho. & implants	Restorative camouflage Ortho. to be decided

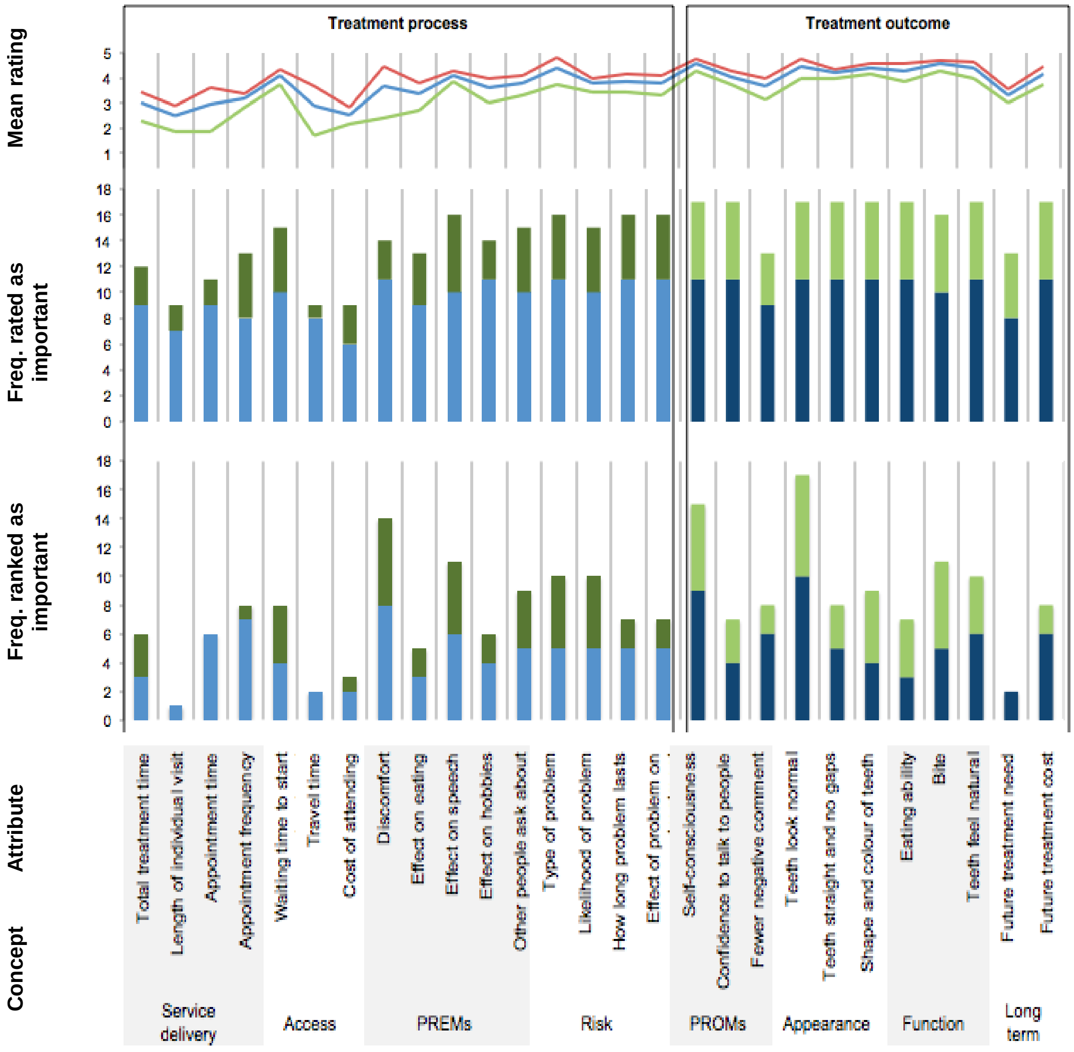
Appendix Figure 1: Flow diagram to show the record selection process



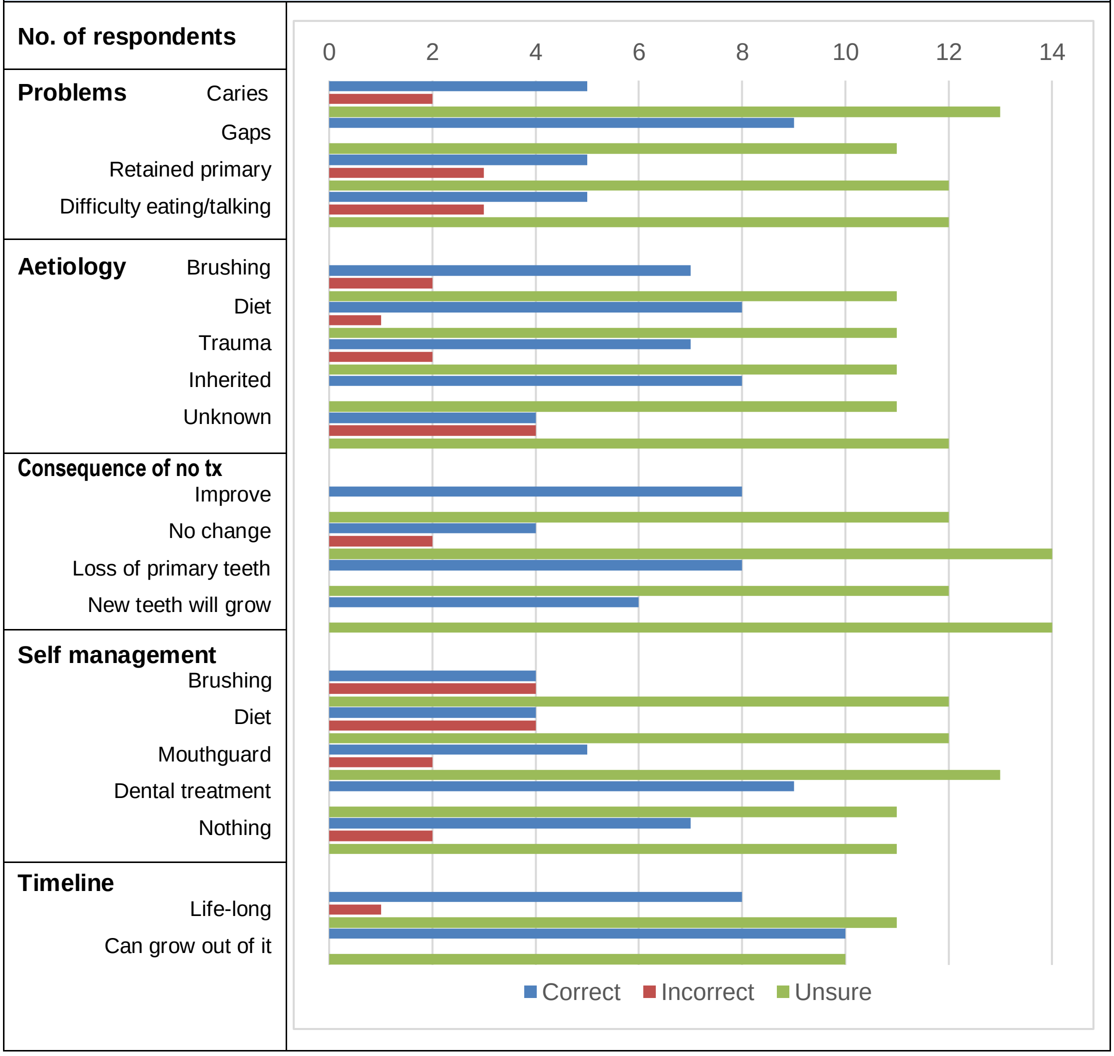
Appendix Figure 2: Selection process for online resources



