Understanding Dental Fear and Anxiety in Children

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Abstract

Background

Dental fear and anxiety (DFA) assessment using standardised self-report questionnaires could improve patient care, facilitate communication, and reduce occupational stressors for dental professionals. However, existing measures have conceptual limitations and barriers to clinical utilisation.

Aim

To further the understanding of DFA assessment in paediatric dental patients.

Method

Firstly, a cross-sectional study was conducted to determine a profile of paediatric dental patients referred with DFA. Additionally, child mental health and health-related quality of life were assessed. Next, a qualitative approach was used to explore children's DFA experiences using a Cognitive Behavioural Therapy (CBT) framework. The findings informed the development on a new child centred DFA measure (Porritt et al. 2018). Subsequently, a web-based version of this measure for use on mobile smart devices was designed and tested.

Results

In Study 1, the profile of 100 children aged 11-16 years found most were female, White ethnicity, and lived in areas with high deprivation (61%, 85%, and 50% respectively). Participants had a range of self-reported DFA severity scores. Most did not have additional psychological difficulties and reported levels of impacts on daily living consistent with community samples. In Study 2, semi-structured interviews were conducted with 13 children aged 11-16 years. The thoughts, feelings, physical symptoms, and behaviours described were characteristic of changes associated with fears and anxieties. In Study 3, 107 children aged 9-16 years participated in the design and/or testing of the web-application. Mode of questionnaire delivery did not affect data equivalence or completeness, or time taken to complete responses. Both questionnaire versions had high acceptability, with no participant preference reported.

Discussion

DFA is a multidimensional experience for children. Further research is needed into the factors that influence children's self-reporting of DFA using standardised questionnaires. The functionality offered by web-applications on mobile smart devices has considerable potential for DFA assessment.

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Abbreviations

ACDAS	Abeer Children Dental Anxiety Scale
A.M.	Miss Annie Morgan, PhD student
A.T.	Mr Ashley Towers, Research Software Engineer
BMP	Behavioural management problems
BPRS	Behavioural Profile Rating Scale
CBT	Cognitive-behavioural therapy
C.C.	Professor Cathy Creswell, Professor of Developmental Clinical Psychology
CDAS	Corah Dental Anxiety Scale
CDHS	Child Dental Health Survey
CEDAM	Children's Experiences of Dental Anxiety Measure
CFSS-DS	Children's Fear Survey Schedule Dental Subscale
CFSS-DS_f	Children's Fear Survey Schedule Dental Subscale (faces version)
Child-OIDP	Child Oral Impacts on Daily Performances
CHU-9D	Child Health Utility 9D
CV	Cognitive Vulnerability
CVM	Cognitive Vulnerability Model
DAQ	Dental Anxiety Question
DCQ	Dental Cognitions Questionnaire
DFA	Dental fear and anxiety
DFS	Dental Fear Survey
dfs	Decayed, filled surfaces in primary teeth
DGA	Dental general anaesthetic
DMFS	Decayed, missing due to caries, and filled surfaces in permanent teeth
dmfs	Decayed, missing due to caries, and filled surfaces in primary teeth

DMFT	Decayed, missing due to caries, and filled permanent teeth
dmft	Decayed, missing, and filled primary teeth
DSM	Diagnosis and Statistical Manual of Mental Disorders
EAS	Emotional, Activity and Shyness Scale of Child Temperament
eCEDAM	Electronic Children's Experiences of Dental Anxiety Measure
E.G.	Dr Ekta Gupta, Research Assistant
FIS	Facial Images Scale
FSSC	Fear Survey Schedule for Children
GDP	General dental practitioner
HADS	Hospital Anxiety and Depression Scale
H.R.	Professor Helen Rodd, Research Supervisor
HRQoL	Health-Related Quality of Life
IDAF	Index of Dental Anxiety and Fear
IDAF-4C+	Index of Dental Anxiety and Fear (Anxiety and Fear Module)
ICD	International Statistical Classification of Diseases and Related Health Problems
I.Q.R.	Interquartile range
J.P.	Dr Jenny Porritt, Research Supervisor
LSOA	Lower Super Output Area
MCDAS	Modified Child Dental Anxiety Scale
$MCDAS_f$	Modified Child Dental Anxiety Scale (faces version)
MDAS	Modified Dental Anxiety Scale
mHealth	Mobile Health
NICE	National Institute for Health and Care Excellence
OHRQoL	Oral Health-Related Quality of Life
ONS	Office for National Statistics

RCADS	Revised Children’s Anxiety and Depression Scale
ROC	Receiver Operating Characteristic
SDQ	Strengths and Difficulties Questionnaire
SFP	Smiley Faces Programme
SFP-R	Revised Smiley Faces Programme
STAI-C	State-Trait Anxiety Inventory for Children
STAI-S	State-Trait Anxiety Inventory State Anxiety Scale
U.K.	United Kingdom
USA	United States of America
VPT	Venham Picture Test
VAS	Visual Analogue Scale
Z.M.	Professor Zoe Marshman, Research Supervisor

Chapter One

1. Introduction

1.1. Background

Dental fear and anxiety (DFA) are common and widespread in children. Findings from the latest Child Dental Health Survey for England, Wales, and Northern Ireland showed that 14% and 10% of children, aged 12 and 15 years respectively, had high DFA levels (Health and Social Care Information Centre 2015a). Moreover, a systematic review of 50 studies from 21 countries suggested DFA is a global problem for children, with an estimated pooled prevalence of 24% (Grisolia et al. 2021).

There is good evidence that DFA has important implications for oral health and daily living of children. DFA creates barriers to children receiving appropriate and timely dental care, whereby children with DFA are less likely to have prevention, diagnosis and early treatment of dental caries (Seligman et al. 2017). This, and other factors, leave children with DFA at high caries risk, and perpetuate disparities in oral health between children with and without DFA (Klingberg et al. 1994b; Ridell et al. 2007; Nuttall et al. 2008; Humphris and Zhou 2014; Seligman et al. 2017; Scottish Dental Clinical Effectiveness Programme 2018). Although there has been limited research into the impact of DFA on children’s daily living, a recent systematic review identified that more severe DFA in children is related to poorer oral health related quality of life (Alharbi et al. 2021). It should also be acknowledged that the impacts of DFA extend to dental professionals. Although, dental professionals describe a sense of professional responsibility to help children with DFA, providing dental treatment for children with DFA can result in significant occupational stressors and economic concerns (Hill et al. 2008; Jones and Huggins 2014).

As DFA is a common and clinically important condition, being able to identify and measure it contributes to research, health service planning and delivery, and patient care. Within clinical practice, DFA assessment can be used to: quantify and understand the nature of DFA responses; inform decision-making about appropriate and timely DFA management; promote good communication; facilitate time management planning; and allow evaluation and monitoring of treatment outcomes (Armfield 2010b; Southam-Gerow and Chorpita 2010a; Buchanan 2012; Whiteside et al. 2016; Alshammasi et al. 2018). Therefore DFA assessment has potential to promote high quality patient care and reduce occupational stressors in dental professionals.

Multiple different questionnaires for use with children to self-report DFA have been described in the scientific literature (Schuurs and Hoogstraten 1993; Newton and Buck 2000; Porritt et al. 2013). However, the construct validity of these existing measures has been brought into question as they do not have a clear conceptual focus and have not been based on a theoretical framework for DFA in children (Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013; American Educational Research Association et al. 2014). That is not-to-say they are not valid, or able to identify DFA in children, but consideration is required about what they are, and are not measuring, or what part of the construct is not being operationalised (Armfield 2010b; Grisolia et al. 2021).

A potential conceptual limitation, is that existing DFA measures for use with children, assess DFA from an adult perspective. For example, questionnaires used with children, for research or clinical purposes, have often been intended for adults, or based on those for adults, but with minor modifications for children. When measures have been developed for children, a top-down approach has been used, whereby the content has been based on the scientific literature and expert opinion. Children's involvement, when reported, has generally been limited to late stages of measure development, and restricted to wording and comprehension issues. Prior to the research presented in this thesis, there were no measures where the content has been derived from qualitative interviews with children with DFA. The use of adult measures with children assumes that the DFA construct captured by the measure is the same in adults and children. Yet there are many factors that differ for adults and children in the dental setting which could influence children's DFA experience. These include: cognitive development, previous learning experiences, dental treatments (e.g. biological approaches to caries management), communication factors, health literacy of parents and children, role of parents and peers, power imbalance of relationships, and issues relating to autonomy and consent for treatment (Alfano et al. 2002; Themessl-Huber et al. 2010; Blomqvist et al. 2013; Gao et al. 2013). Therefore, it is likely that adults and children do have different DFA experiences, but how those differences fundamentally change the validity and relevance of DFA measures has not been explored (Silverman and Ollendick 2005). There is a strong argument that if DFA measures were developed with children, rather than for children, it would improve their validity and relevance (Stevens 2009).

A further criticism of existing measures is that they have a narrow conceptual focus on dental stimuli and situations that represent specific moments of a dental experience (Armfield 2010b; Porritt et al. 2013). If measures contain only a small number of items relating to specific dental stimuli, it is important that those items chosen are relevant to contemporary children's DFA experiences. If children have no knowledge of a questionnaire item, then they would not be able to determine the likelihood or severity of it resulting in DFA (Buchanan 2005). Correspondingly, measures that only capture part of a construct may underrepresent it, and fail to identify all children with DFA (Schuurs and Hoogstraten 1993). This consideration is more important for children with low levels of DFA than for children with high levels of DFA, for whom any dental stimuli would trigger DFA (Schuurs and Hoogstraten 1993). It is also possible that if different measures capture different parts of the DFA construct they would not necessarily identify the same children as having DFA (Locker et al. 1996). To compound this difficulty, studies report using different threshold values to identify DFA in children (Grisolia et al. 2021). As DFA is a continuum, there is no 'gold' reference standard for defining what is clinically important DFA (Kazdin 2005; Grisolia et al. 2021). Typically, DFA measures for use with children have used behaviour management problems (BMPs) as the reference criteria. Using dental BMPs may underrepresent DFA clinical significance as children with DFA behave in different ways within the dental environments, and not all behavioural actions associated with a response to DFA are disruptive (Freeman 2007). The use of BMPs also defines the clinical importance of DFA again from an adult perspective and fails to consider what children themselves think is important about their DFA experience (Porritt et al. 2013). These considerations make comparing DFA studies difficult, as findings may vary depending on the characteristics of the population of children identified with DFA by the particular measure used in the study (Locker et al. 1996). Importantly, this has implications for scientific efforts to understand DFA, and may mask true differences between populations (Locker et al. 1996).

A further consideration is that although standardised questionnaires for DFA assessment in children are widely available, they are not used by dental professionals for patient care (Dailey et al. 2001; Alshammasi et al. 2018). It has been put forward that this could be interpreted as dissatisfaction with existing measures (Schuurs and Hoogstraten 1993). Correspondingly, dental professionals have suggested that DFA questionnaires are not appropriate for clinical practice (Alshammasi et al. 2018). A potential barrier to their use is that they are perceived as being time-consuming to complete and difficult to score during a dental visit (Alshammasi et al. 2018). The use of an electronic questionnaire, completed and automatically scored prior to a dental appointment, could improve convenience of DFA assessment and address these time management concerns for dental professionals (Ventola 2014). To date, only one measure of childhood DFA has been developed for computer application (Buchanan 2005; 2010). However, it is only available on desktop computers which limits its wider usage. Mobile smart devices are already extensively used by health professionals during patient care (Ventola 2014; Marcano Belisario et al. 2015). Therefore, there is considerable scope to use mobile smart devices during DFA assessment in children.

1.2. Aims and objectives

The overall aim of the research described in this thesis is to further the understanding of DFA assessment in paediatric dental patients.

The research objectives to be addressed are as follows:

1. To assess the sociodemographic, quality of life, and child mental health characteristics of paediatric dental patients with DFA.
2. To explore the experiences of DFA in paediatric dental patients to inform the development of a new child-centred measure of DFA.
3. To design and test a web based DFA assessment measure for use on mobile smart devices.

1.3. Thesis structure

Chapter Two is a narrative review on the literature on DFA assessment in children. It is divided into six parts. Firstly, relevant definitions are introduced. The prevalence of dental fear and dental anxiety in children is then described, with reference to demographic and socioeconomic differences. The third section considers the clinical importance and impacts of dental fear and dental anxiety for children and dental professionals. Next, multiple factors involved in the development and maintenance of dental fear and dental anxiety in children are outlined. Finally, the methods of assessing dental fear and anxiety in children are reviewed. Throughout this chapter the conceptual considerations for DFA assessment for children are highlighted.

Chapter Three describes the rationale, aims and objectives of the research described in this thesis. The research was undertaken in three stages, and described in Chapters Four, Five, Six and Seven.

Chapter Four reports a quantitative study to determine a sociodemographic profile of children with DFA, who would be representative of a clinical population of patients for DFA assessment. That is, children who potentially would be users of a new DFA assessment measure. Participants also completed measures to assess child mental health and health related quality of life, and those findings are described. The use of multiple measures has been suggested for DFA assessment (Schuurs and Hoogstraten 1993).

Chapter Five reports a qualitative study where children directly describe their multidimensional DFA experiences using a Cognitive Behavioural Therapy (CBT) model for DFA assessment as a theoretical framework. A CBT model considers fears and anxieties to be composed of cognitive, affective, behavioural, and physiological responses (Kendall 1985; Barlow 2002). It has been suggested that the experience of DFA for a child is composed of these response systems, and the construct validity of assessment measures would be heightened if they were measured (Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013). The findings of this study were used to inform the development of a new DFA assessment measure for children: Children's Experiences of Dental Anxiety Measure (CEDAM).

Chapter Six and Seven describes the design and development of an electronic version of the CEDAM (eCEDAM) for use as a web-app on a tablet computer. In Chapter six, the qualitative approaches used to design and test the web-app for cognitive and usability problems with children are presented. Chapter Seven describes a quantitative evaluation to compare the data agreement, data quality, acceptability, and participant preference between the original paper version of the CEDAM and the eCEDAM.

Chapter Eight discusses the overall research findings, reflects on the study design, and describes the implications of the research presented for patient care and future research.

Chapter Nine summarises the main research findings, study limitations, recommendations, and ethical considerations from the research in this thesis.

Chapter Two

2. Literature review

2.1. Introduction

This chapter describes the scientific literature regarding assessment of dental fear and dental anxiety in children.

2.2. Definitions relevant to dental fear and dental anxiety in children

2.2.1. Dental fear and dental anxiety

The negative feelings associated with dental treatment in children are commonly referred to as dental fear and dental anxiety (Klingberg and Broberg 2007). More precise definitions have suggested that dental fear is an emotional reaction to specific threatening stimuli in the dental situation, whereas dental anxiety describes an apprehension that something dreadful is going to happen in relation to dental treatment (Klingberg and Broberg 2007). Conceptually, dental fear and dental anxiety also exist on a continuum of severity representing greater or lesser degrees of health and adaptation (Freeman 2005a; National Institute of Mental Health 2011; Garvey et al. 2016). However, within the dental literature, the terms 'dental fear' and 'dental anxiety' are frequently used interchangeably (Armfield 2010b). Armfield (2010b) has suggested this inconsistency in usage can be attributed to a lack of conceptual clarity. In this section, considerations for the definitions of fear, anxiety, and phobia, and their relevance to dental fear and dental anxiety in children will be described.

2.2.2. Childhood and adolescence

There is no consensus on what defines childhood and adolescence, with definitions varying for different governments and cultures (Matza et al. 2013). However, the United Nation's Convention on the Rights of the Child (1989), which has been ratified by 196 countries including the United Kingdom (U.K.), defines a child as a person below age 18 years. With respect to the term 'adolescence', the World Health Organisation (2014) suggests that this should be used to describe young people in their second decade of childhood, beginning at age 10 years. Throughout this thesis the term 'children' will be used as it is the more inclusive term and not age range dependent.

2.2.3. Fear and anxiety

Within the psychological literature there is generally a consensus that the emotions of fear and anxiety should be regarded on a conceptual level to be qualitatively different (National Institute of Mental Health 2011). Fear has been defined as a primitive alarm response to real, or imagined immediate danger (National Institute of Mental Health 2011). Fear occurs when there is the perception of a direct threat, although the stressors can be external (e.g. people, situations) or internal (thoughts, images) (Craske 2003). It is associated with: cognitive processing to focus all attention onto the threat; a surge of autonomic nervous system activity, and defensive behavioural actions to facilitate escape and survival from harm (e.g. flight or fight) (American Psychiatric Association 2013). Anxiety is differentiated from fear based on the proximity to a threat. It is described as the reaction to potential threats, where there is the expectation of adversity, but there is no immediate danger present (Craske 2003; National Institute of Mental Health 2011). Izard (2013) suggested this definition is limited, and anxiety is a far more complicated mood state than simply the emotion of fear without a direct threat, theorising that it contains a changeable pattern of multiple fundamental emotions that may include fear, sadness, anger, shame and guilt. In a well-accepted conceptualisation, Barlow (2002) proposed that anxiety is characterised by helplessness, because of a perceived inability to predict or control upcoming situations, suggesting it is comprised of related cognitive and affective components including: hypervigilance for threatening stimuli; cognitive distortions; negative affect; and supportive physiology (Barlow 2000; American Psychiatric Association 2013). Furthermore, anxiety is associated with an attentional shift, whereby the cognitive focus moves from the threat onto oneself, characteristically resulting in an inaccurate and critical assessment of abilities to overcome the threat (Barlow 2002). If anxiety processes become chronic, attempts to cope with the negative affect are characterised by a tendency to avoid entering a state of anxiety, and worrying over future events (Barlow 2002). Worry is a specific cognitive component of the anxiety response (Craske 2003). The process of worry can be adaptive and can act to decrease anxiety by occupying attentional focus and distracting an individual, causing a reduction in physiological responses (Hoehn-Saric and Mcleod 2000). However, when worry becomes persistent, and causes interferences with functioning, it is considered pathological (Barlow 2002).

2.2.4. State anxiety and trait anxiety

The distinction between adaptive and maladaptive dental fear and dental anxiety is challenging in children (Lodge and Tripp 1995). To distinguish an adaptive anxiety reaction from an anxiety disorder, Spielberger (1972) considered anxiety to have two distinct constructs: state anxiety and trait anxiety. Applying state and trait concepts, state dental fear and dental anxiety can be considered the transient fear and anxiety response to a situational stress within a dental context (Buchanan 2012). That is, state anxiety is the anxiety a child is experiencing at a particular moment (Spielberger 1972). It is normal and appropriate to have a state dental fear and dental anxiety response when confronted with an unfamiliar or challenging situation in the dental setting (Gustafsson et al. 2010a). Moreover, a state dental fear and dental anxiety response in a child may be influenced by their stage of development. Typically, developmental fears and anxieties are transitory and follow a pattern influenced by cognitive maturity and learning experiences, with new fears and anxieties developing as children become able to perceive dangers in different situations, but are not yet sufficiently advanced to understand the situation fully (Gullone 2000). For example, young children have predominately fears and anxieties related to their immediate environment (e.g. loud noises), school-age children, who are at a stage where fantasy and reality can be confused, commonly have fears and anxieties relating to the supernatural, whilst in adolescence there is an emergence of fears and anxieties relating to abstract concepts (e.g. environmental concerns, political issues) and social evaluation (Piaget 1964; Gullone 2000). With increasing age there is an overall decrease in the number of fears, although once in adolescence fears become more stable (Gullone 2000). Of relevance, fears and anxieties related to medical situations increase from childhood to young adulthood (Gullone and King 1997). However, as the dental situation is complex, children may experience dental fear and dental anxiety to different elements of the dental encounter depending on their stage of development (e.g. loud noises from dental equipment in young children, bodily injury during dental procedures in older children). For most children this reaction will decrease with cognitive maturation and further dental habituation (ten Berge et al. 2002a). In contrast, trait dental fear and dental anxiety is a stable characteristic, and is considered the predisposition or proneness to react with fear and anxiety (e.g. state response) to the dental situation (Spielberger 1972). Children may experience state dental fear and dental anxiety at different threat imminences (e.g. before a dental visit, during a dental visit) and different intensities (e.g. low to high intensities) regardless of whether it is an adaptive response, related to child development, or due to underlying trait dental fear and dental anxiety (Barlow 2002; De Jongh et al. 2011; National Institute of Mental Health 2011). It has been suggested that the core features that differentiates between adaptive and maladaptive dental fear and dental anxiety is not the intensity of the emotional experiences, but whether the dental fear and dental anxiety is persistent, developmentally inappropriate, and interferes with functioning (Barlow 2002).