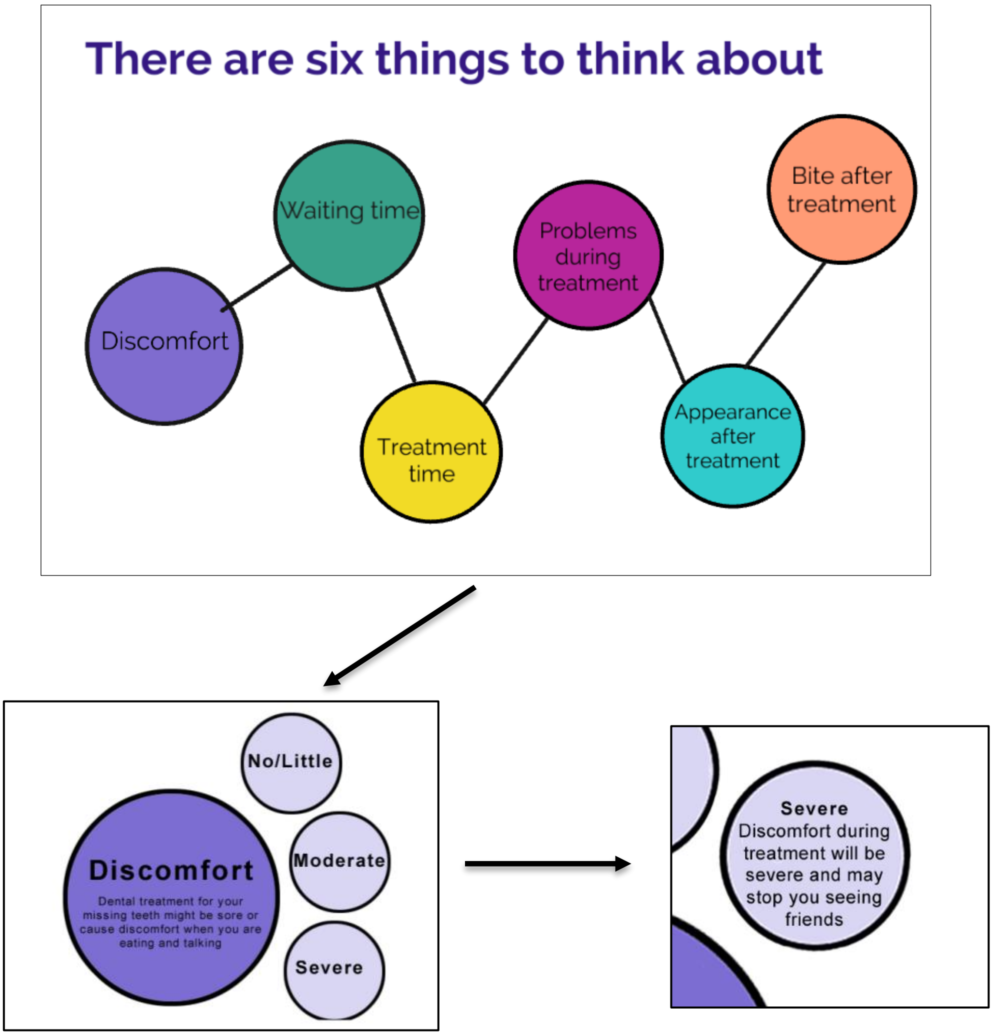
Appendix Figure 3: Aggregate results from attribute rating survey (Blue: Parent (n=11) Green: Adolescent (n=7) Red: All (n=18))



Appendix Figure 4: Answers given to the knowledge questions by the pilot respondents



Appendix Figure 5: Example of interactive animation to introduce attributes and levels



There are six things to think about

Discomfort

Waiting time

Treatment time

Problems during treatment

Appearance after treatment

Bite after treatment

Discomfort

Dental treatment for your missing teeth might be sore or cause discomfort when you are eating and talking

No/Little

Moderate

Severe

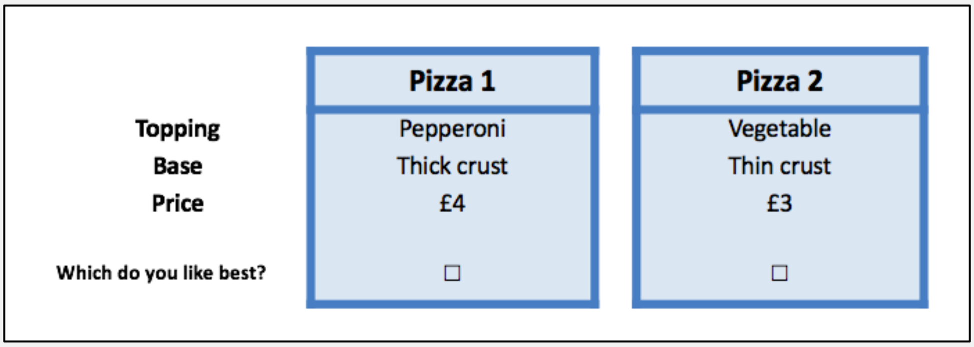
Severe

Discomfort during treatment will be severe and may stop you seeing friends

Appendix Figure 6: Explanation of Choice tasks

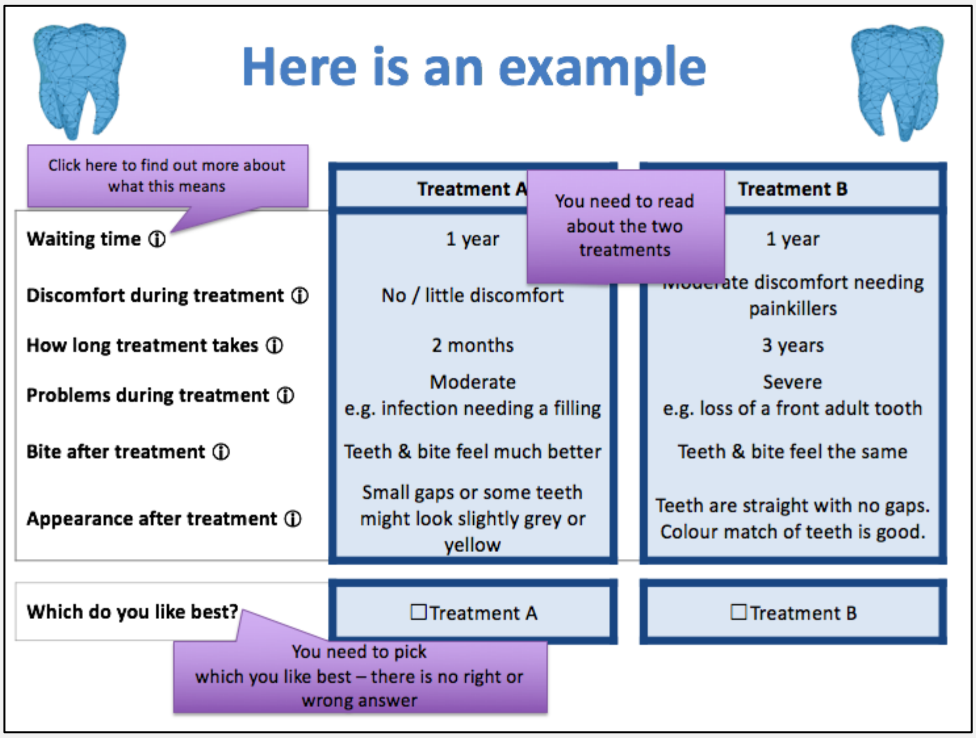
1. Use of a non-dental example to introduce the concept of trade-offs (a)
2. Introduction of the attributes and levels
3. An annotated example of how to answer the question (b)
4. Information boxes for each attribute and attribute-level that could be clicked on to see a full explanation
5. Reassurance that there is no right or wrong answer and acknowledgement that sometimes the choice might be difficult

a)



	Pizza 1	Pizza 2
Topping	Pepperoni	Vegetable
Base	Thick crust	Thin crust
Price	£4	£3
Which do you like best?	☐	☐

b)



Here is an example

Click here to find out more about what this means

	Treatment A	Treatment B
Waiting time ⓘ	1 year	1 year
Discomfort during treatment ⓘ	No / little discomfort	Moderate discomfort needing painkillers
How long treatment takes ⓘ	2 months	3 years
Problems during treatment ⓘ	Moderate e.g. infection needing a filling	Severe e.g. loss of a front adult tooth
Bite after treatment ⓘ	Teeth & bite feel much better	Teeth & bite feel the same
Appearance after treatment ⓘ	Small gaps or some teeth might look slightly grey or yellow	Teeth are straight with no gaps. Colour match of teeth is good.
Which do you like best?	☐Treatment A	☐Treatment B

You need to read about the two treatments

You need to pick which you like best – there is no right or wrong answer

Appendix Figure 7: Participant Identification Centres and geographical area covered

Arrow Park Hospital, Wirral
Birmingham Dental Hospital
Bristol Dental Hospital
Burnley & Blackburn Hospitals
Cardiff Dental Hospital
Chesterfield Royal Hospital
Countess of Chester
Dorset County Hospital
Liverpool Dental Hospital
Montagu Hospital, Mexborough
Musgrove Park & Yeovil
Pinderfields Hospital, Wakefield
Royal Alexandra Children’s Hospital, Brighton
Royal Bournemouth Hospital
Royal Derby Hospital
Salisbury Hospital
Stoke Mandeville Hospital
Wythenshawe Hospital, Manchester

Appendix Figure 8: Preference elicitation survey (adolescent version)

Part One - Information and Consent

1.1 Study Information

This survey is for people with hypodontia and their families. Hypodontia is a dental condition that stops tooth from developing. This causes teeth to be missing.

We would like to know what you think is important about your dental treatment for your missing teeth. This survey is part of a research study at the University of Leeds. The results will be used to help make dental care better for people with missing teeth in the future.

If you live in the UK and are:

- 12-16 years old with naturally missing teeth (hypodontia)

OR

- A parent / guardian of someone with missing teeth

and would be happy to answer some questions please continue.

If you do not have missing teeth this survey will not be relevant to you, but thank you for taking the time to check.

The survey will take approximately half an hour to complete. A £5 e-voucher for Amazon will be given to each person who completes the survey.

If you would like more information, please watch this video about the research.

****LINK TO VIDEO****

1.2 Screening questions (online version only)

1. Are your teeth / your child's teeth developmentally missing (hypodontia)?

Yes	Continue
No	Exit point 1: Thank you for reading about my research. The answers you have provided mean you are not eligible to complete the survey at this time.

2. Do you live in the UK?

Yes	Continue
No	Exit point 1: Thank you for reading about my research. The answers you have provided mean you are not eligible to complete the survey at this time.

1.3 Consent (online version only)

The survey will take about half an hour to complete. It asks you what is important to you when you are thinking about dental treatment for your hypodontia.

Important information:

- It is up to you if you want to take part
- If you are under 16, you need your parent/guardian's consent
- Your answers will not be linked to your name or email address. No-one except the research team will see your answers.
- You can stop answering questions at any time. Any answers that you have given may be used.
- You need to answer all questions to complete the survey and receive the £5 e-voucher
- There are no right or wrong answers
- You can only answer the survey once but other people in your family can also answer it.

Do you want to take part in this survey?

Yes	Continue
No	Exit point 2: Thank you for taking the time to read the information. You have indicated that you do not wish to complete the survey at this time. You can now exit the survey and no answers will be recorded for you.

If you are 16 years old or younger and answering on your own, is your parent / guardian happy for you to take part in this survey?