2.2.5. Dental behavioural management problems in children

Anxiety and fear are associated with behavioural actions to avoid or escape (flight and fight response) the threatening situation (Barlow 2002; American Psychiatric Association 2013). In the dental setting these coping responses can be unhelpful, and may disrupt the provision of dental treatment (Seligman et al. 2017). In children such actions are defined as behavioural management problems (BMPs) (Klingberg and Broberg 2007). In the literature, a distinction is not always made between dental fears, dental anxieties and dental BMPs, with many studies exploring dental fear and dental anxiety based on children referred with dentist-reported uncooperative behaviour. Correspondingly, Wogelius and co-authors (2003) and Ramos-Jorge and co-authors (2006) have identified that dental BMPs are more common in children with high dental fear and dental anxiety levels. However, using dental BMPs as a proxy may underrepresent dental fear and dental anxiety as children behave in different ways within the dental environments, and not all behaviour associated with dental fear and dental anxiety will disrupt dental treatment (Freeman 2007). That is, children with dental fear and dental anxiety may be cooperative for dental treatment. Additionally, dental BMPs may not be fully explained by dental fear and dental anxiety alone. A cross-sectional study reported the prevalence of dental fear and dental anxiety and BMPs in Danish children (n=1281) aged 6 to 8 years (Wogelius et al. 2003). To identify BMPs, children's clinical records were reviewed for entries reporting disruptive behaviour. To assess dental fear and dental anxiety the parent version of the Children's Fear Survey Schedule Dental Subscale (CFSS-DS) was used (DFA score≥38). The CFSS-DS is the most frequently used measure reported in the dental literature to assess trait dental fear and dental anxiety in children (Cuthbert and Melamed 1982; Porritt et al. 2013). It requires children to rate their level of fear to 15 dental situations (e.g. having to open your mouth, the noise of the dentist drilling) using a five-point response scale (1=not afraid to 5=very afraid. The study identified that 18% had a negative dental behavioural incident in their records, only 6% of participants were identified with dental fear and dental anxiety (Wogelius et al. 2003). This finding suggests that not all dental BMPs can be attributed to dental fear and dental anxiety. Admittedly, this study used clinical records and parent reports of dental fear and anxiety as a proxy for their children. Parental reporting has been shown to agree poorly with child self-reports (Gustafsson et al. 2010a; Klein et al. 2015; Patel et al. 2015). As the available evidence suggests that dental BMPs are not always attributable to dental fear and dental anxiety in children they will not be considered as a proxy for dental fear and dental anxiety throughout this review.

2.2.6. Dental phobia

When fears and anxieties persist and interfere with normal functioning they can develop into fear or anxiety-related disorders (American Psychiatric Association 2013; World Health Organisation 2018). Currently, two key manuals provide a categorical classification system for the diagnosis of such disorders in children: The Diagnosis and Statistical Manual of Mental Disorders (DSM) and the International Statistical Classification of Diseases and Related Health Problems (ICD), produced by the American Psychiatric Association and the World Health Organisation, respectively (American Psychiatric Association 2013; World Health Organization 2015). The two systems do not provide identical diagnostic criteria (Clark et al. 2017). Within the fifth edition of the DSM (2013) there are 11 separate anxiety disorders: separation anxiety disorder; selective mutism; specific phobia; social anxiety disorder; panic disorder, agoraphobia; generalised anxiety disorder; anxiety disorder due to another medical condition; other specified anxiety disorder; and unspecific anxiety disorder. According to this classification, the anxiety disorders differ based principally on the object of threat (e.g. social anxiety disorder describes fear or anxiety about one or more social situations in which an individual is exposed to possible scrutiny by others, compared to specific phobia which is fear or anxiety about a specific object or situation) (American Psychiatric Association 2013). Phobia about dentistry is classified as a type of specific phobia. It is characterised by the following diagnostic features: marked fear and anxiety about dental treatment that has been present for at least six months; fear and anxiety always being evoked by dental treatment, although the intensity of experience can vary across different situations and contextual factors; avoidance of dental treatment, or enduring dental treatment only with intense fear and anxiety; and clinically significant distress or impairment in functioning (American Psychiatric Association 2013). Within ICD 10 (1992), dental phobia in children is considered a phobic disorder of childhood if it shows marked developmental phase specificity i.e. the onset is developmentally appropriate, but the fear and anxiety experienced is clinically abnormal. It is developmentally appropriate for a child to have an emotional response when confronted with an unfamiliar or challenging situation in the dental setting (Gustafsson et al. 2010b). For most children this reaction will decrease with further dental visits and increased familiarity (ten Berge et al. 2002a). Therefore, a degree of anxiety and fear should be expected, but it becomes maladaptive when it is persistent, disproportionate to the situation, and it interferes with functioning (Gustafsson et al. 2010b). Outside psychosocial development, dental phobia is categorised by ICD 10 as a phobic anxiety disorder, with a subclassification of specific (isolated) phobia (World Health Organization 1992). The key diagnostic features include dental avoidance or enduring dentistry with dread; anticipatory anxiety on contemplation of the dental situation; anxiety that can vary in severity; and experiencing panic attacks. The key feature of a panic attack is an abrupt surge of intense fear or intense discomfort that reaches a peak within minutes (American Psychiatric Association 2013). They are associated with the following symptoms: palpitations; sweating; trembling or shaking; sensation of shortness of breath or smothering; feelings of choking; chest pain; nauseated feelings; feeling dizzy, light-headed or faint; chills or heat sensations; paraesthesia; derealisation (feelings of unreality); fear or losing control; and fear of dying (American Psychiatric Association 2013).

Both categorical classification systems have practical utility for clinicians as they provide an explicit criterion for the diagnosis of dental phobia. However, conceptually dental fears and dental anxieties exist on a continuum (Freeman 2005a). Using a dimensional approach, dental phobia exists at the extreme end of the continuum, but there is not a singular universal threshold that represents the point when dental fears and dental anxieties are considered a dental phobia (Brown and Barlow 2005).

2.2.7. Blood injury injection phobia

Children with blood injury injection phobia can be misdiagnosed as having dental phobia. Blood injury injection phobia describes a marked fear of witnessing bleeding/injury or receiving an injection or an invasive procedures (Vika and Agdal 2013). Historically, dental phobia has been classified as a subtype of blood injury injection phobia (American Psychiatric Association 1994), although some researchers have advocated for dental phobia to be considered an independent type of specific phobia (De Jongh et al. 1998; Lebeau et al. 2010). Regardless, there is a clinical overlap between blood injury injection phobia and dental phobia, as children with dental phobia may also experience fear and anxiety towards blood injury injection phobia-related stimuli (e.g. sight of blood, local anaesthetic) (Vika and Agdal 2013). However, in contrast to the autonomic hyperarousal associated with fear, blood injury injection phobia is physiologically distinct. It is characterised by a biphasic change in heart rate and blood pressure (Lebeau et al. 2010). This drop in heart rate and blood pressure can lead to an increased fainting tendency (Lebeau et al. 2010).

2.2.8. Summary of definitions relevant to dental fear and dental anxiety in children

Within the dental literature, the terms 'dental fear' and 'dental anxiety' are frequently taken to have identical meaning and used interchangeably. However, the constructs for fear and anxiety, as presented, have significant conceptual differences, although both emotional responses are multidimensional and have cognitive, behavioural, and physiological components (American Psychiatric Association 2013). A dimensional approach to fears and anxieties considers dental fears and dental anxieties to exist conceptually on a severity continuum representing greater and lesser degrees of health and adaption (Freeman 2005a; National Institute of Mental Health 2011; Garvey et al. 2016). Correspondingly, children can experience dental fear and dental anxiety at different threat imminences, different intensities, and different durations. Children may also experience fear and anxiety towards different aspects of the dental situation at the same time. There is a further need to consider this against the context of adaptive responses and child development. Using this dimensional approach, dental phobia exists at the extreme end of the dental fear and dental anxiety continuum. However, there is not a diagnostic threshold on the continuum whereby dental fear and dental anxiety are considered a dental phobia (Brown and Barlow 2005). Although dental fear, dental anxiety and dental phobia may have conceptual differences, there are significant challenges to distinguish between them in children (Gustafsson et al. 2010a; Porritt et al. 2013). Therefore, throughout this review, the comprehensive term 'dental fear and anxiety' (DFA) will be used to describe all forms of dental fears and dental anxieties, including dental phobia, in children (Klingberg and Broberg 2007).

2.3. Prevalence of dental fear and anxiety in children

2.3.1. Systematic reviews

The prevalence of DFA in children has been reported in the dental literature with two key systematic reviews. Klingberg and Broberg (2007) conducted a systematic review of DFA prevalence studies. A single electronic search engine (PubMed, United States National Library of Medicine) was used to identify studies published between 1982 and 2006. The authors limited their search to include only English language publications. Eligibility criteria restricted the number of DFA assessment measures to include: CFSS-DS; Dental Fear Survey (DFS) and Modified Dental Fear Survey (Taani et al. 2005); Corah Dental Anxiety Scale (CDAS), and Modified Dental Anxiety Scale (MDAS). These measures are described in detail in Section 2.6. The CDAS and MDAS are adult measures, although have been employed in child research. These measures were selected as they have DFA threshold values reported. However, limiting the search to certain measures acted to exclude national representative surveys that used other approaches to identify DFA. Case-reports and interventional studies reporting treatment outcomes were also excluded. Later the review was extended to include papers published up until 2011 (Klingberg 2013). In total, fifteen studies were found to meet the eligibility criteria (range: 223 to 3597 individuals). Two publications were based on the same study population so were considered together (Milgrom et al. 1995; Raadal et al. 1995). The children in the study populations were aged from 4 to 18 years. The included studies were conducted in Jordan, New Zealand, North America, Northern Europe, Russia, Singapore, Taiwan, and Turkey. Only five included studies were published within the last 20 years (ten Berge et al. 2002b; Wogelius et al. 2003; Taani et al. 2005; Lee et al. 2007; Akbay Oba et al. 2009). Most studies (n=12) employed a cross-sectional design whereby children were recruited from schools or public dental health services using and non-random sampling strategies. The remaining two studies were a birth cohort (Thomson et al. 1997), and a cohort of school children followed longitudinally (Murray et al. 1989). The CFSS-DS was used as the assessment measure in eight of the studies, although both parental and self-report versions were used. Threshold values to identify children with DFA also varied, even when the same measure was used. Overall, the prevalence of DFA reported varied from 6% to 21%, with a mean prevalence of 12%. Using only children's self-reports (n=10 studies), the mean prevalence of DFA was 13% (range 7% to 20%).

Grisolia and co-authors (2021) also conducted a systematic review and meta-analysis of DFA prevalence studies to determine the global prevalence of DFA in children. Multiple electronic databases (n=7) were searched to identify studies published between 1985 and 2020. The authors also searched for grey literature and hand-searched six dental journals. Eligibility criteria limited DFA measures to those identified as valid and reliable for the study population. In total, 50 studies (reported in 57 papers) were identified for inclusion. The studies were from 21 countries, although most studies were based in Europe (n=20) or South America (n=16). Only five papers were not published in the last 20 years (Chellappah et al. 1990; Milgrom et al. 1992; Klingberg et al. 1994a; Milgrom et al. 1994; Klingberg et al. 1995a). Most studies employed a cross-sectional design (n=47) and non-random sampling strategies. The children in the study populations were aged from 3 to 18 years. The CFSS-DS was the most frequently used measure (n=27), although both parental and self-report versions were used. Other measures were Children's Fear Survey Schedule Dental Subscale (faces version) (CFSS-DS_f), Dental Anxiety Question (DAQ), Dental Fear Survey (DFS), Modified Child Dental Anxiety Questionnaire (faces version) ($MCDAS_f$), and Venham Picture Test (VPT). These measures are described in Section 2.6. The pooled DFA prevalence was 24% (95% Confidence Interval 20%-27%).

2.3.2. Prevalence in the child population in the United Kingdom

There have been two cross-sectional surveys investigating the prevalence of DFA in children living in the U.K. In 2003 a measure of DFA was included in the U.K. Child Dental Health Survey (CDHS) for the first time since 1973 (Nuttall et al. 2008). A single question was incorporated into a parental questionnaire with other items relating to patterns of care, service use and socio-economic status. The global question required parents/carers to rate their child's willingness to attend a dental visit, taking their child's anxiety into consideration, on a five-point response scale for severity of behavioural impact. The question had not been previously validated. The questionnaire was distributed to a random subsample of parents/carers taken from the overall representative sample of children aged 5, 8, 12 and 15 years (Morris et al. 2006; Nuttall et al. 2008). Questionnaires were sent to 5,480 parents/carers from whom 3,342 were returned (61% response rate) (Morris et al. 2006). Only 34 (0.2%) respondents failed to complete the question on DFA (Nuttall et al. 2008). According to their parents, a total of 22%, 25%, 28% and 25% of children aged 5, 8, 12, and 15 years respectively, were affected by DFA. Between 1.0 and 1.5% of parents/carers reported that their child would either only agree to attend a dental visit if they were in pain or would refuse to go altogether because of severe DFA, which is suggestive of high DFA. Although the inclusion of only a single question in the 2003 U.K. CDHS limits the interpretation of the findings, large cross-sectional surveys in other countries have also used similar brief approaches.

In 2013, children aged 12 and 15 years who participated in the CDHS for England, Wales and Northern Ireland, self-completed a paper questionnaire on their perceptions and attitudes towards their dental health (Health and Social Care Information Centre 2015a). This included a standard version of the MDAS measure for DFA (Humphris et al. 2013). The MDAS has previously been used to assess DFA in the U.K. Adult Dental Health Survey (Humphris et al. 2013). It comprises five items relating to anxiety provoking dental situations (e.g. sitting in the waiting room, about to have a tooth drilled), with a five-point response scale (1=not anxious, 5=extremely anxious) (Humphris et al. 1995). A total score of 19 has been used to indicate severe dental anxiety (King and Humphris 2010; Hill et al. 2013). However, its psychometric properties in relation to DFA in British children have not been determined. Moreover, the MDAS threshold score of 19 has been identified as underestimating the prevalence of severe DFA in a child population (Honkala et al. 2014). Overall, nearly all the children aged 12 and 15 years (99.6%) who had a dental examination (n=4950) completed the questionnaire (Health and Social Care Information Centre 2015a; b). High levels of DFA (MDAS total score≥19) were identified in 14% and 10% of children aged 12 and 15 years respectively, whilst over half of the participants (62% and 54% of children aged 12 and 15 years respectively) were identified with moderate levels of DFA (MDAS total score=10 to 18) (Health and Social Care Information Centre 2015a). In the same survey, parents of children (n=2307) aged 5 and 8 years rated their child's DFA on a scale of 1 to 10 (1=not anxious, 10=extreme anxiety). About half (51% and 55% at 5 and 8 years respectively) of all children in the sample were not perceived as having DFA by their parents. However, moderate to extreme levels of DFA were reported for between 17 to 21% of children. Additionally, 1% to 2% of parents reported their child did not attend the dentist, although the actual proportion of those children who did not attend because of DFA was not clear.

2.3.3. Role of age on dental fear and anxiety prevalence in children

Age trends for childhood DFA may aid discrimination between developmental DFA (i.e. DFA that is part of normal child development and will resolve, see section 2.2.4.) and maladaptive DFA during clinical assessment (Silverman and Ollendick 2005). However, cross-sectional studies report conflicting evidence for the influence of age on DFA. Klingberg and co-authors (1994a) and Wogelius and co-authors (2003) have identified that with increases in children's age (aged 4 to 11 years, and 6 to 8 years, respectively) there is a corresponding DFA increase. Dogan and co-authors (2006) reported the opposite; that DFA scores in Turkish children decreased from age 8 to 12 years. Other studies have found no difference in children aged from 4 to 11 years, 8 to 15 years, and 5 to 11 years (ten Berge et al. 2002b; Nakai et al. 2005; Nicolas et al. 2010). Majstrovic and Veerkamp (2005) reported a decrease in DFA between ages 4 and 11 years in children recruited from an outpatient clinic in a Dutch university teaching hospital, but then noted, after 11 years of age, DFA levels increased again. Similarly, U.K. children aged 12 years had the highest prevalence of DFA in the 2003 CDHS, and the highest prevalence within participants who completed self-reports of DFA in the 2013 CDHS (Nuttall et al. 2008; Health and Social Care Information Centre 2015a). Correspondingly, 12 years has been considered as the age dental phobia first presents (Ost 1987). With advancing age it has been proposed cognitive development may result in children having greater awareness of dental treatments, different coping strategies, and increased sensitivity to interpersonal elements of dental interactions, which may increase children's susceptibility for DFA development (ten Berge et al. 2002a). However, chronological age is, at best, a fairly rudimentary index of child development and understanding of the dental situation (Silverman and Ollendick 2005).

The above studies all had a cross-sectional design and only recorded DFA at a single point in time. However, two recent cohort studies have indicated that the natural history of DFA in children is also not straightforward. Klaassen and co-authors (2008) reported the finding of a longitudinal evaluation conducted in the Netherlands that followed a convenience sample of 401 participants. The children were between the ages of 5 to 10 years and 8 to 13 years during the first and second study assessment, respectively. To measure DFA, the parent version of the CFSS-DS was used. The response rate was excellent at 90%. Overall, the proportion of children with DFA decreased, but the mean DFA scores for children with DFA stayed the same, suggesting that DFA in those children remained stable. Tickle and co-authors (2009) reported the findings for children from North-West England followed from ages 5 to 9 years. The DFA assessment was carried out at age 5 years and repeated at 9 years, with parents asked to rate their child's DFA on a single five-point response scale (Milsom et al. 2003). The overall prevalence of DFA was found to have significantly increased, particularly in girls. However, most of the children with DFA at 5 years (54%) no-longer had DFA by 9 years. Between the assessment periods, a notable 34% of children were lost from the study. The children lost to follow-up had significantly higher levels of DFA, parental anxiety, caries experience, and history of extractions when aged 5 years than the continuing study sample. This suggests the reported findings are an underestimation of DFA in children at 9 years.

2.3.4. Role of sex on dental fear and anxiety prevalence in children

In the 2013 CDHS, female participants aged 12 and 15 years, were more likely to have high DFA levels than their male peers (Health and Social Care Information Centre 2015a). Correspondingly, male participants were more likely to report no/low levels of DFA (Health and Social Care Information Centre 2015a). In contrast, for children aged 5 and 8 years, the findings were conflicting; at age 5 years no sex differences were identified, whilst at 8 years, females had lower reported DFA levels than male participants. Regardless of methodological differences, there is some evidence to suggest that sex differences heighten with increasing age. Epidemiological studies from Sweden, Denmark, Turkey and France that included children aged 4 to 6 years and 9 to 11 years, 6 to 8 years, 8 to 12 years, and 5 to 12 years, respectively, identified no differences in DFA between male and female study participants (Klingberg et al. 1994a; Wogelius et al. 2003; Dogan et al. 2006; Nicolas et al. 2010). However, Bedi and co-authors (1992) and Taani and co-authors (2005) did identify sex differences in Scottish children aged 13 to 14 years and Jordanian children aged 12 to 15 years, whereby females had higher DFA levels than males.

2.3.5. Role of social deprivation on dental fear and anxiety prevalence in children

In the U.K., childhood DFA has previously been associated with social deprivation, with children with DFA more likely to be in receipt of free school meals (taken as a proxy measure of social deprivation) (Townend et al. 2000; Nuttall et al. 2008). Similarly, studies in Sweden, North America, Turkey, and Jordan also found social deprivation was associated with higher levels of DFA (Klingberg et al. 1994a; Milgrom et al. 1998; Taani et al. 2005; Dogan et al. 2006). Findings from U.K. epidemiological surveys have demonstrated that children from socially deprived backgrounds are more likely to have experienced dental decay, and have dental decay that is severe or extensive, than children from less deprived backgrounds (Health and Social Care Information Centre 2015c; Public Health England 2020). Children from socially deprived backgrounds are also less likely to engage regularly with dental professionals and attend for routine dental visits, and more likely to attend only when there is a dental problem (Health and Social Care Information Centre 2015c). Problem-orientated attendance is a situation that is likely to increase DFA, rather than provide learning opportunities that would have a positive impact on DFA levels (Liddell and Locker 2000). Therefore, the evidence would suggest that DFA and social deprivation share factors that are associated with poor oral health outcomes. A reasonable assumption is that if a child has severe and extensive dental caries experience, the child might be at an increased risk of negative dental experiences that could contribute to DFA development. This is supported by evidence that children with DFA are more likely to have had restorative experience in the primary dentition (Coxon et al. 2019a). However, the most recent U.K. epidemiological data did not find a relationship between DFA and receipt of free school meals (Health and Social Care Information Centre 2015a). A possible explanation is that dental general anaesthetics (DGA) are increased in children with DFA and from socially deprived backgrounds (Hariharan et al. 2017; Knapp et al. 2020). DFA is not influenced in the short-term by dental treatment with a DGA, and DFA severity remains unchanged post-operatively (Balmer et al. 2004; Klaassen et al. 2009). Moreover, children from socially deprived backgrounds have been reported to have more extractions under DGA than children from affluent background, which may then reduce their need for dental treatment until they have experienced further dental disease (Harper et al. 2020).