Yes	Continue
Not applicable	Continue
I am not sure	Please ask your parent / guardian for their permission to complete the survey.
No	Exit point 3: Thank you for taking the time to read the information. You have indicated that your parent/guardian does not want you to complete the survey at this time. You can now exit the survey and no answers will be recorded for you.

I have asked for your email address so I can send your £5 shopping voucher to you. I will not use your email address for any other reason and I will not share it with anyone else. Please write your email address below.

Free text box

1.4 Version of survey

I am...

Aged 12-16 years old and I have missing teeth (hypodontia). I am completing this survey on my own.	Continue to child version
Aged 12-16 years old and I have missing teeth (hypodontia). I am completing this survey with my parent / guardian.	Continue to child version
A parent or guardian of someone with missing teeth. I am completing this survey on my own.	Continue to parent version
A parent or guardian of someone with missing teeth. I am completing this survey with my child.	Continue to parent version

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Part Two - Demographic information

2.1	How old are you (in years)?	Free text	
2.2	How would you describe your gender?	☐ Male ☐ Female ☐ Other ☐ I don’t want to say	
2.3	Where do you live?	☐ North England ☐ Midlands ☐ South England	☐ Scotland ☐ Wales ☐ Northern Ireland
2.4	How would you describe your ethnic group?	☐ White ☐ White Mixed ☐ Asian / Asian British	☐ Black / Black British ☐ Chinese ☐ Other Ethnic group
2.5	How many adult teeth are you missing? (This does not include wisdom teeth)	☐ 1-2 ☐ 3-6 ☐ More than 6 ☐ I’m not sure	
2.6	Which teeth are you missing? *(Please tick all that apply)*	☐ Top teeth ☐ Bottom teeth ☐ Front of the mouth ☐ Back of the mouth ☐ I’m not sure	
2.7	What are you planning to do when you finish school?	☐ Find a job ☐ Apprenticeship ☐ A levels / College ☐ University	

Part Three - DCE choice tasks

3.1 Introduction to DCE tasks

We would like to know what you think is important about dental treatment.

Next are some questions where we ask you to pick from two different options (Treatment A or Treatment B). These are not real dental treatments.

There are no right wrong answers.

An example (using pizza!) is given on the next page.

Pretend we want to find out what type of pizza people like.

- Two made-up pizzas are given (Pizza 1 and Pizza 2)
- For each pizza there are three features we are interested in: topping, base and price
- People answering the survey pick which they like best by thinking about the three features
- There is no right or wrong answer

	Pizza 1	**Pizza 2**
Topping	Pepperoni	Vegetable
Base	Thick crust	Thin crust
Price	£4	£3
Which do you like best?	☐	☐

In our survey we are asking about imaginary dental treatments for missing teeth.

There are six features in each option to think about:

- Waiting time
- Discomfort during treatment
- How long treatment takes in total
- Problems that you might have during your dental treatment
- How your teeth feel and bite after treatment
- The appearance of your teeth after treatment

***** Interactive animation to introduce attributes and levels *****

How to answer the questions

1. Read through the two options
2. Compare the options using the six features. If you want more information about what something means you can click on the ⓘ
3. Pick which option you like best
4. Please tell us know how easy it was to pick
5. After this, if you don't like either option, you can pick No Treatment. This means you would prefer to have NO dental treatment at all and keep your teeth how they are naturally

***** Annotated example *****

3.2 Option for No Treatment

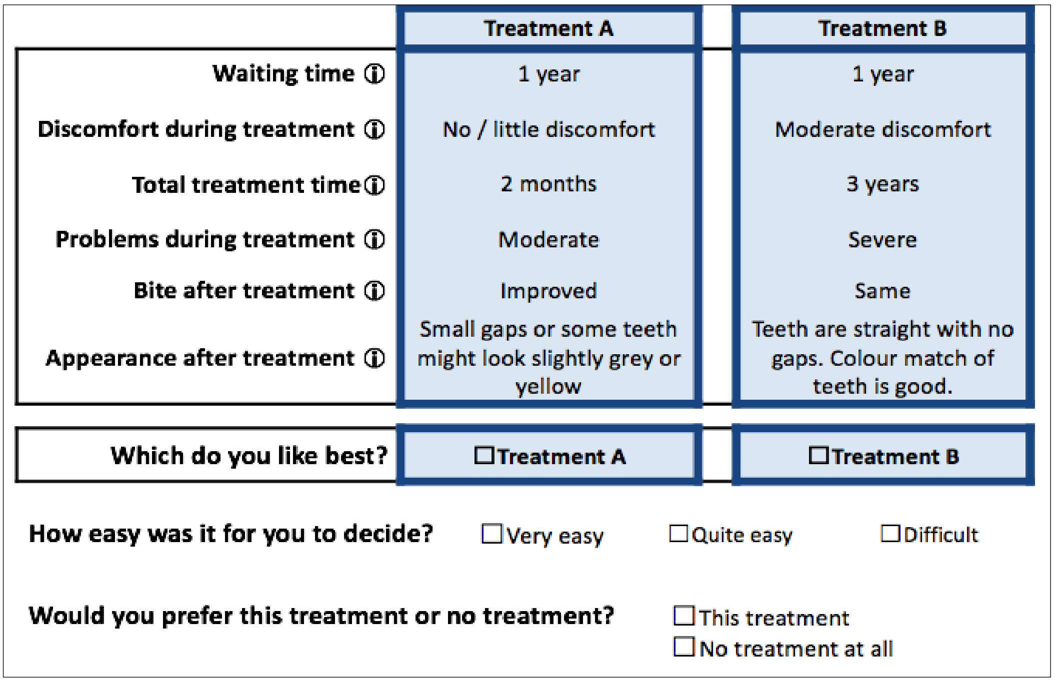
You will also be asked if you like the treatment you picked or if you would prefer no treatment at all.

This would mean your teeth would stay how they are naturally. The dentist would not do anything to change your teeth.

3.3 Choice tasks

***** Seven choice tasks plus one repeat *****

Example task



	Treatment A	**Treatment B**
Waiting time ⓘ	1 year	1 year
Discomfort during treatment ⓘ	No / little discomfort	Moderate discomfort
Total treatment time ⓘ	2 months	3 years
Problems during treatment ⓘ	Moderate	Severe
Bite after treatment ⓘ	Improved	Same
Appearance after treatment ⓘ	Small gaps or some teeth might look slightly grey or yellow	Teeth are straight with no gaps. Colour match of teeth is good.
Which do you like best?	☐**Treatment A**	☐**Treatment B**

How easy was it for you to decide? ☐Very easy ☐Quite easy ☐Difficult

Would you prefer this treatment or no treatment?
☐This treatment
☐No treatment at all

Part Four - Background Information

4.1 Beliefs about dental treatment

We would like to know what you think about dental treatment for your missing teeth.

Please mark whether you agree with the statements below.

	Yes	**No**
Dental treatment is the only option for my missing teeth	☐	☐
Having dental treatment now for missing teeth will reduce my need for more dental treatment in the future	☐	☐
I worry about the risks of dental treatment for missing teeth	☐	☐
Dental treatment for missing teeth can cause new dental problems	☐	☐

4.3 Making choices about dental treatment

We would like you to tell us about making decisions about dental treatment for your missing teeth.

Please answer the questions below.	Yes	No
The following people help me choose treatment for my missing teeth		
Parents / guardian and family	☐	☐
Dentist	☐	☐
Friends	☐	☐
Other people with missing teeth	☐	☐
I feel sure about the best choice for me	☐	☐
I know the benefits and risks of each option	☐	☐
I am clear about which benefits and risks matter the most to me	☐	☐
I have enough support and advice to make a choice	☐	☐

4.4 How you feel about dental treatment

We would like to know how you feel about dental treatment.

Please answer the questions below.	Strongly	Agree	Neither	Disagree	Strongly disagree
I feel anxious about having any dental treatment with my normal dentist					
I feel anxious about having dental treatment for my missing teeth					

4.5 How you feel about your missing teeth

Please mark how much you think your teeth affect you day to day based on:

	A lot	A bit	Not at all
Confidence to smile			
Ability to chew hard food			
Confidence to eat in front of other people			
Ability to speak – for example without a lisp			
Confidence to talk in front of other people			
Other people’s reaction to his/her missing teeth			
Ability to cleaning his/her teeth			
Worry about his/her teeth in the future			

4.6 Your dental treatment

Please can you tell us about your dental treatment for your missing teeth?

Have you started any dental treatment for your missing teeth?	☐ Yes ☐ No ☐ Not sure
Do you still need to agree what type of dental treatment you would like?	☐ Yes ☐ No ☐ Not sure

What type of treatment have you already had for your missing teeth?
Please tick all that apply

☐ Fillings in baby teeth
☐ Extraction of baby teeth
☐ Fillings in permanent teeth
☐ Fixed braces
☐ Removable braces
☐ False teeth that I can take out (a plate or denture)
☐ False teeth glued to other teeth (bridge)
☐ Metal screw placed in the jaw to hold a false tooth (implant)
☐ Retainers
☐ Other (please specify)

In the future, are you planning to have the following treatment?

White fillings to change the shape or size the teeth. This is without any brace treatment first.	☐ Yes	☐ No	☐ Not sure
Braces to close spaces where teeth missing.	☐ Yes	☐ No	☐ Not sure
Braces make the spaces the right size for a bridge (false teeth glued to other teeth).	☐ Yes	☐ No	☐ Not sure
Braces make the spaces the right size for implants (metal screw placed in the jaw to hold a false tooth).	☐ Yes	☐ No	☐ Not sure

Part Six - End of Survey

Thank you for taking part in this survey. You will receive your £5 e-voucher for Amazon in the next few days via email. All your answers will be kept confidential and your email will not be shared with anyone else.

If your parent/guardian has not yet completed this survey, please could you invite them to look at it. We would be very grateful for their opinion too.

Sources of support

If this survey has raised any issues that you would to talk about, Childline offers free advice and support to adolescents for all areas of concern. You can contact Childline free on 0800 1111.

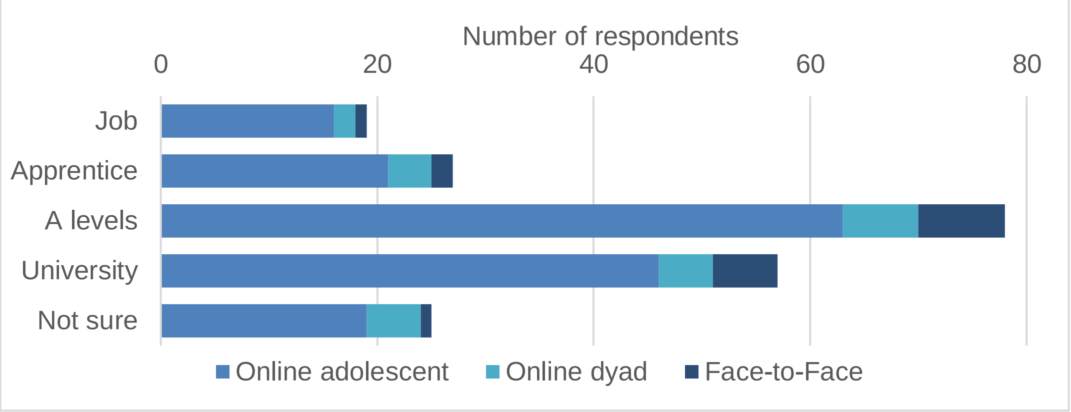
If you have any concerns or questions about missing teeth or this research you can contact Sophy Barber.

sophybarber@nhs.net

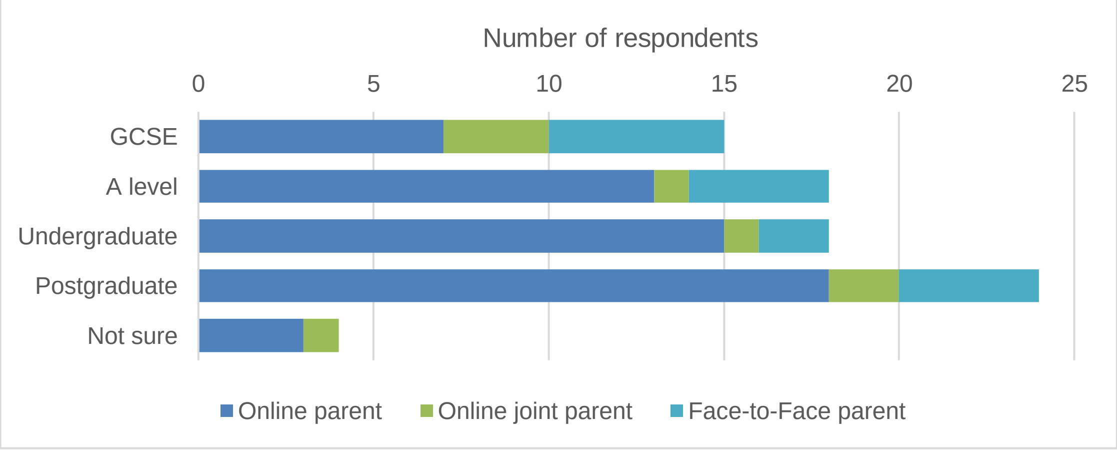
Sophy is part of the research team and also has experience of treating people with dental problems. She can provide general advice about any issues that arise from the survey and signpost to further sources of information and support. She is not able to give specific advice relating to your individual treatment or care – for this you are advised to contact your own dentist