2.3.6. Comparing dental fear and anxiety prevalence between studies

Locker and co-authors (1996) evaluated the concordance of three DFA rating scales and found a very different prevalence rate was delivered by each of the measures, for the same population. An explanation is that different measures operationalise the construct of DFA in different ways. Therefore, direct comparison of prevalence rates between studies that have used different DFA measures is not recommended (Locker et al. 1996).

Assessment measures can be used to quantify a child's magnitude/severity of DFA by determining a score (Silverman and Ollendick 2005). To interpret scores, threshold values can be used to dichotomise individuals to identify those whose DFA is considered clinically important, or to categorise individuals into ranges of different DFA severities (e.g. low, moderate, high). One approach to determining threshold values that has been applied to DFA assessment measures, is the use of receiver operating characteristic (ROC) curve analysis. In ROC curve analysis a reference standard, assumed to be the operational definitions of the construct of interest, is used to dichotomise individuals (e.g. cases, non-cases) (Streiner et al. 2015). Subsequently, a graph of the false positive rate for the assessment measure (e.g. 1-specificity) is plotted against true positive rate (sensitivity) (Streiner et al. 2015). Threshold values are determined by the point on the curve where the overall error rate is the lowest (Streiner et al. 2015). However, to compound the difficulties, there is no 'gold' standard for the reference criterion to determine presence or absence of clinically important DFA, and studies to determine threshold values have reported using different external criteria (Kazdin 2005; Grisolia et al. 2021). Measures for DFA in children have typically used dental BMPs that have been reported by a referring dental professional as their categories (Freeman 2007; Gustafsson et al. 2010a). Although, dental BMPs have important clinical impacts, these are not exclusive to children with DFA and are more likely to represent the dental professional's perception of whether a child's behaviour in the dental setting is cooperative or uncooperative (Klingberg et al. 1994b; Aartman et al. 1996; Wogelius et al. 2003; Freeman 2007). Therefore, when applying the ROC curve analysis, the use of dental BMPs as the reference standard for childhood DFA, may only identify individuals with dental BMPs and childhood DFA. Additionally, Youngstrom (2013) had advocated against the use of a categorical classification system and threshold values suggesting for conditions that exist on a continuum, such as DFA, it creates artificial and arbitrary distinctions between health and illness.

2.3.7. Summary and discussion of prevalence of dental fear and anxiety in children

The evidence suggests that DFA is common in children. There is also evidence that as age increases there is a difference in DFA prevalence for male and female children, with female children reporting higher DFA levels. However, what role age and social deprivation has on DFA prevalence was not clear, this suggests that it is experienced by children of different ages and backgrounds. There are significant limitations to comparing the findings between prevalence studies. Notably, prevalence can vary with the population studied (community or clinical), the sampling strategy (random or non-random), the assessment measure and threshold used to identify DFA, and the source of informant (children, parent, or clinician) (Lodge and Tripp 1995; Grisolia et al. 2021). There may also be cultural differences that may influence DFA when comparing studies between different countries. For example, local health care systems, the availability and acceptance of psychological and pharmacological DFA treatments, trust of dental professionals, importance placed by dental professionals on communication skills, and availability of novel clinical techniques and treatments (e.g. placement of preformed crowns using the non-invasive Hall Technique or wand computerised digital systems for local anaesthesia) (Armfield and Heaton 2013; Milgrom et al. 2013). For the most recent CDHS, DFA was assessed using an adult measure and threshold score. The measure had not been validated for children. The use of adult measures for children assumes there are no differences between the DFA experienced by adults and children. However, children are likely to have a different experience. Possible factors include: the child-dentist relationship and its power balance; the role of parent and peer group; communication factors and health literacy of parents and children; parental advocacy abilities; and modalities of dental investigations and treatments for adults and children (Themessl-Huber et al. 2010; Zhou et al. 2011; Blomqvist et al. 2013; Gao et al. 2013). Further research is needed to understand how DFA is experienced by children.

2.4. The impact of dental fear and anxiety on children and dental professionals

2.4.1. Impact of dental fear and dental anxiety in children on caries experience

Childhood DFA is associated with increased caries experience in the primary dentition (Coxon et al. 2019a). Within the mixed and permanent dentitions, the evidence becomes conflicting. A descriptive analysis of the most recent CDHS for England, Wales and Northern Ireland initially identified that children aged 12 and 15 years with high levels of DFA (MDAS≥19) were more likely to have active dental caries and soft tissue lesions (taken as a measure of signs of acute and chronic dental infection) (Coxon et al. 2019b). However a further regression analysis of the data set found that DFA was not a predictor for caries experience or dental infection. As previously discussed, a consideration in the use of the MDAS in the 2013 CDHS is the appropriateness of using adult threshold values as cut-off scores to identify children with DFA, and whether this would have under-represented the proportion of children with DFA (Honkala et al. 2014). However, similarly, no correlation has been found between decayed, missing due to caries, and filled permanent teeth (DMFT) scores and DFA scores for Jordanian children aged 12 to 15 years, and between DMFT/ decayed, missing due to caries, and filled primary teeth (dmft) and DFA scores for Greek children aged 6 to 12 years (Taani et al. 2005; Boka et al. 2017). In contrast, Akbay Oba (2009) found significant differences in the decayed, missing due to caries, and filled permanent tooth surfaces scores (DMFS) and decayed and filled primary tooth surfaces (dfs) scores for Turkish children aged 7 to 11 years with DFA than those without DFA. When only decayed teeth are included, U.K. studies have demonstrated that childhood DFA is associated with increased prevalence of untreated carious lesions (Townend et al. 2000; Nuttall et al. 2008). Whereas children with more filled tooth surfaces are less likely to have DFA (Boka et al. 2017). Moreover, there is also evidence that DFA occurs less frequently in children who have no caries experience, and that DFA during adolescence is a risk factor for high caries experience (DMFT≥10) in young adults (Julihn et al. 2006; Grisolia et al. 2021). To exacerbate any negative oral health impacts, children with DFA are less likely to have dental treatments that prevent dental caries (e.g. fissure sealants) and more likely to need treatments for extensive dental caries (e.g. endodontic treatment and extractions) (Ridell et al. 2007; Nuttall et al. 2008). In the most recent Adult Dental Health Survey for England, Wales and Northern Ireland, adult participants identified with DFA (MDAS≥19) were more likely to have untreated dental decay and soft tissue lesions than individuals without DFA (Heidari et al. 2015).

2.4.2. Avoidance of dental care

There is evidence that the prevalence of missed appointments is higher in children with DFA when compared to children without DFA (Skaret et al. 1999; Wogelius and Poulsen 2005). Delays in obtaining dental care can influence the possible management options for children (Newton et al. 2012). For example, if a child is only brought for a dental visit when they are experiencing dental pain or infection then potentially more challenging dental treatments for extensive dental caries are required. As children are not independently able to decide whether to attend dental appointments, missed appointments suggest there is decision-making by parents to avoid taking their child. A qualitative study that explored parental motivations behind these decisions found that parents felt overwhelmed and unable to convince their child with DFA about the need/importance of dental care (Hallberg et al. 2008). Other parents have reported a reluctance to put their child through unpleasant dental experiences that they felt they had endured themselves, whilst others described avoiding taking their child because they did not want to pass their DFA onto them (Smith and Freeman 2010). Consequently, avoidance of dental appointments in childhood DFA may demonstrate a response to DFA by parents and carers rather than by the child themselves.

In children, avoidance of dental care can manifest as dental BMPs (see section 2.2.5.). For example, evidence indicates that anxiety increases the likelihood of children refusing treatment by 170 times (Humphris and Zhou 2014). BMPs are not isolated to children, with adults with DFA reporting crying in the dental surgery, and displaying physical aggression towards dental professionals (Cohen et al. 2000). However, behaviours that prevent dental treatment can have negative implications for childhood oral health status. Children with a history of BMPs (parent-reported) are twice as likely to have dental caries at age 5 years than children without BMPs (Wigen et al. 2009). Additionally, children with reported BMPs are less likely to have had dental radiographs, and more likely to have restorative treatment completed without local anaesthetic (Klingberg et al. 1994b). If dental professionals are unable to provide dental treatment children may be referred to secondary or tertiary dental services with longer time periods until treatment is provided (Harris et al. 2008). However, Klingberg and co-authors (1994b) also identified that for 12% children who had a BMP, their dental treatment was postponed indefinitely.

When young adults begin to take responsibility for their own oral health care there is a linear increase in frequency of missed dental appointments (Skaret et al. 1999). A study in Norway, where all children are given free dental care during childhood, highlighted the risk factors for avoidant dental behaviour during adolescence. Questionnaires were sent to young adults aged 20 years who had attended public dental health services between the ages of 12 and 18 years (Skaret et al. 2000). To assess DFA the DFS was used. The DFS is a 20 item measure that assesses DFA related to dental avoidance (2 items), physiological arousal (5 items), fear and anxiety provoking stimuli and dental procedures (12 items), and a global DFA item (Kleinknecht et al. 1984). Users rate their response to each item on a five-point Likert scale (1=not at all, 5=very much). The total DFS scores can range from 20 (no/minimal DFA) to 100 (most severe DFA). A score equal or above 60 has been suggested as a threshold value for severe DFA (Milgrom et al. 1988). Overall, 754 questionnaires were returned (78% response rate). A subsample of 124 participants who had missed over 20% of their appointments (categorised as avoidant behaviour) were identified from clinical records. The variables found to be independently associated with a high frequency of missed appointments included: forgetting to attend (odds ratio=6.8); negative beliefs about dentists (odds ratio=3.4); high dental DFA (odds ratio=2.6); giving dental treatment a low priority (odds ratio=2.5); and high caries experience (odds ratio=2.4). 'Forgetting to attend' was also associated with high levels of DFA. Significantly more young adults who reported avoidant dental behaviour rated their oral health as poor compared to those that attended for regular dental care (Skaret et al. 2007).

2.4.3. Impact of dental fear and anxiety on children's daily life

The impact of oral conditions on individual's daily living is most often assessed using measures of health-related quality of life (HRQoL). HRQOL has been defined as, 'a combination of a person's physical, mental and social well-being; not merely the absence of disease' (National Institute for Health and Care Excellence (NICE) Glossary. © 2020, letter=h). Measures of HRQoL can be generic, or site, or disease specific. To date, DFA in children has only been evaluated with respect to oral health-related quality of life (OHRQoL). Locker and Allen (2007) define oral OHRQoL as, 'the impact of oral disorders on aspects of everyday life that are important to patients and persons, with those impacts being of sufficient magnitude, whether in terms of severity, frequency or duration, to affect an individual's perception of their life overall'. In the adult population, there is a substantial difference in OHRQoL scores between individuals with and without DFA (McGrath and Bedi 2004). In the U.K., adults with high levels of DFA have been shown to be amongst those with the poorest OHRQoL, even when accounting for confounding factors (age, gender, and social class) (McGrath and Bedi 2004).

There has been a paucity of research about the impact of DFA on daily living in children. However, the Child Oral Impacts on Daily Performances (Child-OIDP) measure was included in the CDHS 2013 for children aged 12 and 15 years (Health and Social Care Information Centre 2015a). This measure of OHRQoL requires users to score their difficulty with daily activities (eating, speaking, cleaning their teeth, relaxing/sleeping, smiling, laughing, or showing their teeth without embarrassment, doing schoolwork, and enjoying being with other people) and feeling different. Each item is scored on a three-point scale (0=not at all, 3=a lot) and a total score summed (Ravaghi et al. 2016; Coxon et al. 2019b). The English version of the Child-OIDP has been validated (Yusuf et al. 2006). Overall, children with severe DFA (MDAS≥19) had significantly poorer OHRQoL compared to children without severe DFA (Coxon et al. 2019b). Additionally, children with severe DFA reported more difficulty eating, cleaning their teeth, smiling, laughing, or showing their teeth without embarrassment, and enjoying being with other people (Coxon et al. 2019b). They also reported they felt different more often (Coxon et al. 2019b).

Alharbi and co-authors (2021) recently published a systematic review and meta-analysis to determine the association between DFA and OHRQoL in children. Multiple electronic databases (n=8) were searched to identify studies published before 2020. The authors also hand-searched reference lists. In total, five studies were identified for inclusion in the meta-analysis. The studies were from Europe, India, and the Middle East. The studies were all published between 2015 and 2019. Most studies employed a cross-sectional design (n=4). The children (n=7466) in the study populations were aged from 8 to 15 years. The measures used to assess DFA were the CDAS, CFSS-DS, DAQ, FIS, and MDAS. These measures are described in Section 2.6. All measures of OHRQoL included were determined to be valid and reliable. The overall effect size was 0.06 (95% Confidence Interval 0.04-0.08, fixed effect model). This suggests there is an association between DFA and OHRQoL in children, whereby more severe DFA is related to poorer OHRQoL.

2.4.4. Long-term consequence of childhood dental fear and anxiety

Participants that are part of a birth cohort study in New Zealand have been followed to explore patterns of change for DFA during adolescence and early adulthood, and have provided evidence that if DFA is still present at age 15 years it can become a long-term difficulty (Locker et al. 2001b; Poulton et al. 2001; Thomson et al. 2009). The cohort comprised 1037 individuals born in Dunedin between 1972 and 1973 (Locker et al. 2001b). As part of a battery of medical and psychological measures, DFA was assessed at 15, 18, 26 and 32 years using the CDAS (Locker et al. 2001b; Thomson et al. 2009). The CDAS is the most frequently used measure for adult DFA in the dental literature (Armfield 2010b). It comprises four potentially fear and anxiety provoking dental scenarios with a five-point response scale. However, a well-accepted limitation is that for items two, three and four the corresponding response scale is not ordinal, and has responses that are not mutually exclusive (Armfield 2010b). Within the study DFA was identified as a CDAS score ≥13 (Locker et al. 2001b; Thomson et al. 2009). Locker and co-authors (2001) reported that overall a third of participants experienced DFA during the follow-up period. At age 15, 11% (n=72) of participants had DFA. Between ages 15 and 26 years, DFA did show high remittance rates (54%), but for some individuals their DFA persisted (22%), whilst for others it remitted only to recur later in life (24%). The authors found that the outcome could not be predicted by sex or severity of DFA at age 15 years.

Thomson and co-authors (2009) conducted statistical modelling to identify developmental trajectories for DFA following a further assessment point at age 32 years. Data for DFA were available for 828 (86%) participants for at least three of the four time points. Most participants (78%) were identified as non-dentally fearful and anxious at age 15 years; and remained so for all the assessment points. For the 73 individuals with DFA at age 15 years, 82% were classified within a stable-anxious trajectory, whereby their DFA did not change between age 15 and 32 years, and 18% were classified within a recovery group, as their DFA remitted. The remaining participants (14%) were not dentally fearful or anxious at age 15 years but developed DFA in late adolescence or adulthood. One predictor of DFA trajectory was caries prevalence at age five years, whereby children in the stable anxious (persistent) group had the highest mean dmfs scores at age five years, whilst the stable non-anxious (no/low dental anxiety) had the lowest mean dmfs/DMFS scores across all time points. Therefore, most children with DFA in adolescence will become adults with DFA, and early dental experiences may have long-term DFA implications for individuals.

2.4.5. Impact of dental fear and anxiety on dental professionals

The impact of DFA on dental professionals has been explored in questionnaire-based studies with general dental practitioners (GDPs) from Denmark, Germany, Norway, and the U.K. (Moore and Brodsgaard 2001; Hill et al. 2008; Diercke et al. 2013; Strom et al. 2015). Dental professionals surveyed report a sense of responsibility for helping patients with DFA (Hill et al. 2008). Irrespectively, treatment of dentally fearful and anxious children and adults was described as difficult, tiresome, time-consuming, and a source of occupational stress (Moore and Brodsgaard 2001; Hill et al. 2008; Strom et al. 2015). Less than half (n=193, 42%) of the U.K. GDPs surveyed by Hill and co-authors (2008) reported finding treating patients with DFA an enjoyable experience, although the proportion of GDPs who find providing dental treatment on patients without DFA an enjoyable experience was not provided. Kent and Blinkhorn (1991) suggests that the reported difficulties in treating patients with DFA could partly be attributed to the emotional burden of dental professionals being seen as the cause of the distress. Correspondingly, qualitative research has found that specialist dentists report that it can be difficult to separate themselves from the emotional experience of a patient interaction, and the emotional burden of appearing calm under pressure (Murray et al. 2016). Private GDPs in Denmark identified that they perceived dentally fearful and anxious patients to be unreliable, due to the frequency of missed appointments, cancelled appointments and emergency attendances, and considered them to be a poor economic risk (Moore and Brodsgaard 2001). Economic concerns were also expressed by U.K. GDPs who described a lack of remuneration for the extra time required to treat patients with DFA as having negative consequences for quality of dental care provided (Hill et al. 2008). However, it was acknowledged that developing a reputation for treating dentally fearful and anxious patients had positive impacts for establishing a dental practice (Moore and Brodsgaard 2001). Therefore, DFA has a wide variety of impacts on dental professionals and for service provision.

2.4.6. Summary and discussion of clinical relevance of DFA in children

As presented, there is good evidence that DFA has important oral health implications. Children with DFA have an increased prevalence of untreated dental caries (Townend et al. 2000; Coxon et al. 2019a). Moreover, children can enter a cycle whereby multiple factors perpetuate disparities in oral health between children with and without DFA (Scottish Dental Clinical Effectiveness Programme 2018). These include children with DFA not being brought to dental appointments, demonstrating behaviours that disrupt the provision of dental treatment, being less likely to have investigations and treatments to identify and prevent dental caries, but being more likely to have treatments for extensive dental caries, or receiving no dental treatment at all (Klingberg et al. 1994b; Ridell et al. 2007; Nuttall et al. 2008; Humphris and Zhou 2014; Seligman et al. 2017). Therefore evidence suggests that DFA is not associated with an increase in dental caries prevalence per se, but it is associated with an increase in caries risk status (Scottish Dental Clinical Effectiveness Programme 2018). DFA creates barriers to children receiving appropriate and timely dental care, whereby children with DFA are less likely to have prevention, diagnosis and early treatment of dental caries that is present (Seligman et al. 2017). It should also be acknowledged that without intraoral dental radiographs for caries diagnosis it is likely that dental caries prevalence in children is being underestimated (Innes et al. 2020). Longitudinal studies have demonstrated that DFA in children is a long-term condition and associated with DFA in adulthood (Thomson et al. 2009). Therefore, DFA assessment and identification is important so that children can receive an appropriate management approach, and the long-term oral health implications of DFA are prevented or mitigated (ten Berge et al. 2002a).

A deficiency in the evidence base for childhood DFA is the lack of research interest into the relationship between DFA and daily living (i.e. HRQoL/OHRQoL) in children. Of note, measures of OHRQoL assess the oral symptoms, functional limitations, and social and emotional well-being of oral conditions on children's daily lives (Jokovic et al. 2004). As DFA is principally a psychological condition and not an oral condition, measures of OHRQoL may fail to explore its full range of impacts. Consequently, the use of generic HRQoL measures should be considered. Moreover, there is currently no reference standard to identify individuals with clinically important DFA, or agreement on how clinical importance is determined (e.g. impacts on the child, impacts on the dental treatment a child receives). Incorporating measures of HRQoL/OHRQoL, could add to the meaningfulness of DFA clinical assessment (Silverman and Ollendick 2005). For example, to determine threshold values based on the impacts on daily living, or to determine whether a treatment approach for DFA has improved children's daily life (Silverman and Ollendick 2005).

2.5. Acquisition and maintenance of dental fear and anxiety in children

As presented, DFA is prevalent in children, associated with poor outcomes for oral health and significant impacts on daily living, and is a condition that can endure from childhood into adulthood. Therefore, it is important to understand how it develops and is being maintained in children over time (Porritt et al. 2013). To date, there is no single mechanism to explain how DFA develops and is maintained in children (Porritt et al. 2012). Weiner and co-authors (1986) proposed that DFA can be acquired broadly by two mechanisms: exogenous and endogenous. In the exogenous category, DFA is acquired through learning experiences, whilst endogenous acquisition refers to dispositional factors, described as a constitutional vulnerability to anxiety disorders (Locker et al. 1999). However, current research suggests DFA is a complex, multidimensional construct, with interrelated factors involved in its development and maintenance (Liddell and Locker 2000; ten Berge 2001; Seligman et al. 2017). In this section, factors that are involved in the development and maintenance of DFA in children will be presented.