Appendix Figure 9: Parent and adolescent self-report actual or planned level of education

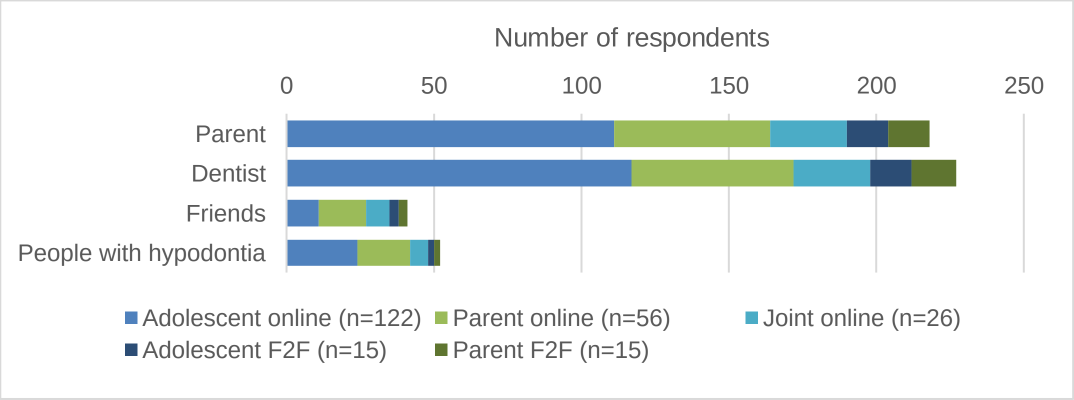
a) Self-reported expected level of education for adolescent with hypodontia



b) Parents’ self-reported level of education

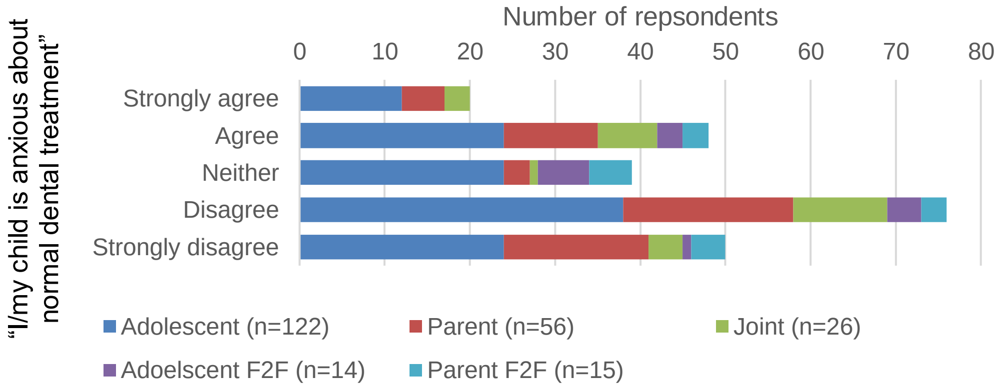


Appendix Figure 10: People who help with decision-making for hypodontia

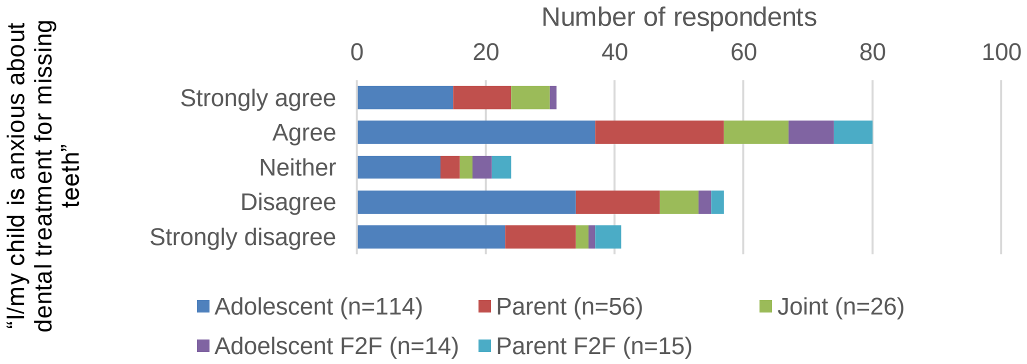


Appendix Figure 11: Adolescent and parent ratings for dental anxiety

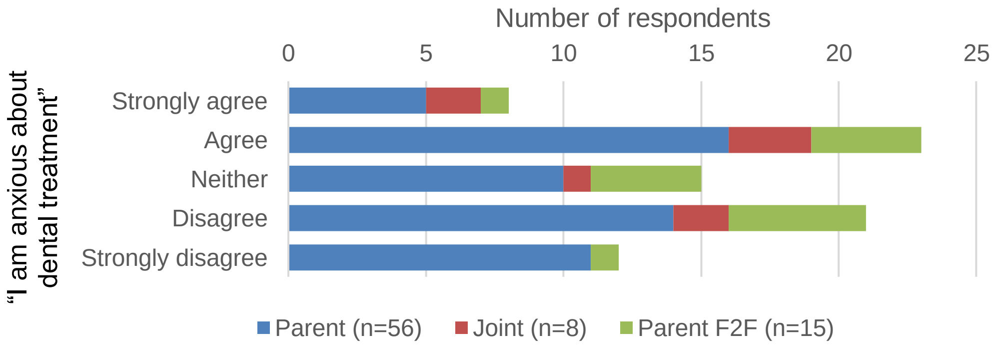
a) **Adolescent anxiety about normal dental treatment with own dentist, as reported by adolescents and parents**