2.5.1. The role of learning in the development of DFA in children

Research concerning the aetiology of childhood DFA has provided some support for the role of learning (behavioural) pathways. Rachman (1977) proposed that fear acquisition is based on three negative learning pathways: directly, as a result of conditioning, or indirectly, due to social learning by vicarious acquisition (observational learning) and informational acquisition.

2.5.1.1. Direct learning pathway

The original classical conditioning learning theory is based on Pavlovian principles and is derived from a series of experiments involving an infant child ('Little Albert') who was conditioned to fear a rat. Watson and Rayner (1920) demonstrated that if a neutral stimulus becomes paired with an aversive stimulus, a conditioned fear response can occur if the neutral stimulus is encountered again (i.e. the neutral stimulus has acquired the capacity to elicit fear) (Rachman 1977). The strength of fear can be determined by the number of times the association occurred, and the intensity of the experience in relation to the stimulus (Rachman 1977). The conditioned learning can generalise to the situation, or the environment, associated with the conditioned stimulus, whereby the fear response is elicited with or without the specific conditioned stimulus present (Mowrer and Lamoreaux 1951). Therefore, if an individual has a negative or traumatic dental experience, they will learn to associate aspects of the dental situation (e.g. smell, stimuli, the dentist, dental treatment) with the negative experience, and a fear response will be developed when they are presented with that dental situation/stimuli again (Buchanan 2012). Extending the theory, Mowrer (1951) demonstrated that fear learning can be reinforced by experiencing a reduction in fear by avoidance of the conditioned stimulus. Safety behaviours (avoidance, escape) provide temporary relief from distressing fear and anxiety responses, but whilst these can produce short term reductions in anxiety, these behaviours can be a major contributor to the longer term maintenance of fear and anxiety, because the individual does not acquire positive experiences that can actually demonstrate to them they are able to effectively able to cope with the fear provoking stimuli (Rachman et al. 2008).

As evidence to support the conditioning pathway children and adults with DFA often recall negative dental experiences (Berggren et al. 1997; Lin et al. 2014). De Jongh and co-authors (2003) suggested that difficult dental encounters could be divided into four categories: pain or feelings of helplessness; behaviour or personality of the dental professional; serious treatment failures or clinical errors; and feelings of embarrassment. Locker and co-authors (1999a) identified that a majority of adults (75%, n=1065) had painful experiences, whilst 1 in 7 adults (13%, n=185) had embarrassing experiences. Adults also report that their memories of previous traumatic experiences return to them during subsequent dental visits (De Jongh et al. 2003). Similarly, children aged 7 to 14 years with DFA recount three times more negative dental visits than children without DFA (Townend et al. 2000). Potentially negative dental experiences in children appear common, with 36% (n=95) of a randomly selected sample of Swedish children aged 15 years reporting having experienced pain during their last dental visit (Stenebrand et al. 2013), and 11% and 62% of children (n=1564) from schools in Singapore reporting they had been hurt, or were made to feel guilty by the dental team about how they looked after their teeth during their most recent dental visit, respectively (Milgrom et al. 1992).

Negative learning associated with invasive medical and surgical experiences has also been implicated in the acquisition of DFA in children (Karjalainen et al. 2003). In one prospective study simple medical procedures such as getting nose-drops or having a suppository, were found to be associated with DFA in children aged 3 years (Klaassen et al. 2002). Similarly having a history of medical conditions (e.g. recurrent middle ear infections, pharyngitis, sinusitis, juvenile diabetes) during childhood (from age 3 years) was associated with DFA at 9 years (Karjalainen et al. 2003). However, overall the evidence is conflicting. Some children who have been exposed to frequent medical or surgical interventions, such as for treatment of cleft lip and palate or congenital heart disease, have been shown to have a higher level of DFA, whilst no difference has been found in childhood cancer survivors, children born extremely or very preterm, or for children with haemophilia (Wogelius et al. 2009; Brogardh-Roth et al. 2010; Vogels et al. 2011; Dogan et al. 2013; Hollis et al. 2015). A possible explanation is that medical and surgical experiences may generalise to the dental setting. For example, fear and anxiety that is acquired due to direct conditioning following negative experiences of medical and surgical treatment could be transferred when a child requires similar dental treatment (Karjalainen et al. 2003). Conversely, treatments required for other conditions may provide positive learning experiences and promote the development of helpful coping strategies in children, which may also then be used for dental care (Wogelius et al. 2009).

Although there is evidence to support direct conditioning as important in DFA development in children, retrospective reports of past trauma are subjective, and it is known that memories are not always accurate. A sample of adults with and without DFA were asked to rate their pain experience after dental treatment (Kent 1985). Immediately after the treatment had finished it was found that the groups had similar scores. However, three months later when asked to remember their pain again, the group with DFA reported more severe pain than they had described at the time. There is also evidence that memory biases of pain experienced by children are mediated by levels of anxiety, whereby children who experience intense anxiety with a painful event are more likely to recall a higher level of pain than the pain they reported experiencing at the time (Noel et al. 2012). However, Townend and co-authors (2000) and ten Berge and co-authors (2002a) have suggested that actual experiences have a minor role in the acquisition of DFA in children, and it is the perception of having experienced a negative dental visit, rather than the objective treatment experience itself, that is important.

2.5.1.2. Indirect learning pathways

To date, DFA in children has most often been attributed to the direct conditioning pathway with less evidence to describe the role of indirect social learning pathways for fear acquisition (Locker et al. 1999a; Townend et al. 2000). However, from an evolutionary perspective, it is social learning that is considered of greater importance for human survival when compared to the potential hazards of classic conditioning (trial-and-error) learning (Bandura 1965).

Social learning encompasses two pathways: children acquire DFA vicariously by observing DFA responses in another individual (the model), or as a result of what they have read, seen or heard from parents, family members, peers, teachers, or the media (Townend et al. 2000).

The mechanism for fear acquisition through observation can be conceptualised as a form of conditioning whereby a neutral stimulus is paired with the negative response of the model (Askew and Field 2008). Parents are considered the most likely candidate from whom DFA is learnt by children. Themessl-Huber and co-authors (2010) conducted a systematic review and meta-analysis of English and German language studies published between 1968 and 2007 to investigate the relationship between child and parent DFA. In total, 43 papers involving children aged 2 to 19 years met the inclusion criteria. Most studies (n=38) were conducted in Europe and North America. The findings suggest that if children are aged under 8 years, parental anxiety plays a significant role in child DFA. However, for older children the evidence was less clear. Also, established assessment measures were used by only 30 included studies.

Vicarious learning requires the observation of fear responses in another individual. Townend and co-authors (2000) demonstrated that children are poor at recognising DFA in their mothers in a study involving 60 patients aged 7 to 14 years attending a U.K. Dental Hospital. Clinical observation was used to assess DFA in participants, with severity rated on a Visual Analogue Scale (VAS). Maternal behaviours were rated using a modified Dydadic Prestessor Interaction Scale, which is an observation based measure (Bush et al. 1986). It contains seven items representing dimensions of parenting behaviour which relate to mother-child interaction in a dental environment identified during a pilot trial (fine motor agitation, ignoring, empathetic comment, empathetic gesturing, humorous gesturing, information to the child, and information to the dentist) (Townend et al. 2000). It was found that mothers of children with DFA were more likely to behave in an obviously agitated manner, ignore their child, and make empathetic gestures during a dental visit than mothers of children without DFA. However, when participants were asked to rate their mother's worry on a ten-point visual analogue scale (0=not worried; 10=very worried) the modal response was 1.0, with no significant differences for children with and without DFA. This suggests the children did not seem to notice the maternal distress and agitation, and the majority believed their mother was not anxious at all.

Less evidence is available to support the informational pathway in DFA acquisition in children. Crego and co-authors (2013) demonstrated that children with DFA know more family members with DFA than children without DFA. Similarly, Locker and co-authors (1999a) found that half of adults (56%, n=62) who report child-onset DFA had a parent or sibling also with DFA. However, these children could potentially have been subjected to vicarious learning experiences and/or negative information. A study of videos relating to DFA posted online, however, did find that children with DFA described that their friends had given them the expectation that dental care would be traumatic (Gao et al. 2013). The role of social media as an information source for the development of DFA in not known.

2.5.1.3. Threat value of stimuli in the dental setting

The learning pathway for fear acquisition explains how certain stimuli gain threat value (e.g. as a result of direct negative experiences, vicarious acquisition or informational transmission) (Craske 2003). The dental visit is a complex situation with many components. Correspondingly, individuals can experience fear and anxiety towards different stimuli or aspects of dental care. McNeil and Berryman (1989) suggested that fear of pain, fear of being closed in, and fear of injury form part of a general construct for DFA in young adults. Following a literature review, Oosterink and co-authors (2008) established a hierarchy of 67 different DFA provoking stimuli within the dental setting for adults. The situations rated as most likely to cause DFA were: ‘having some gum burnt away’; ‘having root canal treatment’; and ‘insufficient local anaesthetic’. Interestingly, adults both with and without DFA identify the same dental situations as causing fear and anxiety, although differ in the severity that is experienced (Gale 1972).

Children report higher levels of fears and anxieties about specific dental equipment or procedures, compared to the overall dental experience (Rantavuori et al. 2004). Children aged 12 to 15 years from Northern Jordan identified that the sight and sensation of a dental needle, sight, sound and feeling of the dental drill, and sitting in the dentist's chair were all anxiety-provoking stimuli; whilst children from Finland (12 and 15 years) and Estonia (aged 8 to 10 years) rated fear of pain, drilling, and local anaesthetic highly based on item scores for the original and modified versions of the CFSS-DS (Taani 2002; Rantavuori et al. 2004; Olak et al. 2013). Other specific negative stimuli that have been identified include the characteristic smell of Eugenol from a dental clinic, which was judged to be unpleasant by young adults (Robin et al. 1999).

Social anxiety relating to interpersonal aspects of the dental situation, such as fears and anxieties of negative evaluations by a dental professional, have also been identified by children and young adults as important (Kleinknecht et al. 1973). Taani and co-authors (2005) found that children fear negative comments and being reprimanded by dental professionals about their oral hygiene. Cognitive development may result in adolescents being at increased vulnerability to fears of social evaluation. During adolescence there is an increase in abstract reasoning as children begin to conceptualise other people's thoughts and take their perspectives (Piaget 1964). Along with this ability, a form of adolescent egocentricism develops, whereby children are more likely to evaluate themselves against 'ideal' standards and believe others are as preoccupied with their appearance and actions as they are themselves (Elkind 1967). Furthermore, Elkind (1967) suggested that during adolescence children construct and react to an imaginary audience where they are the centre and focus of attention, which can lead to feelings of self-consciousness, as any self-critical thoughts are anticipated in others at the same time.

2.5.1.4. Moderating learning experiences

As alluded to, not all children who have had a negative dental experience go on to acquire DFA. Learning experiences are dependent on contextual factors in which the learning occurs, including the intensity of the experience in relation to the stimulus, and an individual's prior history with the stimulus (Rachman 1977; Craske 2003). Davey (1992) proposed that if an individual encounters an unconditioned stimulus before it is related to a negative experience, then the individual is less likely to develop the conditioned learning which will then trigger the fear response when the stimulus is encountered again. This suggests that previous learning experiences about the stimulus have a moderating effect, a process termed latent inhibition. Clinical evidence for the latent inhibition effect in the acquisition of DFA in children comes from the findings that Dutch children aged 5 to 10 years with a dental history of previous non-invasive dental visits are less likely to experience high levels of DFA when compared to children who had earlier experience of invasive dental treatment (ten Berge et al. 2002a). This would suggest that if a child has a history of positive or neutral dental visits, then those experiences will act as a learning defence against fear acquisition (ten Berge et al. 2002a). It is possible that previous dental visits may support children to develop realistic expectations and coping skills for dental treatment (ten Berge et al. 2002a; Carrillo-Diaz et al. 2012). Additionally, frequent dental attendance may also reduce the probability of a traumatic dental experience (Seligman et al. 2017).

It is also by no-means a certainty that DFA acquired due to the direct conditioning pathway will remain problematic to an individual. It is possible to promote habituation in children through frequent exposure to the dental environment (Carrillo-Diaz et al. 2012). Rantavuori and co-authors (2002) identified that although a negative experience was a strong predictor for DFA, its effect diminished over subsequent dental visits. Similarly, other studies have demonstrated a reduction in DFA levels during successive dental visits (De Menezes Abreu et al. 2011; Ramos-Jorge et al. 2013). Krikken and co-authors (2015) demonstrated the importance of habituation to the dental environment in a prospective study involving a sample of Dutch children (n=200) with cleft lip and palate. Participants completed the CFSS-DS, to assess DFA at the start of the study (T1; age range 4 to 18 years) and again 36 months later (T2; age range 7 to 21 years). Data were available for 102 participants at both time points. DFA was found to be significantly lower at T2 (mean total CFSS-DS score=24.7) compared to T1 (mean total CFSS-DS score=23.2) for the study population. It was identified that the decrease in DFA scores could be largely attributed to a reduction in mean scores for CFSS-DS items relating to routine dental care (e.g. having somebody put instruments in your mouth) (Krikken et al. 2015). This suggests learning following a negative experience is not indelible, and further positive or neutral learning experiences aided recovery from DFA in children.

2.5.2. Genetic factors

Learning theories cannot fully account for the selective nature of objects and situations that elicit phobia, and the difficulties in addressing phobias as compared to conditioned fears. For example, conditioning theory suggests that the nature of the neutral stimulus is irrelevant, and that any stimulus can become conditioned if paired to a negative experience (Seligman 1971). However, phobias are largely selective and, according to Seligman (1971) related to survival of the human species through evolution (e.g. dangerous predators), rather than contemporary threats (e.g. guns) (Mineka and Öhman 2002). Consequently, phobias are 'prepared' to be learnt by humans and are easily acquired. The preparedness theory accounts for the role of conditioning by suggesting that conditioning still needs to occur, but as humans are biologically primed there is a reduction in the number of times the conditioning association needs to occur, and the intensity of the experience in relation to the stimulus, for a phobia to be learnt. Therefore, the preparedness theory proposes genetic transmission; that humans have evolved an inherited predisposition to be 'prepared' to acquire phobias to certain stimuli (Seligman 1971). Genetic factors for phobias have likely been successful during evolution because they promote survival (Kendler et al. 2008). Although, DFA is generally considered to be an acquired, the preparedness theory suggests a genetic basis for DFA, with many dental stimuli (e.g. pain, blood, and injections) considered as ‘prepared’ factors (Bracha et al. 2006; Willumsen et al. 2013).

Twin studies support a genetic contribution to the development of anxiety disorders, whereby monozygotic twins are more strongly concordant than dizygotic twins (Torgersen 1983). Ray and co-authors (2010) investigated concordance for DFA between monozygotic and dizygotic twins in a longitudinal study. The 1480 paired participants were part of a Swedish twin study on child development. DFA (present; not present) was assessed using a questionnaire based on DSM IV criteria for dental phobia (American Psychiatric Association 1994). DFA and Dental Fear Intensity (0=no fear; 10=very afraid) was measured at two points when the participants were aged 13 to 14 years and 16 to 17 years. The prevalence of DFA at the first and second time points was 5% and 4% for male participants, and 8% and 7% for female participants. The authors found that for a female the risk of having DFA if your sibling did was higher for monozygotic than dizygotic twins, but that this was not the case for males, suggesting the hereditability of DFA is stronger in females than males. Interestingly, hereditability was not stable and decreased between the time points. For both male and female participants, Dental Fear Intensity was highly correlated in monozygotic twins, but not correlated in dizygotic twins. Therefore, there may be a genetic predisposition or susceptibility to DFA that explains why monozygotic twins experience DFA at similar levels, but dizygotic twins do not. A consideration to the findings was that DFA was identified based on a criterion for dental phobia. This would identify only participants with highly problematic DFA, and potentially identify participants with less severe DFA as not having DFA at all. Regardless, the evidence would support further studies to consider the possibility of a genetic vulnerability for DFA.

2.5.3. Cognitions, and factors that influence cognitions, in DFA development

As presented, there is evidence to support learning pathways in the development of DFA in children, with the research to date suggesting direct conditioning experiences may be of greatest importance, with less evidence for the role of indirect learning pathways (Seligman et al. 2017). The pattern of learning is also significant with previous positive and neutral dental experiences acting as a moderator of DFA development in children. However, what constitutes a negative dental experience is subjective, and it is a child's perception of that experience that is crucial (Townend et al. 2000; ten Berge et al. 2002a). That is, a dental experience that is perceived by one child as negative, and for them can act as a DFA direct conditioning event, may not be perceived the same way by another child (Seligman et al. 2017). Therefore, a key consideration are the factors that can influence how a child perceives a dental experience, and the individual differences between those children that do, and do not, acquire DFA (ten Berge et al. 2002a). In this section cognitive, temperamental, psychological, and coping factors will be presented.

2.5.3.1. Cognitions in dental fear and anxiety development in children

Beck and Clark (1997) placed cognitive processes at the centre of anxiety disorders. Similarly, Armfield (2010c) has proposed that key to DFA acquisition are cognitions, rather than learned experiences. Cognitive theories consider that components of the cognitive system mediate affective, physiological, and behavioural responses associated with fear and anxiety (Beck and Clark 1988). That is, what an individual thinks affects their emotional and physiological states and alters their behaviours (Williams and Garland 2002). Cognitive components include cognitive structures or schemata, cognitive distortions, and cognitive products. Schemata are internal (mental) frameworks, representing a body of knowledge and life experience, which act to aid interpretation of the world by guiding judgements (Martin et al. 2007). Although some schemata are innate, most develop based on genetic factors and learning experiences (Birney and Sternberg 2011). Negative life experiences lead to the formation of schemata that incorporate negatively biased beliefs and lead to biased interpretations of situations (Beck and Haigh 2014). Characteristically, negatively biased schemata are relatively impenetrable to new information (Beck and Haigh 2014). The second components are cognitive distortions, defined as cognitive processes that lead to distorted interpretations of stimulus information (Beck and Clark 1988). For example, overgeneralisation, where a single event is considered as representative of others, and catastrophizing, where there is an exaggeration of possible negative outcomes. Finally, cognitive products (e.g. thoughts, images) are the outcomes of cognitive processes. During anxiety negative thoughts, images, and self-talk can become pervasive and intrusive. The foundation of cognitive theories for anxiety is that there is an overestimation of threat and danger, and an underestimation of personal ability to cope with the situation (Beck et al. 2005). These negative cognitions may have a causal and maintaining role in anxiety disorders (e.g. DFA).

To date, little is known about the cognitions related to childhood DFA. Brown and co-authors (1986) identified that 64% of children (n=487) aged 8 to 16 years reported experiencing catastrophizing thoughts when asked about receiving a local anaesthetic injection for dental treatment, although the number decreased with increasing age. To explore whether anxious children think differently to non-anxious children, Prins (1985) conducted interviews with 40 children from the Netherlands, aged 8 to 12 years, who were due to have dental treatment in the paediatric department of a university teaching hospital, and invited them to discuss their self-talk (internal dialogue accessible to conscious awareness) in relation to dental scenarios (e.g. on the way to the dentist, lying in the dental chair). The study population included children with DFA (n=20) and children with no/low levels of DFA (n=20). DFA was assessed using a 5-point Likert scale (score≥4, dentist-reported). Negative cognitions were significantly associated with high levels of DFA, where children reported thoughts about pain, difficult encounters, and escaping their appointment. In comparison, children with low levels of DFA had more variable self-talk patterns that included negative and positive cognitions about aspects of the dental encounter, such as missing school, and the benefits of dental care. Admittedly, both research studies were published 35 years ago, and therefore the cognitions reported by the child participants in these studies may not represent the DFA cognitions and experiences of contemporary children.

More recently, Carillo-Diaz and co-authors (2012) explored the relationship between treatment experiences and negative cognitions in a convenience sample of children (n=147) who attended a University Dental Clinic in Spain aged between 8 and 16 years. Generally, the study population had low levels of DFA, with only 20 children identified with high DFA (MDAS: faces version, score≥19). Participants recorded their cognitions based on the negative scale of an earlier measure developed by Kent (1985a) to explore negative and positive cognitions in adults. Children recorded the perceived likelihood and averseness of four negative situations (e.g. 'On today's visit the dentist will be very critical of the way you look after your teeth'). As confirmation of the role of negative cognitions, higher scores for likelihood and averseness of negative dental situations were associated with higher DFA levels. However, the relationship with treatment experiences was more complicated. Children who had experience of dental fillings had lower expectations of having a negative dental experience, whilst children who had dental extraction experience had increased negative cognitions about future dental visits, suggesting the nature of the dental visit is important. This finding is in keeping with previous research that identified a difference in the perceived invasiveness of restorations and extractions (ten Berge et al. 2002a).

2.5.3.2. Cognitive Vulnerability Model

Armfield (2006; 2008) has proposed a cognitive model for DFA development. It is based on theories of information-processing (Beck and Clark 1997). Key to the Cognitive Vulnerability Model (CVM) is a theorised schema for vulnerability. This contains perceptions that a dental stimulus is dangerous, unpredictable, uncontrollable, and disgusting. According to the CVM, on encountering a dental stimulus (e.g. negative dental experience) there is immediate autonomic activation of a fear response (i.e. flight, fight). At the same time there is a cognitive evaluation of the perceived dangerousness, disgustingness, unpredictability, and uncontrollability of the dental stimulus. The cognitive evaluation is influenced by perceived coping skills. If the dental stimulus is perceived as being dangerous, disgusting, unpredictable and uncontrollable, coupled with a perceived inability to cope with the situation, the vulnerability schema is activated, and leads to a sense of vulnerability. This drives the cognitive, physiological, and behavioural responses associated with DFA.

The inclusion of perceptions of danger, predictability and controllability in the CVM are unsurprising as they all key to concepts of fear and anxiety (Bandura 1988; Barlow 2002; Beck 2005). As previously presented, a fear response occurs when there is the perception of an imminent threat, whilst anxiety is the response to potential threats, but where there is no immediate danger present (National Institute of Mental Health 2011). Perceptions of unpredictability and uncontrollability are important in models that characterise anxiety as a state of helplessness (Barlow 2002). Unpredictability describes a lack of knowledge concerning some aspect of a situation (Miller 1980). Individuals use learning from past experiences and information they have acquired, to prepare for an experience, and increase the likelihood of a desired outcome (Grupe and Nitschke 2013). Correspondingly, if a situation is perceived as unpredictable it diminishes the opportunity for preparation (Grupe and Nitschke 2013). Generally, predictability is preferred over unpredictability (Miller 1980). However, there are differences in how individuals perceive the predictability of a situation that can influence fear and anxiety responding (Zvolensky et al. 2000). Armfield and Mattiske (1996) have suggested that those who perceive a situation as unpredictable are more likely to experience fear and anxiety than those who perceives it a predictable. Control is described as the ability to influence personally salient events and outcomes (Armfield 2006). In social-cognitive theory, self-efficacy to control potentially dangerous situations influences anxiety (Bandura 1988). For example, people who perceive they can control a potentially threatening experience have reduced anxiety. Generally, a sense of control can be achieved cognitively or behaviourally (Bandura 1988). However, what is important is the perception of control, rather than the cognitive or behavioural action (Bandura 1988). Logan and co-authors (1991) determined that DFA is associated with a high desire for control, but a low perception of actually being in control. Additionally, the combination of painful dental experiences and the perception of uncontrollability has been found to result in a 14 times increased likelihood of DFA in children aged 13 to 15 years from Singapore (Milgrom et al. 1992). However, if an individual perceives they have control over a stressor their DFA is reduced (Thompson 1981). Unpredictability and uncontrollability are not independent constructs. For example, increased control and being able to do something about a stressor, would increase predictability, whereas increased predictability and knowing more about a stressor, does not necessarily result in increased control (Miller 1980). Recently, there has been an interest in the role of an additional emotion, disgust, alongside fear and anxiety, in anxiety disorders (Armfield 2006). Disgust is the emotional response towards potential contamination (Cisler et al. 2009). It is characterised by cognitions related to the threat of contamination or disease, deceleration in heart rate, and avoidance of the disgusting stimulus (Cisler et al. 2009). There is evidence that disgust contributes to spider phobia and blood injury injection phobia (Cisler et al. 2009).

To explore the applicability of the CVM in children, Crego and co-authors (2013) explored the relationship between DFA and perceptions of dangerousness, disgustingness, unpredictability, and uncontrollability in a representative sample of Spanish children (n=185) aged between 8 and 18 years, and their mothers (n=97) and fathers (n=88). To assess cognitions related to CVM ('I feel trapped or helpless when sitting in the dental chair'; 'Being at the dentist's will turn my stomach') participants completed a 12-item measure. This was based on an adult questionnaire that was adapted for children in the study (Armfield et al. 2008). A four-point Likert agreement scale was used as the response format (1=absolutely disagree; 4=completely agree). A total score was obtained by summing the scores for individual items (range 12 to 48), with higher scores suggesting increased Cognitive Vulnerability (CV) associated with DFA. To assess DFA children and adults both completed the MDAS (faces version and non-faces version, respectively). Children were not dichotomised into DFA and no DFA groups, and the proportion of children identified with high levels of DFA was not reported. Overall, the children in the study population had low levels of DFA (mean=11.0; S.D.=4.9). CV scores were significantly correlated with DFA scores, with adolescents (aged 13 to 18 years) having the strongest correlation. Children's CV score was correlated with that of their mother and father. Moreover, DFA score in children aged 8-13 years was correlated to CV score in their mother and father. Regression analysis demonstrated that CV score in children was also the strongest predictor for DFA score (compared to age, sex, previous negative experience, negative affectivity, and number of fearful relatives). Perceptions of dangerousness, disgustingness and uncontrollability were identified as independent predictors of DFA, although unpredictability was not significantly associated with DFA. Similarly, Armfield and co-authors (2008) also found that unpredictability did not have an independent relationship with DFA in adults, suggesting this was due to high collinearity with the other perceptions in the CVM. It should also be noted this study does not explore causality and the role of the CVM in DFA development, although the findings do support a relationship between CVM cognitions and DFA in children.

Further research involving the same study population considered the applicability of the CVM to DFA. Children completed two questionnaires: Index of Dental Anxiety and Fear core module (IDAF-4C+), with minor changes to wording for children, and the 12-items measure for CVM perceptions described above. The IADF-4C+ contains eight items relating to the affective, cognitive, behavioural, and physiological components of DFA response (Wide Boman et al. 2015). Each item has a five-point Likert agreement response scale (1=disagree, 2=strongly disagree). A DFA score is determined by the mean score across the eight items (i.e. range=1 to 5). The scores for DFA and cognitive vulnerability were calculated from the mean score for the respective items. Participants also rated four negative dental situations (e.g. being criticised by the dentist) for their perceived likelihood of each negative situation occurring and its perceived aversiveness. In the study populations low levels of DFA (7%, n=16, IADF-4C≥3) were identified. DFA was found to be significantly positively correlated with cognitive vulnerability cognitions, perceived probability of negative events, and aversive expectations in children. Multiple regression analysis demonstrated that these three factors were also significant predicators of DFA. Moreover, if negative dental situations were determined to be both highly likely and aversive it was significantly associated with DFA and greater perceptions of cognitive vulnerability.

2.5.3.3. Temperamental factors

Temperament emerges shortly after birth and is defined as the constitutionally-based differences in behavioural characteristics between individuals that are relatively stable across different situations and over time (Birney and Sternberg 2011). It has likely genetic and environmental mediators (Rapee 2002). Childhood temperament is an important contributor to the foundation of personality in adults (Goldsmith et al. 1987), with theorists proposing a substantial overlap between temperamental traits and the five personality traits that are suggested to describe individual differences in all adults (extraversion, agreeableness, conscientiousness, emotional stability/neuroticism and openness to experience) (Lonigan et al. 2011). Temperament as a construct is considered multidimensional (Lonigan et al. 2011). Generally, there is agreement that any conceptualisation would include features of emotionality (positive and negative affect), sociability (preference for being with others rather than alone) and activity (preferred levels of activity) (Rutter 1987). However, some theorists argue for a model of temperament that it is based on individual differences in reactivity (negative affectivity and extraversion) and self-regulation (effortful control) (Goldsmith et al. 1987). Whilst other theorists have proposed a fourth temperament: impulsivity, the tendency towards impatience and lack of perseverance (Goldsmith et al. 1987). Additionally, some conceptualisations consider the traits of shyness and sociability as distinct (Mathiesen and Tambs 1999). Shyness refers to feeling anxious and distressed, and behaving in an inhibited manner when confronted with strangers and unfamiliar people, with a tendency towards avoiding social interactions (Mathiesen and Tambs 1999).

Kagan and co-authors (1999) followed healthy children from age 4 months to 7 years to explore the role of temperament on the development of anxiety. At age 4 months (n=462) the infants were classified based on motor activity and frequency of crying and fretting into temperamental categories that included: high reactive (vigorous motor activity, high frequency of fretting and crying); and low reactive (low motor activity and low frequency of fretting and crying) (Kagan et al. 1999). Overall 22% of infants were high reactive and 40%

were low reactive (the remaining 38% of infants were classified in other temperamental groups) (Kagan et al. 1998). At age 14 and 21 months, children from the high reactive infant temperamental category were more likely to show high levels of fear to unfamiliar situations, objects and people (behavioural inhibition e.g. avoidance) than children from the other temperamental categories (Kagan et al. 1998). By age 7 years, the study population had decreased to 164 children. Children who had been classified as highly reactive infants were more likely to demonstrated symptoms of anxiety (n=23, 45%) during laboratory tests (e.g. subdued when interacting with an unfamiliar adult) than children from the low reactive group (n=9, 15%). This suggests that a highly reactive infant temperament, whereby many of whom also showed an inhibited profile at age two years, is a predictor for children at an increased risk of developing anxiety in childhood. However, most children (n=27, 53%) in the high reactive category did not have anxiety symptoms.

Klingberg and Broberg (1998) investigated the relationship between temperament and DFA in a sample of 124 children aged 5 to 7 years and 10 to 12 years from Sweden. The children represented a sample taken from a large cross-sectional study (Klingberg et al. 1994a). The sampling procedure had been carried out to select children to participate in a study to validate a pictorial measure for dental fear (Klingberg et al. 1995b). Within the sample group 21% were identified with DFA (CFSS-DS, parent version, score≥38). The parents of the participants also completed the Emotional, Activity and Shyness Scale of Child Temperament (EAS). This contains 20 items based on the temperamental dimensions of: emotionality; activity; shyness and sociability (Mathiesen and Tambs 1999). A clinician also independently rated shyness by observing behaviours. Children with DFA had significantly higher mean negative emotionality scores and shyness scores than other children. Negative emotionality refers to a tendency towards distress in an infant, and fear, anger and becoming upset easily in childhood (Gustafsson et al. 2010b). It can result in refusal behaviours (Gustafsson et al. 2010b). Negative emotionality also correlates with DFA and generalised anxiety and depression in adult populations, suggesting it has a role in levels of general psychological distress (Lundgren et al. 2007). Arnup and co-authors (2007) conducted a similar study, also based in Sweden, but compared children (n=50) aged 8 to 12 years who had been referred to a specialist paediatric dentistry clinic with behavioural management problems, to a control group of children (n=117) attending for routine follow-up in a public health dental clinic. To assess temperament, the EAS-I survey (parental-response) was employed. As well as measuring emotionality, activity, and shyness, it includes an additional five items to measure impulsivity. As with DFA, children referred with BMPs also showed significantly higher mean negative emotionality scores than children in the control group (Klingberg and Broberg 1998). As there is an overlap between DFA and BMPs it is worth noting that children in the study group also had significantly higher mean DFA scores (CFSS-DS, parental version, score≥38) compared to the control group. Mean scores for impulsivity were also significantly different, but there were no differences for activity, shyness, or sociability. Impulsivity is associated with a lack of patience, so it is not surprising that children with BMPs in the dental setting would have higher scores than those in the control group. Therefore, these studies suggest that temperamental features may have a role in the development of DFA and BMPs in some children. Both children with DFA and BMPs expressed negative emotionality, but children with DFA were more likely to be shy, whilst children with BMPS demonstrated impulsivity.

2.5.3.4. Psychological difficulties

Anxiety and depression are common in children in the U.K. (Ford et al. 2003). Findings from a national survey of child mental health, involving 9117 children aged 2 to 19 years, found that overall 8% of children aged 5 to 19 years had an emotional disorder (Health and Social Care Information Centre 2018). Stenebrand and co-authors (2013) evaluated the relationship between DFA (DFS, score≥60) and general anxiety and depression in a representative sample of children aged 15 years (n=263) from southern Sweden. The study population had a low prevalence of DFA (7%; n=14). Participants completed the Hospital Anxiety and Depression Scale (HADS) to identify those with or without clinically significant anxiety and depression. The HADS contains 14 items, divided into two subscales, relating to the cognitive manifestations and emotional symptoms of general anxiety and depression (White et al.

1999). Overall, there was a significant difference in the mean general anxiety symptom score for participants with DFA compared to those without DFA. Moreover, logistic regression analysis demonstrated that general anxiety predicted DFA, whereas depression was not predictive.

Locker and co-authors (2001a) compared the prevalence of psychological disorders in young adults with or without DFA (n=1037) who were part of the Dunedin birth cohort study in New Zealand. DFA was identified in 13% of participants (CDAS, score≥13). Overall 44% of the participants had one or more psychological disorders, with 22% diagnosed with an anxiety disorder and 17% with a mood disorder. Participants with DFA were more likely to have a psychological disorder than those without DFA. This was mostly accounted for by participants (n=36) with high levels of DFA (CDAS score≥15) whereby nearly three-quarters (73%) of the participants had a least one diagnosed psychological disorder. Amongst those with high DFA, 59% had anxiety or a mood disorder and 19% had anxiety and a mood disorder. However, most of the participants with a psychological disorder did not have DFA. Although participants with DFA who had a psychological disorder at 18 years were also more likely to have DFA at 26 years, anxiety disorders did not predict outcome.

The association between levels of DFA and psychological difficulties was also investigated in a convenience sample of 128 children aged 4 to 11 years receiving dental treatment in a specialist paediatric dentistry clinic in the Netherlands (Versloot et al. 2008). The parent version of the Strengths and Difficulties Questionnaire (SDQ) was used to evaluate psychological variables. The SDQ is a measure of social, emotional, and behavioural function in children aged 4 to 17 years (Goodman 2001). It is available as parent or teacher versions for children aged 4 to 17 years, and a self-report version for children over 11 years old (Goodman 2001). It has been used for: screening; clinical assessment; as an outcome measure; and as a research tool (Goodman 2001). The SDQ asks questions relating to 25 positive and negative psychological attributes divided between five scales (emotional symptoms, conduct problems, hyperactivity, peer relationship problems and prosocial behaviour) with a three-point response scale for agreement. The total scores for each subscale, excluding prosocial behaviour, are combined to give a total difficulties score. A validation study with children aged 5 to 15 years in the U.K. demonstrated that scores above the 90th centile of a community sample predicted a raised probability of a psychiatric disorder (OR=15.7 for parent version and OR=6.2 for self-report version) (Goodman 2001). The measure was also shown to have satisfactory reliability (Goodman 2001). Overall, parents reported that 35% of children in the sample had DFA (CFSS-DS, parent-report, score≥32), whilst 9% had an SDQ total difficulties score suggesting a raised possibility of a psychiatric disorder. Subscale specific scores suggesting a clinical disorder were reported as 6%, 13%, 6%, and 9% for emotional symptoms, conduct problems, hyperactivity, and peer relationship problems, respectively. A significant correlation was found between DFA score and the total difficulties score, emotional problems score and peer problems. This suggests that children with higher scores for DFA had lower levels of psychological functioning and more social and peer problems. However, due to the design of this study the research does not provide insight into the nature of the relationship between these factors.

Although there has been little research interest into psychological comorbidities in children with DFA, studies with adults have indicated that individuals with psychological conditions are more likely to have severe levels of DFA, increased prevalence of dental extractions, be more difficult to treat, and have poorer treatment outcomes (Makkes et al. 1987; Aartman et al. 1999; Locker et al. 2001a). Due to the cross-sectional nature of studies evaluating DFA and psychological conditions, it is difficult to know what condition develops first (Locker et al. 2001a). That is, whether psychological difficulties contributed the DFA development. Regardless, psychological comorbidities may contribute to the maintenance of DFA (Locker et al. 2001a).

2.5.3.5. Coping in dental fear and anxiety development in children

Research into coping applied to dentistry considers the construct to encompass two areas: coping responses and copying styles (Buchanan 2017). Coping responses are the cognitive and behavioural strategies a child employs during a dental encounter (Buchanan 2017). Generally, behavioural coping strategies predominate in young children, with an increase in cognitive strategies occurring in line with cognitive development (Versloot et al. 2004). Coping styles refers to individual differences in preference for coping responses and is considered a relatively stable personality characteristic (Heszen-Niejodek 1997). That is, although there are a multitude of different coping responses used by children in a dental setting, those used by an individual are consistent with their dispositional coping style (Heszen-Niejodek 1997).

Coping responses can be broadly categorised as adaptive (e.g. helpful) or maladaptive (e.g. unhelpful) (Buchanan 2017). The use of different coping strategies is influenced by personal factors (cognitive development, temperament) and learning factors (previous experience, social support from parents and dental professionals to help children learn from difficulties adaptively) (Versloot et al. 2004; Krikken et al. 2015; Zimmer-Gembeck and Skinner 2016). Watson (2010) explored the strategies that children use to cope with DFA during a dental visit in a qualitative study involving a convenience sample of 54 children (aged 7 to 11 years) from a primary school in New Zealand. Five coping response themes were identified: seeking reassurance (parents, dental nurse, siblings); taking control (asking questions, asking for pain-relief, using cognitive self-control to stop negative cognitions); escape (crying, tantrums, hiding, being aggressive, employing diversionary thinking); physical interventions (tensing body, gripping arm rests, closing eyes); and passivity (magical thinking/wishing). However, there has been relatively little research into the coping strategies employed by children with high levels of DFA. The available evidence suggests DFA interferes with adaptive coping and potentiates maladaptive responses, whereby children with DFA have a propensity towards unhelpful coping responses in dental situations (e.g. closing mouth, getting angry with the dentist) (Van Meurs et al. 2005). Correspondingly, there is evidence that children with DFA are more likely to refuse dental treatment (Humphris and Zhou 2014). The way a child responds when faced with an adverse dental situation is important in the maintenance of DFA in children. That is, if a child employs a maladaptive coping response (e.g. an action that stops dental treatment) they do not have the opportunity for further learning experiences, and this can lead to the exacerbation of the DFA experienced (Zimmer-Gembeck and Skinner 2016). This is supported by evidence that children who demonstrated BMPs are more likely to have their treatment abandoned by dental professionals (Klingberg et al. 1994b). Moreover, if a maladaptive coping response is successful, at least in the short term, in achieving a reduction in DFA (e.g. if treatment is abandoned), it is more likely to be used as a coping response again and acts to maintain the DFA experienced in the longer term (Heszen-Niejodek 1997). Conversely, if a child employs an adaptive coping response to a negative dental experience it can promote further positive learning and recovery from DFA, with the development of dental resilience for future challenging dental experiences (Zimmer-Gembeck and Skinner 2016). Therefore, the learning experiences associated with coping responses act as a moderating factor for DFA in children (ten Berge 2001).

Miller (1987) has suggested there are two different coping styles for the extent of information individuals prefer about threats, monitoring (seeking out and attending to threat-related information) and blunting (avoiding threat-related information and preferring to be distracted). Generally, children experience less anxiety if interventions that are congruent with their coping style are employed (Christiano and Russ 1998). A tendency for monitoring or blunting informational coping style influences an individual's coping responses and their effectiveness (Watson 2010). A key determinant for the adaptiveness of a coping response employed is the perceived controllability of the stressor situation. In a controllable situation information-seeking is an adaptive response whereby it acts to increase predictability and facilitate change; however in uncontrollable situations information-seeking has less value, and blunting to avoid experiencing anxiety may be more effective (Miller 2015). In uncontrollable situations a high monitoring profile is associated with increased patient demands, negative cognitions (notably intrusive and repetitive thoughts about negative threat information), and reports of increased symptoms experienced and procedural and postoperative pain (Miller 1995).

To date there has been little research into the role of informational coping style as a vulnerability factor for the development of DFA in children. Miller and co-authors (1995) identified that children with a high monitoring profile had increased anxiety compared to children with a low monitoring profile in the dental setting. More recently, Campbell and Buchanan (2016) explored the monitoring and blunting informational coping style preferences of Scottish children (n=20, mean age=13.4 years) who attended a pre-sedation clinic in a dental hospital. Participants completed a new coping measure (Monitoring Blunting Communication Tool Dental) to determine how a child copes generally, and specifically with dental treatments (e.g. 'having an injection in the gum'; 'having a filling in a tooth'). To assess DFA the $MCDAS_f$ was used. This is an eight-item questionnaire that assesses severity of DFA to common dental situations (e.g. having an injection in the gum) using a five-point response scale (1=relaxed/not worried to 5=very worried). The $MCDAS_f$ is based on the MCDAS but has the Facial Images Scale (FIS) added to the response format. The FIS comprises a row of five faces (1=very happy face; 5=very unhappy face). The study population had high levels of DFA ($MCDAS_f$, mean score=30.4). Overall, children reported they were more likely to use blunting coping responses than monitoring coping responses (50%; n=10 and 30%, n=6 respectively). The remaining four children identified themselves with a mixed coping style profile. This suggests that children with DFA have monitoring, blunting, or monitoring and blunting coping styles. However, the study included a small sample size, and did not investigate which informational coping styles may be associated with better outcomes (e.g. completion of treatment, lower level of DFA).