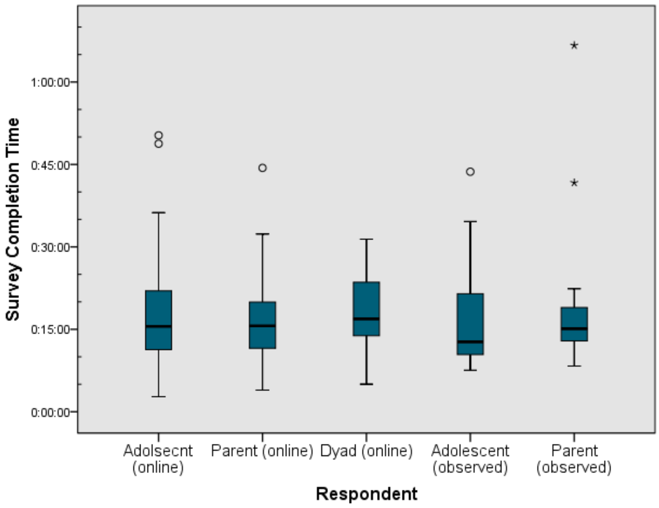
b) **Adolescent anxiety about dental treatment for hypodontia, as reported by adolescents and parents**



c) **Parental anxiety about dental treatment**

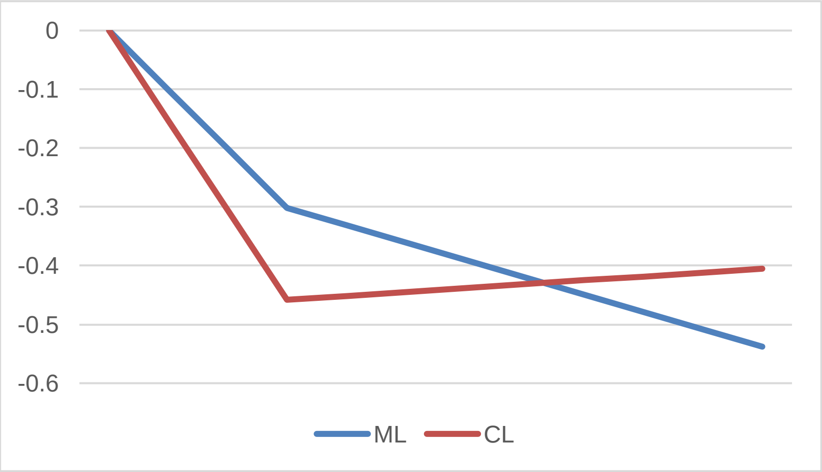


Appendix Figure 12: Survey completion time for respondent groups

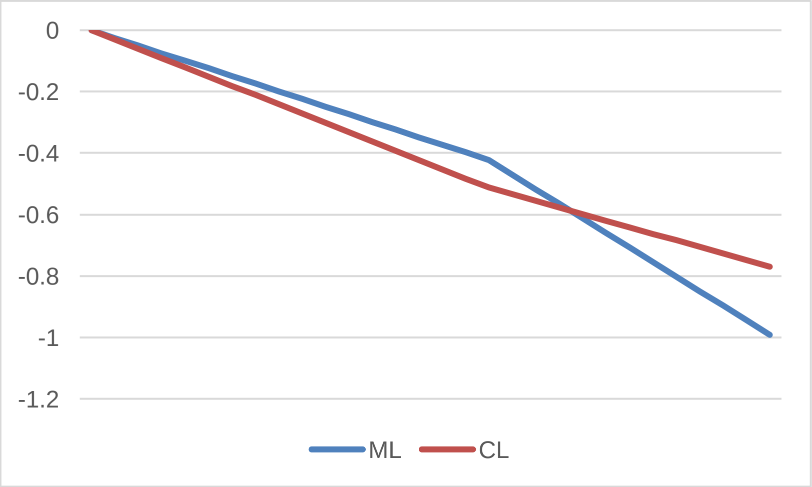


Appendix Figure 13: Examination of functional form of 3-level time attributes

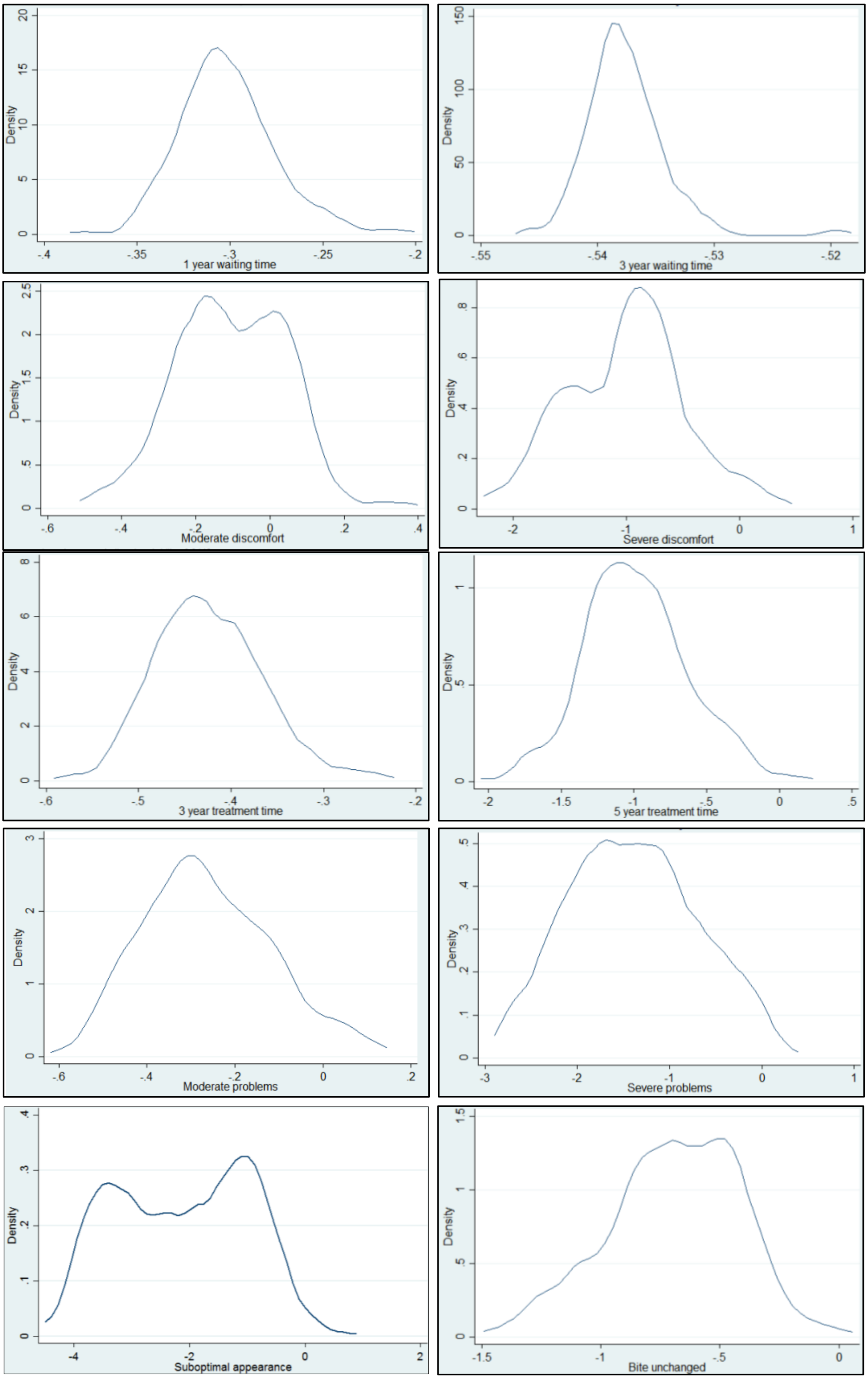
a) Waiting time



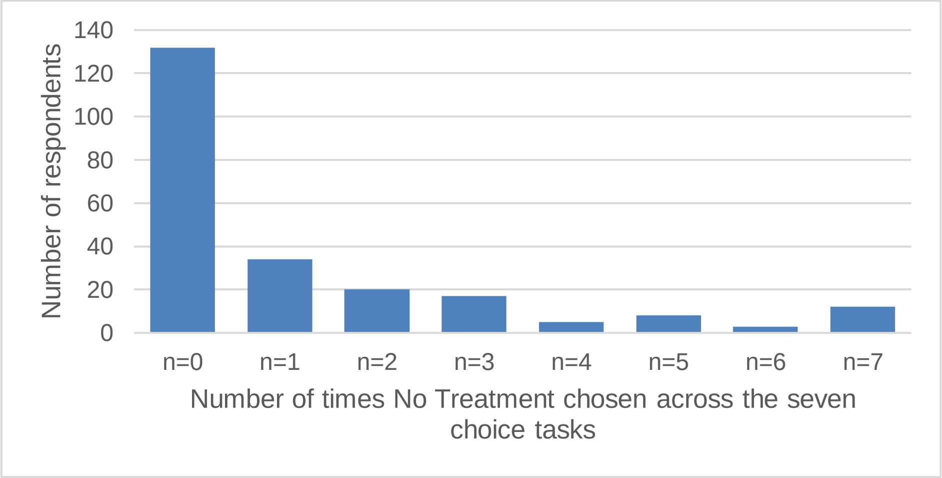
b) Treatment time



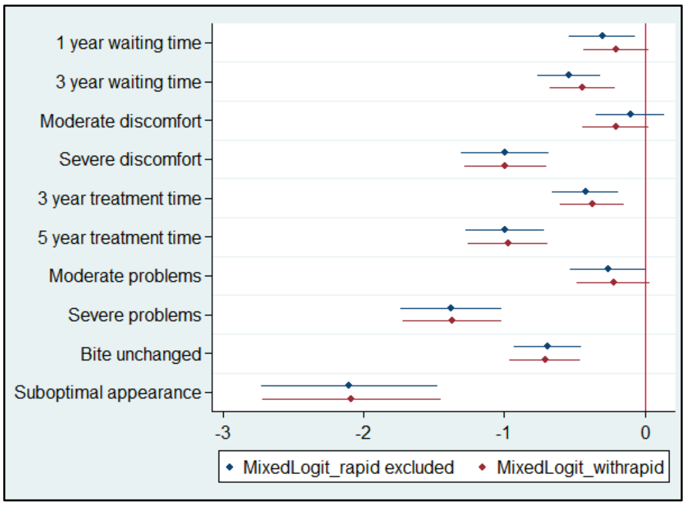
Appendix Figure 14: Distribution of individual-level coefficients for attributes



Appendix Figure 15: Selection of No Treatment by respondents across all choice tasks



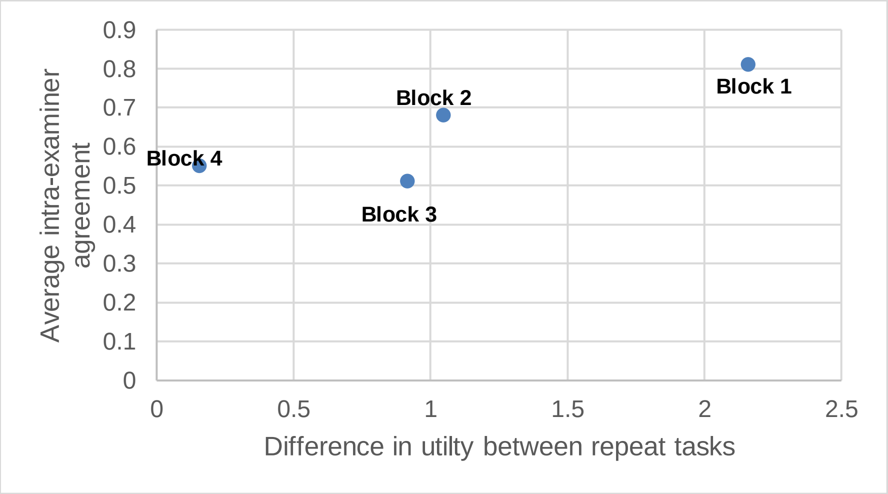
Appendix Figure 16: Sensitivity analysis of effect of exclusion by survey completion time. Attribute-levels are reported relative to the reference level with 95% CI. Estimated using mixed logit model



Preference weights for attribute-levels are given relative to the omitted level for that attribute, which has a value of zero (red line). Reference levels are

- Discomfort: none/mild
- Treatment time: 2 months
- Waiting time: 3 months
- Problems: none/mild
- Bite: improved
- Appearance: improved

Appendix Figure 17: Correlation between intra-examiner agreement and difference in choice task utility for repeat task (Pearson Correlation = 0.85)



Appendix Figure 18: Number of choice tasks in agreement per respondent (out of seven) in the initial and repeat survey (n=23)