2.5.4. Discussion of acquisition and maintenance of DFA in children

As presented, there is no single mechanism to explain how DFA develops in children. There is evidence that DFA in children often includes a direct, or, less likely, an indirect, conditioning event. That is, a dental experience that a child perceives as negative (e.g. painful or traumatic events) (ten Berge et al. 2002a; De Jongh et al. 2003). Admittedly, research into the role of negative dental experiences in childhood is mostly based on retrospective accounts provided by adults; or based on the assumed differences in invasiveness of certain dental procedures. There is also evidence that memories of pain and trauma are not always accurate (Kent 1985; Noel et al. 2012). Regardless, children with DFA often report negative dental experiences (Townend et al. 2000). Importantly, there are individual differences in perceptions of negative dental experiences, and a dental experience that is perceived by one child as negative, and for them can act as a DFA conditioning event, may not be perceived the same way by another child (Seligman et al. 2017). There is increasing evidence that it is children's perceptions about a dental experience that are crucial, with support for the idea that it is cognitions that are key to DFA development and maintenance (Armfield 2010c). The CVM has advanced this theory. In the model is a vulnerability schema containing perceptions that a dental stimulus is dangerous, unpredictable, uncontrollable, and disgusting, and the perception of being unable to cope with it (Armfield et al. 2008). Briefly alluded to, once activated, the vulnerability schema drives the cognitive, physiological, and behavioural responses associated with DFA (Armfield et al. 2008). Research to understand the negative cognitions children experience in relation to DFA, and the implications of cognitive development on how they are expressed, is needed (Alfano et al. 2002). Although learning and cognitive pathways have an important role in DFA development, there are other factors (previous learning, temperament, psychological difficulties, coping abilities, family influences) that can influence children's learning and cognitions about both themselves and the dental experience. To date, there has been little research into the complex relationships between these factors.

In children, DFA is common. Importantly, DFA traps children in a cycle of poor oral health, that can have permanent implications. Additional consideration is the associated burden to children, families and health services (Seligman et al. 2017). There is clinical potential in identifying children at risk of DFA so that a preventive approach can be implemented, and the negative implications are mitigated (Southam-Gerow and Chorpita 2010a; Youngstrom 2013).

For example, as the evidence suggests that children with psychological difficulties are more likely to have severe DFA and worse treatment outcomes, the question of whether dental professionals should also assess child mental health during routine medical screenings is raised (Makkes et al. 1987; Aartman et al. 1999; Locker et al. 2001a). Assessment of psychological difficulties in children with DFA could also enable dental professionals to instigate an appropriate child mental health referral (Kani et al. 2015). However, not all children with mental health problems have DFA, and further research is needed into the clinical usefulness of including it during DFA assessment. Key to further DFA research is DFA assessment and the ability to identify DFA in children (Seligman et al. 2017).

2.6. Assessment of dental fear and anxiety in children

Assessment is the systematic approach to measure a characteristic (e.g. DFA) in an individual (American Educational Research Association et al. 2014). Generally, assessment informs decision-making related to: screening/identification; treatment planning; and outcome measurement (Southam-Gerow and Chorpita 2010a). In addition to uses within epidemiological and research studies, assessment of DFA within clinical practice has a number of goals: (1) to quantify and understand the nature of DFA responses e.g. whether intensity or severity of DFA is outside developmentally appropriate norms, and its symptoms and functional consequences; (2) to inform the clinical encounter by promoting decision-making about appropriate and timely DFA management; (3) to promote good communication with the child and their family, and between dental professionals during patient referrals and clinical correspondence; (4) to facilitate time management planning; and (5) in the evaluation and monitoring of treatment outcomes (e.g. monitor clinical changes during a treatment course) (Armfield 2010b; Southam-Gerow and Chorpita 2010a; Buchanan 2012; Whiteside et al. 2016; Alshammasi et al. 2018). An additional purpose of clinical assessment is prevention of DFA, whereby children with risk factors for acquiring DFA are identified (Southam-Gerow and Chorpita 2010a; Youngstrom 2013).

There have been multiple reviews in the dental literature describing and evaluating the different assessment approaches for childhood DFA (Aartman et al. 1996; Aartman et al. 1998; Newton and Buck 2000; Al-Namankany et al. 2012b; Porritt et al. 2013). These methods can be broadly divided into measurement of physiological or behavioural fear and anxiety responses, and the use of formal psychometric measures (standardised questionnaires completed by a parent/carer as a proxy respondent on behalf of the child or by the child themselves). In this section, DFA assessment in children will be discussed.

2.6.1. Measurement of physiological response

The physiological responses associated with fear and anxiety facilitates the body to react mentally and physically to potentially dangerous situations (Hoehn-Saric and Mcleod 2000). Active fear behaviours (e.g. flight or fight) are characterised by increased muscle tension and broad sympathetic and parasympathetic activity. This results in: increased heart rate, contractility, and excitability (causing extra-systoles); increased blood pressure; increased respiration rate and oxygen consumption; increased sweat gland, gastrointestinal and bladder activity; and cutaneous vasoconstriction with decreased skin temperature and piloerection at base of hair follicles (Hoehn-Saric 1998; Kreibig 2010). In contrast to the physiological flight or fight responses, anxiety describes a state of readiness with physiological systems becoming prepared to facilitate escape from perceived danger. Therefore, there is similar sympathetic and parasympathetic activity, but with reduced intensity. With respect to DFA, children and young adults have reported experiencing muscle tenseness and increases in heart rate, breathing, sweating, and the sensation of increased salivation (Kleinknecht et al. 1973).

Physiological arousal is an adaptive response to acute stressors. Correspondingly, elevated physiological arousal may be expected in anxiety disorders. However, after controlling for baseline differences, there is no difference in the nature of the physiological response to acute stressors between individuals with and without an anxiety disorder, although it can take longer for baseline levels to return to resting with an anxiety disorder (Hoehn-Saric and Mcleod 2000). An exception is in the case of a specific phobia (e.g. DFA), whereby an elevated physiological response occurs when a phobic individual is exposed to a personally-relevant phobic stimulus (Hoehn-Saric and Mcleod 2000).

Blood pressure, heart rate, muscle tension, respiration rate, salivary biomarkers, skin conductance and sweat tests have all be used as indices to measure the physiological arousal associated with DFA (Aartman et al. 1996). An advantage of physiological assessment is that it is objective and not dependent on subjective reporting (Southam-Gerow and Chorpita 2010b). Although, physiological assessments have a sound theoretical basis (i.e. fear and anxiety physiological responses), a potential limitation for widespread use by the dental professional is that specialist equipment may also be required (Buchanan and Niven 2002). Recent advances in the medical use of wearable technology (e.g. smartwatches, fitness band consumer electronics) may offer a solution, whereby physiological measurements are obtained with relative ease (Buchanan and Niven 2002; Düking et al. 2020). Regardless, a significant limitation with the use of physiological readings is that measurements are non-specific, and it is not possible to attribute the results solely to DFA. For example, the equipment itself could evoke an anxiety response (Aartman et al. 1996). Consequently, physiological measures have limited clinical applications in routine DFA assessment.

2.6.2. Observation of behavioural response

Behavioural theories consider fear and anxiety as being part of a defensive motivational system that functions to activate different behavioural responses to a perceived threat (Barlow 2002). The behaviours associated with fear and anxiety correspond to ethological models of a predatory threat imminence continuum (Fanselow and Lester 1988). Responses to high imminence threats (fear) are qualitatively different to responses to low imminence threats (anxiety) (National Institute of Mental Health 2011). Along the continuum defensive behaviours are determined by the spatial distance of the prey to the predatory threat, predator and prey characteristics, and context of the predatory threat (Craske 2003; Kozlowska et al. 2015). In humans, threat imminence is also based on psychological perceptions of the threat (e.g. appraisal of coping resources) (Craske 2003). Applying the threat imminence continuum to a DFA analogy, children are the prey and the dental experience the predatory threat. The initial pre-encounter stage describes a situation of increased prey vulnerability with potential for a predator. It is characterised by alertness and vigilance, considered analogous to anxious worry, to facilitate avoidance of predators (Craske 2003). Mammals may also respond by becoming attentively immobile (Kozlowska et al. 2015). For prey, immobility reduces the likelihood of detection by a predator as the mammalian visual cortex primarily detects moving prey (Bracha 2004), but still allows for an active response if required. During the post-encounter stage (e.g. danger imminent) defensive behaviours that decrease the likelihood of contacting the threat and allow escape predominant (e.g. flight). Lastly, circa-strike describes survival behaviours following contact with the predator (e.g. fighting). There is also evidence that in the face of imminent mortal danger (e.g. sexual assault), when restraint prevents escape, tonic immobility can occur as a final step in the threat imminence continuum (Humphreys et al. 2010). This survival response is characterised by physical immobility, muscle rigidity, fixed and unfocused stare, suppressed vocalisations, and tremors in the extremities (Humphreys et al. 2010).

In childhood DFA, defensive behaviours can manifest as dental BMPs. For example, in an investigation to determine a profile of Scottish nursery school children receiving fluoride varnish applications, it was identified that children with initial anxiety (anxiety related behaviour in first 20 seconds of treatment) were more likely to: shake their head; cry; sit up; hide their face; and turn their head away (Humphris and Zhou 2014). Anxiety increased the likelihood of refusing treatment by 170 times (Humphris and Zhou 2014). However, not all DFA related behaviours are disruptive, and children's coping style and behavioural expressions vary between children and across stressful dental situations (Weinstein et al. 1996; Freeman 2007). Children with high levels of DFA have been found to be more likely to report the use of unhelpful behavioural coping strategies during dental visits (e.g. getting angry with the dentist, closing their mouths) (Van Meurs et al. 2005). Additionally dental BMPs are more common in children with high DFA levels (Wogelius et al. 2003). However, children with DFA may also present with withdrawal and passivity in the dental environment (Klingberg and Broberg 1998). Behavioural responses may be influenced by children's emotional awareness and ability to regulate their emotions in a socially appropriate manner (Rieffe et al. 2008; Gullone et al. 2010). Harris and co-authors (1986) identified that children from six years understood that expressed emotions are often tempered compared to the actual emotion experienced. A further consideration is that children with DFA have also been found to have difficulties in other behavioural areas which may influence their behaviour in the dental setting (Krikken et al. 2010).

ten Berge and co-authors (1999) evaluated emotional and behavioural problems in children aged 4 to 11 years old from the Netherlands. Children receiving dental treatment in a specialist clinic (n=203) were allocated to a DFA group, whilst children from another study (n=1172) were identified as a control group. All parents completed the emotional and behavioural problem scale of the Child Behaviour Checklist based on the previous six months (Achenbach and Ruffle 2000). The scale comprises 118 items related to nine subscales of problem behaviour including: social withdrawal; somatic complaints; anxiety/depression; social problems; thought problems (e.g. having obsessional thoughts); attentional problems; delinquent behaviour (e.g. stealing); aggressive behaviour; and sex problems. It has a three-point agreement response scale. The items are summed to provide a score for each subscale, internalising and externalising problems and a total problems score. Overall, children with DFA had significantly higher mean scores for total problems, internalising and externalising problems, and all the subscales, except for sex problems, than the community sample. Approximately, 20% of children with DFA had a total problem score that suggested a clinical problem requiring treatment (score above 98th percentile for normative population). With respect to individual subscales, between 2% and 8% of children with DFA had a score in the clinical problems range.

A number of behavioural rating scales have been described that typically assess children's level of cooperation with dental treatment (Aartman et al. 1996). Frequently reported scales include: Frankl Rating Scale (Frankl et al. 1962); Houpt Four-Point Categorical Rating Scale (Houpt et al. 1985); Global Ratings Scale (Chambers et al. 1981); and the Behaviour Profile Rating Scale (Melamed et al. 1975). The most widely used is the Frankl Rating Scale (Frankl et al. 1962; Aartman et al. 1996). The scale is completed by a person (e.g. dental professionals or researcher) observing the child. The child's behaviour is rated on a four-point scale (definitely negative, negative, positive, definitely positive) in different clinical situations (e.g. being separated from a parent, examination, dental prophylaxis, dental radiograph, departure). An overall score is obtained by summing the score for each of the clinical situations (Aartman et al. 1996). Alternatively, behaviour can be classified based on the number of positive and negative scores (Frankl et al. 1962). However, a review of behavioural ratings scales identified the Behavioural Profile Rating Scale (BPRS) as the preferred behavioural measure (Melamed et al. 1975; Aartman et al. 1996). The scale is comprised of 27 items relating to children's DFA related behaviour in the dental setting (e.g. inappropriate mouth closing). Each item is weighted depending on its perceived disruptiveness. An observer scores the frequency of each behaviour over successive three-minute periods. An item score is obtained by multiplying the weight factor of the item with its frequency of occurrence. The total score is determined by summing the item scores and dividing by the number of three-minute periods included. Although the BPRS is complicated to use and score, it has the advantage of assessing children at regular intervals and obtaining a more precise behavioural assessment.

With respect to DFA, a fundamental difficulty with using behavioural rating scales is that they do not discriminate between DFA and BMPs. Typically behavioural ratings focus on cooperativeness in the dental setting as perceived from the perspective of a parent/carer, dental professional or researcher e.g. reports of behaviours visible to others (Aartman et al. 1996). This is often in the context of the ease in which dental treatment is completed (Aartman et al. 1996). However, children with DFA can behave in different ways within the dental environment (Klingberg et al. 1994b; Wogelius et al. 2003; Freeman 2007). Additionally, much of the research on children's behaviour in the dental setting has involved young children, whereas there has been little research interest into the behaviours of adolescents (Klaassen et al. 2003). There are also multiple factors that can influence DFA related behaviour in children including temperament, coping style, emotional awareness and regulation and behavioural problems (Klingberg and Broberg 1998; ten Berge et al. 1999; Gullone et al. 2010). However, observation of children's behaviour as an assessment of DFA may be required for children who lack verbal communication skills (e.g. very young children, children with cognitive impairments) (McGrath 1987).

The available evidence suggests that dental professionals' ratings of children's behaviour correlate only moderately well with children's own self reports of their DFA (Townend et al. 2000). Similarly, there is poor agreement between assessments of problem behaviour between parents and children (Seiffge-Krenke and Kollmar 1998). Additionally, different observers may interpret children's behaviours differently, although this can be overcome through training and calibration (McGrath 1987). Consequently, children with challenging behaviour may be incorrectly diagnosed with DFA, and children with DFA who do not behave in an outwardly way, may have their DFA go unrecognised (Klaassen et al. 2003).

2.6.3. Use of psychometric measures

Youngstrom (2013) has suggested that assessment measures should address at least one of the following: prediction of the construct of interest (i.e. screening/identification); prescription (the property of informing the choice of treatment modality, identifying moderators for treatment); or process (variables that quantify meaningful treatment outcomes). Generally, measures contain items/prompts and a format to quantify the response, from which a score is determined (American Educational Research Association et al. 2014). Assessment measures should be developed using an appropriate theoretical framework for the construct of interest (e.g. DFA in children) (American Educational Research

Association et al. 2014). If assessment measures are without a clearly articulated theory they lack construct validity and may fail to measure the construct of interest (Cronbach and Meehl 1955). Correspondingly, the construct the scale is measuring should be specified e.g. the construct interpretation that will be made based on the item responses (American Educational Research Association et al. 2014). Assessment measures should have acceptable validity and reliability (Freeman 2005b). Validity refers to the degree to which evidence and theory support the interpretation of test scores for the proposed uses of the test (American Educational Research Association et al. 2014). Validity can be determined in different ways. These include: criterion validity (demonstrated by comparing the results of the measure to a 'gold standard' measure, which purports to assess the same construct of interest); concurrent validity (type of criterion validity, whereby the measure and the 'gold standard' are administered at the same time); construct validity (describes the ability of the measure to test the underlying construct of interest e.g. measure applied to two groups who are known to differ in the construct of interest); and convergent validity (supports construct validity as can be demonstrated by scores on measure being evaluated are highly correlated to scores on a test thought to measure similar or related concepts) (Roach 2006). Reliability refers to the consistency of a person's score on a measure (Mash and Hunsley 2005). It can be measured in two ways: internal consistency (whether all elements of a measure contribute in a consistent way to the data obtained); and test-retest reliability (if similar results would be obtained if an individual completed the measure a second time) (Mash and Hunsley 2005). With respect to reliability, Newton and Buck (2000) have proposed that internal consistency, as determined by Cronbach alpha, is considered satisfactory if alpha >0.6, and high if alpha >0.8, and that test-retest is satisfactory if the correlation coefficient is >0.8, and high if the correlation coefficient is >0.9.

Self-report measures (e.g. standardised questionnaires) assess the patient's perspective, whereas physiological and behavioural approaches do not provide information about the subjective experience of DFA (Rose and Devine 2014). For children, DFA assessment questionnaires may be completed by a parent/carer, or by a child themselves. However, recent studies have highlighted poor agreement between parent proxy-report and child self-reports of DFA (Gustafsson et al. 2010a; Klein et al. 2015; Patel et al. 2015). Klein and co-authors (2015) found that parents of highly dentally fearful and anxious children tended to underestimate their child's levels of DFA, whilst parents of children with low levels of DFA would overestimated it. In contrast, Gustafsson and co-authors (2010a) identified poor agreement between parents and children with DFA generally. It is more difficult for parents to appreciate the severity of emotional problems (e.g. anxiety, depression) compared to more outwardly obvious behavioural problems (Fox et al. 2008). Correspondingly, Patel and co-authors (2015) found that parents failed to recognise DFA in half of all children who identified themselves with DFA. Further evidence that children and parents are poor at identifying DFA is provided by the findings of Luoto and co-authors (2010) who asked parents-child (aged 11 to 16 years) dyads to identify whether the other person in the pair was afraid of dental care. Generally, agreement, demonstrated by Kappa values, for parents and children was poor. Moreover, parental sensitivity for correctly identifying their child with DFA was <0.39 for mothers and <0.37 for fathers, respectively. However, sensitivity increased slightly if the parent experienced DFA themselves (sensitivity=0.43-0.50). Children's sensitivity for recognising a parent with DFA was 0.38. Children with DFA also had low sensitivity for parental DFA (sensitivity=0.17-0.46). The findings for childhood DFA are consistent with those for anxiety and depression. A meta-analysis of studies published in the psychological literature between 1967 and 1985 (n=119 included studies) found the correlation between children's self-reporting and parental proxy-reporting of emotional problems to be poor (Achenbach et al. 1987). Additionally, there is evidence that distress and related factors can influence parents' perceptions of child mental health (Berg-Nielsen et al. 2003). Parent proxy-reporting is recommended if a child is too young or too unwell to complete a questionnaire themselves, or as part of multi-informant assessment to supplement child self-report assessments by providing complementary information, but not as a substitute for child self-report if a child is able to complete a measure (Christine and Morse 2001). Therefore, dental professional should not rely on parental reporting alone.

Varni and co-authors (2007) have demonstrated that children can reliably complete self-report questionnaires about their health if it is appropriate for the age range of interest. Wooley and co-authors (2004) discussed the importance of measures also demonstrating developmental validity, whereby children in the targeted age range can read and comprehend the questions and response alternatives. Additionally, children should be able to comprehend the assessment requirements (Gullone 2000). Freeman (2005b) has advocated that assessment questionnaires for children should be child-centred, and quick and easy to use.

There are multiple self-report questionnaires available to assess DFA in children (Aartman et al. 1998; Newton and Buck 2000; Al-Namankany et al. 2012b; Porritt et al. 2013). Types of self-report DFA questionnaires can be broadly categorised as state measures or trait measures. State measures assess anxiety at the time they are completed e.g. how anxious a child is feeling at a particular moment (Buchanan 2012). However, state measures can also be used at different points during a treatment episode to monitor anxiety levels, and immediately following a treatment episode to assess anxiety retrospectively (Buchanan 2012). In contrast, trait anxiety describes individual differences in proneness to experience state anxiety, and is a relatively stable personality characteristic (Spielberger 1972). Generally, trait measures for childhood DFA have required children to evaluate their severity of fear and anxiety to specific stimuli, procedures and situations, or assess their emotional responses associated with DFA (Armfield 2010a). Most measures then provide an overall DFA score by summing the scores for individual items (Armfield 2010b).

2.6.4. State measures for dental fear and anxiety assessment in children

2.6.4.1. Venham Picture Test

The Venham Picture Test (VPT) is a state measure of situational anxiety that comprises a picture selection task which has been shown to be suitable for children aged from three years (Venham and Gaulin-Kremer 1979). The use of a pictorial scale overcomes the difficulties of a numerical rating scale in young children (Howard and Freeman 2007) Children select a cartoon image from each of eight image pairs. Each paired image represents an emotional state (e.g. happy, sad, scared, crying, scared motion) associated with the dental setting. The pairs show two male children experiencing the same emotional state, but one is less anxious, and one is more anxious. The male child in the images has been designed to have a disproportionately large head, so as to draw attention to the facial expressions (Venham and Gaulin-Kremer 1979). Respondents select for each paired image the child (less anxious, more anxious) that most reflects their current emotional state. A total score is derived from the frequency that the more anxious cartoon image is selected (range 0 to 8). The conceptual framework for the selected emotional states and their relevance for DFA is children has not been provided. As the VPT assesses state anxiety, it may determine a child's 'state' at the time of the test, which it is not possible to attribute exclusively to state DFA (Porritt et al. 2013). Additionally, the images used have been criticised for: being ambiguous, as it is not always clear what emotional state is being illustrated; having only male figures, potentially alienating female users; and the images being highly stylised (Buchanan and Niven 2002). Additionally, scared motion (child running away) could be considered as behaviour. Within the dental situation, children may expect to be reprimanded if they ran away which potentially could influence their selection. The VPT has demonstrated high internal consistency (determined by Kuber-Richardson Formula 20 and Cronbach alpha) and a test-retest reliability of r=0.7 (Venham et al. 1977). Although concurrent validity has been reported by inter-scale correlations with trait measures, Aartman and co-authors (1998) identified that correlations with other state measures are required. Children were not active participants in the development of the measure (Venham and Gaulin-Kremer 1979).

2.6.4.2. Facial Images Scale

The Facial Images Scale (FIS) is a picture scale that assesses state anxiety (Buchanan and Niven 2002). It is suitable for children from age three years (Buchanan and Niven 2002). Children are asked to choose one face from a row of five faces (very happy face to very unhappy face) that best matches how they are feeling at that time. The illustrations are simple line drawings.

The scores range from 1 to 5 (1=very happy; 5=very unhappy). The FIS was found to be significantly correlated with the VPT, suggesting concurrent validity (Buchanan and Niven 2002). As the FIS is a one item state measure reliability estimates have not been provided (Alshammasi et al. 2018). The FIS has also been used as the response format within measures of trait DFA, including: the Smiley Faces Programme (Buchanan 2005); Revised Smiley Faces Programme (Buchanan 2010); Modified Child Dental Anxiety Scale faces version (Howard and Freeman 2007), and a modified version of the Corah Dental Anxiety Scale (Dogan et al. 2006). As with the VPT, the conceptual framework for the selected images, and their relevance for DFA is children has not been provided (e.g. are those the facial expressions children associate with anxiety). Nor were children included in its development. Additionally, as the FIS assesses state anxiety, it will assess a child's 'state' at the time of the test, which it is not possible to attribute exclusively to state DFA, nor inform the dental team what the child is experiencing anxiety about (Porritt et al. 2013; Alshammasi et al. 2018).

2.6.5. Trait measures for adults used for DFA assessment in children

2.6.5.1. Corah Dental Anxiety Scale

The CDAS is the most frequently used DFA measure (Armfield 2010b). It was first described in 1969, although details outlining its theoretical basis have not be published (Corah 1969). It is comprised of four scenarios: going to the dentist tomorrow; sitting in the dentist office; sitting in the dentist chair and waiting for drilling and sitting in the dentist chair waiting to have teeth scraped and polished. The first item has a different five-point response scale to the final three items. However, the second response scale is not ordinal, and has responses that are not mutually exclusive (Armfield 2010b). For example, 'tense' as point three of the scale versus 'anxious' as point four of the scale. A total score is calculated by summing the individual scores for each item. Scores range from 4 (no DFA) to 20 (most severe DFA). Corah (1969) reported high internal consistency (determined by Kuber-Richardson Formula 20), satisfactory test-retest reliability, and a significant correlation with dentist's ratings of DFA. In a review of the literature, Schuurs and Hoogstraten (1993) found that the CDAS has demonstrated high internal consistency (determined by Cronbach alpha), construct validity, and satisfactory test-retest reliability with adult populations. The psychometric properties have been reported in children, where internal consistency is high, but with a range of test-retest reliability correlations (r=0.66 to 0.95) in a small sample of children (n=40) aged 9 and 13 years (Wong et al. 1998).

2.6.5.2. Dental Anxiety Question

The Dental Anxiety Question (DAQ) is a single item questionnaire. It comprises the question, 'Are you afraid of the dentist' and a four-point response scale (1=No; 4=Yes, very) (Neverlien 1990). It has demonstrated good correlation with the CDAS in Norwegian children aged 10 to 12 years (Neverlien and Johnsen 1991). The DAQ is easy to administer, however, as there is only a single question no information is obtained about dental stimuli or situations that may trigger DFA, or factors that are acting to maintain DFA over time (Newton and Buck 2000; Porritt et al. 2013).

2.6.5.3. Dental Cognitions Questionnaire

The Dental Cognitions Questionnaire (DCQ) is a self-report measure that assesses subjective cognitive responses for adults, which has been employed with children (De Jongh et al. 1995; Mansell and Morris 2003). It comprises 38 statements relating to dental treatment in general (e.g. 'Dentists don't care when it hurts') and negative thoughts during dental treatment (e.g. 'Everything goes wrong'). The items for the questionnaire were formulated following semi-structured interviews with adults with DFA (De Jongh and Ter Horst 1993). As the item were developed with adults it is not known how relevant and pertinent, they are to children's DFA experiences. Each statement has a binary (yes/no) response format. A score is determined by summing the number of statements with a positive response (range 0 to 38). A separate believability score is determined by asking questionnaire users to record a percentage score for the strength of their belief of each statement at that moment. The DCQ has demonstrated high internal consistency, satisfactory test-retest reliability, and satisfactory concurrent and discriminate validity with adults (De Jongh et al. 1995).

2.6.5.4. The Modified Dental Anxiety Scale

The MDAS was developed to overcome the response scale problems with the CDAS. It comprises five items: four based on the scenarios in the Coral Dental Anxiety Scale; and an additional item on DFA related to receiving a local anaesthetic injection (Humphris et al. 1995). Each item is scored on a five-point response scale (1=not anxious; 5=extremely anxious). A total score is calculated by summing the individual scores for each item. Scores range from 5 (no DFA) to 25 (most severe DFA). The MDAS had demonstrated high internal consistency, high test-retest reliability, and concurrent validity (Newton and Edwards 2005). To date, the MDAS has been translated into 28 different languages (Humphris 2021). A total score of 19 has been recommended as the threshold value to identify adult individuals who require specialist intervention for DFA (Humphris et al. 1995). This threshold value has been confirmed by ROC curve analysis conducted with a sample of U.K. university undergraduates and postgraduates, where it was found a score below 19 suggests an individual is not dentally phobic (King and Humphris 2010). However, the population was identified with a low prevalence of dental phobia. The psychometric properties and threshold levels have not been determined for children.

2.6.5.5. The Index of Dental Anxiety and Fear

The Index of Dental Anxiety and Fear (IDAF) is a relatively new assessment measure of DFA for use with adults. It is comprised of three modules: Anxiety and Fear Module (IDAF-4C+); a Phobia Module; and a Stimulus Module (Armfield 2010a). The IDAF-4C+ is the core module in the measure. It contains eight items relating to the affective ('I feel afraid or fearful when visiting the dentist'), cognitive ('I think that something really bad would happen to me if I were to visit the dentist), behavioural ('I delay making appointments to go to the dentist') and physiological ('My heart beats faster when I go to the dentist') components of the DFA response (Wide Boman et al. 2015). The item content was initially generated from theoretical research, and existing self-report measures for fear and anxiety and DFA (Armfield 2010a). Each item has a five-point Likert agreement response scale (1=disagree, 2=strongly disagree). A DFA score is determined by the mean score across the eight items (i.e. range=1 to 5). The IDAF-4C+ has demonstrated high internal consistency, high test-retest reliability, and convergent validity in Australian adults (Armfield 2010a). The IDAF has been translated into multiple languages. The psychometric properties of the Turkish version of the IDAF in relation to children aged 12 to 14 years have been reported (Buldur and Armfield 2018). The Turkish version children was found to have high internal consistency, high test-retest reliability, and demonstrated good correlation with the CFSS-DS in a study with children (Buldur and Armfield 2018).

2.6.6. Trait measures for dental fear and anxiety assessment in children

2.6.6.1. Abeer Children Dental Anxiety Scale (ACDAS)

The ACDAS is a relatively new DFA questionnaire for children aged 6 to 16 years (Al-Namankany et al. 2012a). It comprises three parts. The first part consists of a self-report questionnaire to identify DFA. It contains 13 items and requires children to rate their response to different situations in the dental setting. The item content was developed from in the measure. However, some items have questionable relevance for dentistry ('Having a pinch feeling in the back of your hand'). Responses to items are provided on a three-point faces and written scale (1=happy face; "happy", 2=neutral face; "ok", 3=scared face; "scared"). A total score is determined by summing the scores for the individual items. Scores range from 13 to 39, with higher scores reflecting higher DFA severity. The second part is described as a cognitive test. Children provide dichotomous answers to three questions relating to their feelings of shyness and thoughts of losing control. The final part requires parents to rate their expectations of their child's behaviours, and the dental professional to rate their experience of the child's behaviour. However, the response scale used is based on affect rather than behaviour (e.g. happy, scared). The second and third parts of the test are not included in the score for DFA.

The ACDAS has demonstrated high internal consistency and concurrent validity as determined by inter-scale correlations with the CFSS-DS (Al-Namankany et al. 2012a). A threshold value of 26 was suggested following ROC curve analysis to discriminate between no DFA and DFA, using the threshold value for the CFSS-DS of 36 as the reference standard (Al-Namankany et al. 2012a).

2.6.6.2. Children's Experiences of Dental Anxiety Measure (CEDAM)

The CEDAM is a child-centred measure of DFA for children aged 9 to 16 years (Porritt et al. 2018). Recently, an eight-item short-form of the CEDAM (CEDAM-8) has also been developed (Porritt et al. 2021). The content of the CEDAM and CEDAM-8 was based on qualitative interviews with children with DFA that were conducted for Study 2 of this thesis (see Chapter 5). For chronological narrative purposes it is described fully in Chapter 6. However, it has been included in the literature review for completeness.

2.6.6.3. Children's Fear Survey Schedule Dental Subscale (CFSS-DS)

The most frequently used measure is the CFSS-DS (Cuthbert and Melamed 1982; Porritt et al. 2013). It is suitable for children aged 5 to 15 years (Cuthbert and Melamed 1982), and has been translated into multiple different languages. It is based on the Fear Survey Schedule for Children (Scherer and Nakamura 1968), although the development of the dental subscale has not been clearly described. Children were not involved in the development of the measure (Porritt et al. 2013). The CFSS-DS requires children to rate their level of DFA to 15 dental situations (e.g. 'having to open your mouth', 'the noise of the dentist drilling') using a five-point severity response scale (1=not afraid to 5=very afraid). Scores range from 15 (no DFA) to 75 (most severe DFA). Threshold values of between 23 and 36, determined by ROC curve analysis, have been suggested to identify clinically relevant DFA (reference standard defined as BMPs) for age (4 to 19 years) and sex subgroups of Swedish children (Gustafsson et al. 2010a). Using a similar approach, a threshold score of 37 has been identified for clinically relevant DFA in a sample of Greek children aged 6 to 12 years (Boka et al. 2017). The CFSS-DS has demonstrated high internal consistency and test-retest reliability (Alvesalo et al. 1993; Klingberg 1994; ten Berge et al. 2002c). Factor analysis (principle component) of the Finnish version identified a three factor structure accounting for 54% of the variance: (1) fear of highly invasive procedures, such as injections and drilling; (2) fear of potential victimization, including fear of strangers, choking and hospitals; and (3) fear of less invasive procedures, such as opening the mouth and being examined by the dentist (Alvesalo et al. 1993). Factor analysis (principle component) of an identical Dutch parent version identified that the items load onto a four factor structure, accounting for 60% of the variance e: (1) fear of general, less invasive dental treatment: (2) fear of medical aspects; (3) fear of drilling; and (4) fear of strangers (ten Berge et al. 2002c). Factor analysis in the British version identified a two-factor structure accounting for 64% of the variance: (1) previous dental experience; and (2) items relating to the dental examination. However, some of the items have questionable relevance for contemporary paediatric dentistry practice (e.g. level of anxiety about people in white uniforms, going to hospital). Moreover, U.K. parents have objected to an item that asks children to rate their fear of having a stranger touch them, although this finding has not been reported in studies from other countries (Al-Namankany et al. 2012a).

To overcome the limitations of the CFSS-DS, an eight-item short form of the measure has been suggested (Folayan and Otuyemi 2002). More recently, a revised Finnish version of the CFSS-DS has been developed (Rantavuori et al. 2005). This new version comprises eight of the original items (excluding the general fear items) and three new items (fear of dentistry in general, fear of suction and fear of pain). The response format was also altered to include an option for 'no experience' (score=1). It was found to have a two-factor structure: fears related to invasive dental treatment (treatment of dental decay) and fears related to dental visits in general (Rantavuori et al. 2012). Carson and Freeman (1997) also evaluated the CFSS-DS for use by dental professionals to assess child DFA and identified high levels of agreement between children and dental professionals (dental nurse and dentist) item scores.

2.6.6.4. Dental Fear Survey

The Dental Fear Survey (DFS) is a 20-item measure with five-point response scale (1=no fear/reaction to 5=great fear/reaction) (Kleinknecht et al. 1973; Kleinknecht et al. 1984). Its conceptual framework is based on learning theory, but the final questionnaire contains items that assesses DFA responses related to dental avoidance (2 items), physiological arousal (5 items), fear and anxiety provoking stimuli and dental procedures (12 items), and a global DFA item (Kleinknecht et al. 1984). The DFS was not developed with the intention of providing a DFA score, but as a tool to support dental professionals understand DFA in their patients (Kleinknecht et al. 1973). However, total scores (20 to 100) are frequently reported (Armfield 2010b). The DFS was developed for use with children aged 11 to 16 years and college students in the United States of America (Kleinknecht et al. 1973). However, children were not involved in its development. Some items (e.g. put off making a dental appointment) may not be relevant to children who are not yet responsible for their own healthcare. Internal consistency has been reported to be high (Schuurs and Hoogstraten 1993). In adult studies, the DFS has demonstrated concurrent validity as assessed by inter-scale correlations with the CDAS, and construct validity as it has been shown to discriminate between groups with DFA and groups without DFA (Johansson and Berggren 1992; Kvale et al. 1997). A threshold value of 53 for high DFA has been suggested for Brazilian young adults using a ROC curve analysis (based on binary response to the question, ‘Are you fearful of going to the dentist?’ as the reference standard) (Oliveira et al. 2015). A three-factor structure (avoidance, physiological arousal and fear associated with specific dental stimuli and procedures) has been confirmed in North America, Singapore, and Brazil (Kleinknecht et al. 1984; Milgrom et al. 1990; Cesar et al. 1993). Taani and co-authors (2005) have proposed a modified 15 item version of the DFS that excludes the items relating to physiological arousal, although its psychometric properties have not been reported.

2.6.6.5. Modified Child Dental Anxiety Scale

The MCDAS is an eight item self-report measure that assesses severity of DFA in relation to typical dental situations (e.g. having an injection in the gum, having a filling) in children aged 8 to 15 years (Wong et al. 1998). It also contains one item to assess overall DFA. It is based on the scenarios within the CDAS, and, as with the MDAS, was developed to overcome its problems, most notably inconsistency within the response format (Humphris et al. 1995). Consequently, the MCDAS has a five-point severity response scale (1=relaxed/not worried to 5=very worried) (Wong et al. 1998). The response scale was developed with children (Wong et al. 1998). Total scores range from 8 (no DFA) to 40 (most severe DFA). The MCDAS has demonstrated high internal consistency (determined by Cronbach alpha), and satisfactory concurrent validity, as assessed by inter-scale correlations with the CDAS and CFSS-DS, but with variable test-retest reliability (0.53 to 0.98) (Wong et al. 1998). High inter-scale correlation was also found with the MDAS in children aged 13 to 15 years in Kuwait (Honkala et al. 2014). To date, threshold values have not been reported. One limitation of the MCDAS is that it appears to generate a high number of incomplete questionnaires (Arch et al. 2001). One possible explanation is that children may lack an understanding of some of the included dental situations (e.g. scale and polish, inhalation sedation) (Buchanan 2005).

Howard and Freeman (2007) made improvements to the MCDAS response format by adding the FIS to the numerical scale. The faces version of the MCDAS ($MCDAS_f$) is suitable for children aged 8 to 12 years. The $MCDAS_f$ has demonstrated high internal consistency, test-retest reliability, and concurrent validity, as assessed by inter-scale correlations with the CFSS-DS (Howard and Freeman 2007). Exploratory factor analysis identified two factors: (1) examination (e.g. going to the dentist generally, having your teeth looked at); and (2) treatment (Howard and Freeman 2007). A threshold value of 26 has been reported using referral for DFA as the reference standard for ROC curve analysis (Howard and Freeman 2007). An alternative version of the questionnaire, without the items for inhalation sedation and general anaesthesia, has been recommended for use by the Scottish Dental Clinical

Effectiveness Programme (2012), although its psychometric properties have not been reported to date.

2.6.6.6. Smiley Faces Programme (SFP) and Revised Smiley Faces Programme (SFP-R)

The SFP is a four item measure based on the MDAS, excluding the scale and polish item, which was poorly understood by children during pilot testing (Buchanan 2005). It has a computer-animated, seven-point faces response set. The faces scale is interactive, whereby children can increase the happiness or sadness of a neutral face. The SFP generates score between 4 (no DFA) and 28 (most severe DFA). It is suitable for children from age 6 years. The SFP has demonstrated high internal consistency, satisfactory test-retest reliability, and concurrent validity, as assessed by inter-scale correlations with the CFSS-DS and the MCDAS (Buchanan 2005). Of note, during its validation study there was no missing data for any of the participants, which was partly attributed to the use of a computer programme that makes it more difficult for an item to be missed unintentionally (Buchanan 2005).

The SFP-R includes an additional item about tooth extraction and an improved animated faces response set (Buchanan 2010). Children were involved in these changes and contributed to the development of the measure. Scores for the SFP-R range from 5 (no DFA) to 35 (most severe DFA). It is suitable for children from 4 to 11 years old. The SFP-R has demonstrated satisfactory internal consistency, test-retest reliability and concurrent validity as assessed by inter-scale correlations with the MCDAS (Buchanan 2010).

2.6.7. Use of DFA assessment measures by dental professionals

The available evidence suggests that DFA questionnaires are poorly utilised by dental professionals during patient assessment (Dailey et al. 2001; Alshammasi et al. 2018). Alshammasi and co-workers (2018) conducted qualitative interviews with postgraduate students, pre-specialty trainees, specialty trainees, and consultants in Paediatric Dentistry, and found that dental professionals are highly sceptical about the benefits of using DFA questionnaires, preferring to use their own clinical experience during patient assessment. Concern was also expressed during the interviews that a child would not be able to self-report their dental DFA reliably. Additionally, DFA questionnaires were perceived as impractical, and considered to be time consuming, difficult to score, and required further training to use. Traditionally, dental professionals have relied on personal data collection techniques, such as history-taking, for DFA assessment (Landgraf 1999). Whilst there is evidence to support the use of clinical judgement, standardised assessment approaches can improve the specificity and comprehensiveness of the information obtained (Holmes and Girdler 2005; Whiteside et al. 2016). As support for the use of self-report assessment measures, it has been demonstrated that when a child and a dental professional complete the same measure for state anxiety agreement between them is only poor to moderate (Buchanan and Niven 2003; Barros and Buchanan 2011). Moreover, clinical judgement in DFA assessment is better correlated with decision-making about interventions (e.g. the treatment modality to facilitate dental treatment) rather than identifying DFA per se (Dailey et al. 2002; Holmes and Girdler 2005; Barros and Buchanan 2011). This suggests that the use of a DFA assessment measure would provide a dental professional with additional knowledge regarding the DFA experienced by a child and support their own clinical judgement during decision-making (Alshammasi et al. 2018).

In the study by Alshammasi and co-workers (2018) the clinical application of questionnaires was also challenged by the study participants, with DFA questionnaires perceived as actually promoting DFA by introducing negative dental situations and terminology to children. However, there is evidence that this is not the case and that DFA self-assessment can have a positive influence on anxiety levels in adults. Dailey and co-authors (2002) conducted a randomised controlled trial involving eight general dental practices in North Wales. All participants competed the MDAS and short form of the State-Trait Anxiety Inventory State Anxiety Scale (STAI-S) in the waiting room prior to their first treatment visit. The long-form version of the STAI-S is a 20 item measure with a four-point response scale to determine intensity of anxiety at a particular moment (e.g. I am worried, I feel calm)) (Spielberger 1983).

The short form contains six of the items (Marteau and Bekker 1992). Only participants with severe DFA (MDAS score≥19; score or 5 for a single item) were included. The participants were then randomly allocated to the control group (MDAS given to the dental practice receptionist) or intervention group (MDAS given to the dentist). At the end of the treatment visit the participants completed the short form of the STAI-S again. Overall, the intervention group showed a significantly greater reduction in state anxiety than the control group. This suggests that the reduction in state anxiety was related to the participants' awareness that the dentist had received their questionnaire.

2.6.8. Electronic DFA assessment measures for children

Children have expressed a preference for the use of electronic questionnaires over written questionnaires to assess DFA (Jones and Buchanan 2010). Similar findings have been reported in other studies, whereby most children (range 77% to 87%) reported a preference for electronic assessment questionnaires (Bushnell et al. 2003; Wood et al. 2011). This is not surprising as mobile smart devices (smartphones, tablets) are widely used by U.K. children. In 2019, 68% and 55% of children aged 5 to 15 years reportedly used a tablet device or smart phone, respectively, to go online at home (Ofcom 2020). Mobile smart devices have operating systems that have been devised to be used with small software applications (i.e. apps). Correspondingly, almost all individuals with a tablet device use apps (Ofcom 2015). To date, only one measure of childhood DFA has been developed for computer application (Buchanan 2005; 2010). However, it is only available on desktop computers and not developed for mobile smart devices, limiting its wider usage.

Electronic assessment measures have a number of advantages over traditional pen and paper approaches, including: improved patient reporting of symptoms; increased disclosure of sensitive issues (e.g. mental health concerns); improved reporting of patient symptoms in clinical records by health professionals; and reduced numbers of missing items and data entry errors, with implications for data completeness and quality (Black and Ponirakis 2000;

Pakhomov et al. 2008; Vinney et al. 2012; Baggott et al. 2015; Bradford and Rickwood 2015). Additionally, mobile smart devices have advanced computing and connectivity capabilities (Marcano Belisario et al. 2015). Therefore, there is considerable scope for different clinical options and developments. These could include: linking measures to patient electronic healthcare records for clinical decision-making; adding alerts for clinicians (e.g. high scores, severe specific symptoms, change in symptoms); development of remote data entry (e.g. using downloadable app and a web-based data collection portal); inclusion of communication coaching, which has been shown to increase patient reporting of symptoms; incorporation of interactive voice response technology (e.g. low health literacy and visually impaired patient groups); and the use of cloud-based systems where results can be accessible to different health care providers (Basch 2014; Berry et al. 2014; Coons et al. 2015; Kao et al. 2015; Muehlhausen et al. 2015)

2.6.9. Summary and discussion of dental fear and anxiety assessment in children

Assessment of DFA in children has multiple uses. As it is a common, and clinically important condition, being able to identify and measure DFA in children is needed for research and survey purposes. Measurement of DFA prevalence in local populations can also aid planning of dental services for communities. For individual children, DFA assessment can facilitate treatment planning, promote good communication with dental professionals, and allow monitoring of treatment outcomes. For dental professionals, it can promote time-management and reduce potential occupational stressors of providing dental treatment when an insufficient appointment length has been allocated. There is also clinical potential in being able to identify children at risk of DFA development, although, to date, there has been little research into DFA risk assessment in children.

To assess DFA different methods have been described in the literature. The available evidence supports the use of self-reported questionnaires, completed by the child themselves, as the most valid and reliable approach (Porritt et al. 2013). As demonstrated during this review, there are multiple different self-reporting DFA measures in the scientific literature that have been used with children. Broadly, measures can be divided into those that assess state DFA, and those that assess trait DFA. Assessing state DFA has clinical utility for dental professionals. They can be used to identify children experiencing DFA at the time the measure is completed; and be used to monitor changes in DFA intensity during a dental visit, if used at different time points (Buchanan 2012). However, state DFA is not necessarily DFA that is problematic outside of the moment it is being experienced. For example, individuals with low levels of DFA may experience high intensity state DFA in a stressful dental situation, but once that is resolved return to their normal low DFA levels. Therefore, to assess DFA that is persistent, and interferes with functioning, measures of trait DFA are used. Trait measures can be employed outside the dental setting and in different contexts (Porritt et al. 2013).

There are multiple self-report scales and questionnaires described in the scientific literature that have been used with children for DFA assessment. Importantly, the quality and clinical utility of data derived from DFA assessment is dependent on the validity and reliability of the measure used (American Educational Research Association et al. 2014). Key is that measures have construct validity (American Educational Research Association et al. 2014). Consideration should be given to what is, and what is not, being assessed by DFA measures, and whether they capture what it is about DFA which is important and relevant to children (Armfield 2010b; Porritt et al. 2013).

Measures should also ideally be developed using an appropriate theoretical framework for the construct of interest (American Educational Research Association et al. 2014). A criticism of the available DFA measures is that they do not have a clear theoretical framework for DFA in children. The content of existing measures has frequently been based on DFA questionnaires for adults; or generated using a top-down approach from reviews of the literature and expert opinion. To date, there are no measures where the content has been derived from qualitative interviews with children with DFA. The use of adult measures with children assumes that the DFA construct captured by the measure is the same in adults and children. Yet there are many factors that differ for adults and children in the dental setting which could influence children's contemporary DFA experiences. Adult DFA measures may also have inappropriate items, wording, and response scales for children (Stevens 2010). A further criticism of existing measures is that they generally have a narrow conceptual focus on a variety of dental stimuli, procedures and situations which may trigger a DFA response during specific moments (Armfield 2010b; Porritt et al. 2013). There is evidence that DFA acquisition in children often includes a conditioning event, and that many dental stimuli have threat potential. However, measures with a narrow focus that only consider part of the construct, may not identify all individuals with DFA. This concern is potentially less crucial for children with severe DFA, than for those with a less global DFA experience, whose DFA may be missed or underestimated with measures that underrepresent the construct (Schuurs and Hoogstraten 1993). A further problem of having a high number of measures that do not tap into the same construct, is that it is difficult to compare findings between research studies and surveys which may restrict the potential for scientific progress (Schuurs and Hoogstraten 1993; Grisolia et al. 2021).

To overcome the conceptual limitations of DFA measures, a new theoretical model for DFA proposes that it is perceptions, notably perceptions of vulnerability, rather than learned experiences, that drive DFA, and the cognitive, affective, behavioural, and physiological responses associated with it (Armfield et al. 2008). Correspondingly, there has been a renewed interest in the application of bio-informational theory in DFA assessment (Schuurs and Hoogstraten 1993; Armfield 2010b). This conceptualisation considers the overall emotional fear and anxiety experience as a tripartite construct that is composed of behavioural (overt-motor), physiological (somato-visceral), and cognitive (verbal-report) responses (Lang 1968). Barlow (2002) extended this definition to place a greater emphasis on the subjective experience of affect. A cognitive-behavioural therapy (CBT) assessment model also incorporates this integrative tripartite view of fear and anxiety being composed of cognitive, behavioural, and physiological responses (Kendall 1985). It has been suggested that the experience of DFA for a child is composed of these highly interactive response systems, and construct validity of assessment measures would be heightened if they were measured (Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013). Assessment of DFA responses could also improve clinical utility of measures by determining how children's cognitive, affective, behavioural, and physiological responses are interacting to maintain their DFA over time (Porritt et al. 2013). To date, there has been little research into these responses in children, and how they form and shape what children with DFA experience. Further research would be needed to inform development of a measure accordingly.

2.7. Summary

On review of the literature relating to DFA assessment in children the following points can be made:

1. DFA is a common and clinically important condition in children.
2. Assessment of DFA is needed for research, epidemiological surveys, health service planning and delivery, and for the provision of treatment for individual children.
3. Within clinical practice, DFA assessment can be used to: quantify and understand the nature of DFA responses; inform decision-making about appropriate and timely DFA management; promote good communication; facilitate time management planning; and allow evaluation and monitoring of treatment outcomes.
4. Self-reported questionnaires, completed by children themselves, are the most valid and reliable assessment approach.
5. The construct of DFA in children is multidimensional and complex. The construct validity of existing DFA assessment measures for children has been criticised as they do not have a clear conceptual framework for the construct of DFA in children. Measures have a narrow conceptual focus, and content that has also been based on DFA measures for adults, or developed with a top-down approach, based on the scientific literature and expert opinion. To date, there are no measures where the content has been derived from qualitative interviews with children with DFA.

6. A measure based on a theoretical model that considers the DFA construct to be composed of cognitive, affective, behavioural, and physiological response systems has been proposed to improve the construct validity of DFA assessment in children. There is a need for further research to explore the experiences of DFA in children based on this theoretical model, which could inform the development of a new measure. The validity and relevance of assessment measures could be improved if children were involved in the development process.
7. The use of multiple measures that address related constructs has been suggested to improve the construct validity of DFA assessment. Further research to determine the clinical utility of incorporating multiple assessment measures during DFA assessment in children is required.
8. Dental professionals have negative perceptions on DFA self-report questionnaires for children and low motivation for their use. The use of electronic devices, such as mobile smart devices, has considerable potential to promote patient and clinician engagement in DFA assessment, and improve the quality and completeness of data collected. Further research into the use of electronic devices during DFA assessment in children is needed.

2.8. Publications arising from this chapter

Morgan, AG and Porritt, JM. (2017) 'Background and prevalence of dental fear and anxiety', in Campbell C. (ed). *Dental Fear and Anxiety in Pediatric Patients - Practical Strategies to Help Children Cope*. Basel, Switzerland: Springer International Publishing, pp. 3-19.

Morgan, AG. (2017) 'Dental fear and anxiety assessment in children', in Campbell C. (ed). *Dental Fear and Anxiety in Pediatric Patients - Practical Strategies to Help Children Cope*. Basel, Switzerland: Springer International Publishing, pp.31-42.

Chapter Three

3. Aims and objectives

3.1. Rationale and aims and objectives for research presented in this thesis

During Chapter Two, a narrative review of the literature on the prevalence, clinical implications, development and maintenance, and assessment of DFA in children was described. As presented, DFA in children is multidimensional and complex. A key finding identified throughout the review was the conceptual limitations of the existing DFA assessment measures in children. Notably, measures lack a conceptual framework for the construct of DFA in children, have been developed from an adult perspective, and typically have a narrow focus on specific dental procedures or situations. Prior to this research presented in this thesis, there were no existing DFA measures that had been developed based on qualitative interviews with children. A potential consequence of the conceptual difficulties identified in DFA assessment, is that measures may underrepresent the construct, or different measures may operationalise it in different ways (Schuurs and Hoogstraten 1993). That is, not all children, or populations of children that share DFA characteristics, are identified by DFA assessment measures (Locker et al. 1996). Consequently, it is difficult to compare findings between research studies or surveys, which may restrict scientific progress (Locker et al. 1996). Although, standardised DFA assessment has potential to improve patients care it is also seldom used by dental professionals during routine clinical practice (Dailey et al. 2001; Alshammasi et al. 2018). Specific barriers are the time to complete and score questionnaires during allocated patient appointment times (Alshammasi et al. 2018). Development of electronic DFA assessment questionnaires, for use of mobile smart devices, has considerable potential to address these concerns and promote dental professional engagement with DFA assessment.

The overall aim of the research described in this thesis is to further the understanding of DFA assessment in paediatric dental patients.

The research objectives to be addressed are as follows:

1. To assess the sociodemographic, quality of life, and child mental health characteristics of paediatric dental patients with DFA.
2. To explore the experiences of DFA in paediatric dental patients to inform the development of a new child-centred measure of DFA.
3. To design and test a web based DFA assessment measure for use on mobile smart devices.

The research was undertaken in three stages, in the next section the rationale and specific research aims and objectives for each stage are described.

3.2. Study 1: Examining sociodemographic factors, quality of life and mental health characteristics in paediatric dental patients with dental fear and anxiety

3.2.1. Rationale for Study 1

The use of standardised questionnaires to assess DFA in paediatric dental patients has multiple clinical applications (see Section 2.6) which would improve patients care, promote good communication between patients, families, and dental teams, and reduce occupational stressors by facilitating time management planning for dental professionals (Buchanan 2012; Jones and Huggins 2014). The use of multiple questionnaires, and making efforts to identify substantiating evidence for DFA, has been suggested to increase coverage of the construct for DFA in children (Schuurs and Hoogstraten 1993; Locker et al. 1996; Seligman et al. 2017). This approach also has potential to develop DFA assessment by considering it in broader terms and across different contexts (e.g. consider treatment factors, or how DFA impacts children's daily lives). It should be acknowledged that the use of multiple measures would increase the response burden placed on children and dental professionals (Yan et al. 2020). The evidence suggests that dental professionals have negative perceptions about DFA self-report questionnaires for children, and low motivation for their use in paediatric dentistry (Alshammasi et al. 2018). Therefore, clear evidence of clinical utility would be needed to justify the recommendation for multiple measures in DFA assessment.

Incorporating measures of HRQoL in DFA assessment is potentially a clinically useful approach, as it would assess the impact of DFA on children's daily living. Measurement of HRQoL could be used in development of a new measure to determine threshold values for clinical importance of DFA based on its impacts on children outside the dental setting (Silverman and Ollendick 2005). The use of a preference based HRQoL measure would also facilitate economic evaluations of DFA interventions (Stevens and Ratcliffe 2012). Measuring HRQoL in children with DFA is broadly supported by the literature. The available evidence suggests that there is a weak association between higher levels of DFA and worse OHRQoL in children (Alharbi et al. 2021). Children with severe DFA have also reported more difficulty eating, cleaning their teeth, smiling, laughing, or showing their teeth without embarrassment, and enjoying being with other people (Coxon et al. 2019b). In adults, there is much stronger evidence of an impact on daily living, whereby U.K. adults with high levels of DFA have been shown to be amongst those with the poorest OHRQoL, even when accounting for confounding factors (age, sex, and social class) (McGrath and Bedi 2004). However, DFA is not principally an oral condition and not all children with DFA have poor oral health. Consequently, measures of OHRQoL may be less relevant, or fail to explore its full range of impacts, in children. In the absence of a DFA-specific measure, the use of a generic HRQoL measures could overcome this limitation. To date, it is not known if a generic HRQoL measure has construct validity for the impact of DFA on daily living in children.

Mental health screening for children during DFA assessment also warrants consideration. Firstly, emotional disorders are common in U.K. children (Health and Social Care Information Centre 2018). Furthermore, the available evidence suggests children with high levels of DFA are more likely to have psychological difficulties (Versloot et al. 2008). It has also been suggested that anxiety disorders can contribute to the maintenance of DFA, resulting in it being more difficult to treat and associated with poor treatment outcomes (Makkes et al. 1987; Aartman et al. 1999; Locker et al. 2001a). Mental health screenings could potentially be used to identify children with a high need of specialist DFA and child mental health management (Locker et al. 2001a). Treatment of any psychological difficulties could also be potentially helpful for DFA management (Locker et al. 2001a). However, not all children with mental health problems have DFA, and further research is needed into the clinical usefulness of including it during DFA assessment.

3.2.2. Aims and objectives for Study 1

The aim of this study was to determine a profile of paediatric dental patients aged 11 to 16 years referred for management of DFA in a secondary/tertiary care clinical setting who would be potential users of a new DFA assessment questionnaire in clinical practice.

The specific objectives were:

- To assess the sociodemographic characteristics in paediatric dental patients with DFA to identify a patient profile for DFA assessment
- To assess the child mental health characteristics in paediatric dental patients with DFA
- To evaluate the relationship of DFA on daily living in paediatric dental patients using a generic HRQoL measure for children

3.3. Study 2: Exploring children's experiences of dental fear and anxiety

3.3.1. Rationale for Study 2

Most published research into DFA has not involved children as important and active partners in that research (Marshman et al. 2007; Marshman et al. 2015). That is, research has been *with* children, in the sense they are self-reporting their DFA, but the questionnaires being completed have been developed for adults, or from the adult perspective on DFA in children (Marshman et al. 2007). Additionally, such measures may also have inappropriate items, wording, and response scales for use with children (Stevens 2010). Therefore, our understanding of DFA has been influenced from the standpoint of adults. As it is children who are experiencing DFA, it is important to ascertain their expert knowledge and perspective on their own experiences (Larsson et al. 2018). Therefore, research involving qualitative interviews or focus groups with children with DFA has been recommended (Matza et al. 2013). Qualitative methods facilitate a more comprehensive, adaptable, and individual research approach to understanding the breadth of children's experiences (Stewart et al. 2008). Admittedly, DFA is not only experienced by children, but also at the same time by parents/carers and dental professionals. However, obtaining the perspectives of such stakeholders will not replace the relevance of research that can arise if children fully participate themselves (Larsson et al. 2018).

The use of a theoretical conceptual framework that considers children's multidimensional emotional DFA experience as composed of cognitive, affective, behavioural, and physiological responses has been proposed for a new measure of DFA, with the suggestion it would provide a more complete understanding of the construct (Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013). A CBT assessment and management model incorporates this conceptualisation of DFA (Williams and Garland 2002). A CBT model would provide a structured assessment approach that considers the thoughts, feelings, physical symptoms, and behaviours a child with DFA experiences and how they interact to maintain DFA overtime (Buchanan 2017). To date, there has been very little research to explore these responses in children, and how they form and shape what children with DFA experience. Further research could expand our understanding of DFA in children and inform development of a new measure accordingly.

3.3.2. Aims and objectives for Study 2

The aim of this study was to explore the DFA experiences of paediatric dental patients with DFA aged 11 to 16 years to inform development of a new child-centred measure of DFA. Specific objectives were:

- To conduct qualitative interviews with paediatric dental patients with DFA
- To utilise a CBT assessment model as a theoretical framework to inform the qualitative interviews and understanding of children's multidimensional DFA experiences

3.4. Children's Experiences of Dental Anxiety Measure

As previously introduced, the CEDAM and CEDAM-8 are new DFA questionnaires that were developed to address the conceptual limitations of existing DFA measures for children identified during the review of the literature (Porritt et al. 2018; Porritt et al. 2021). It is based on the Five Areas™ CBT theoretical framework for the assessment and treatment of anxiety (Williams and Garland 2002). This model identifies the unhelpful thoughts, feelings, behaviours, and physical symptoms that are acting to maintain DFA in children, rather than stimuli and situations in the dental setting that may, or may not, elicit a DFA response in an individual. The fifth domain or area described by the model is the situational factors (e.g. factors external to an individual) that has resulted in anxiety. The content of the CEDAM was derived from the qualitative interviews conducted with children in Study 2. Children's anxiety experiences were mapped onto each of the internal domains (i.e. excluding situational factors) of the framework. Children were also involved in cognitive pretesting and piloting of the measure to improve its developmental validity. It is the first DFA measure for children that has been fully developed with children. A full description of the measure is provided in Chapter 6.

3.5. Study 3: Development and testing of a web-based version of the Children's Experience of Dental Anxiety Measure for clinical assessment of paediatric dental patients aged 9 to 16 years

3.5.1. Rationale for Study 3

Mobile smart devices offer new opportunities for DFA assessment in children. To date, the CEDAM has only been available in paper format. However, children have expressed a preference for the use of electronic questionnaires to assess DFA (Jones and Buchanan 2010). Similar findings have been reported in other studies, whereby children reported a preference for electronic assessment questionnaires over written data collection (Bushnell et al. 2003; Wood et al. 2011). This is not surprising as mobile smart devices (smartphones, tablets) are widely used by U.K. children (Ofcom 2020). Previous research has suggested that dental professionals have negative perceptions on DFA self-report questionnaires for children and low motivation for their use (Alshammasi et al. 2018). Electronic assessment measures have a number of advantages over traditional pen and paper approaches, including: improved patient reporting of symptoms; increased disclosure of sensitive issues (e.g. mental health concerns); improved reporting of patient symptoms in clinical records by health professionals; and reduced numbers of missing items and data entry errors, with implications for data completeness and quality (Black and Ponirakis 2000; Pakhomov et al. 2008; Vinney et al. 2012; Baggott et al. 2015; Bradford and Rickwood 2015). The use of electronic devices, such as mobile smart devices therefore have considerable potential to promote patient and clinician engagement in DFA assessment.

Coons and co-authors (2009) have published best practice guidelines for the evidence required to demonstrate comparability between an electronic measure and a paper-based measure. The recommendations are based on the extent a written questionnaire needs to be modified to migrate it onto an electronic platform. If substantial changes are not made (e.g.

wording of items) full psychometric testing is not necessary (Coons et al. 2009). However, the minimum requirement is for cognitive and usability testing to be conducted with users (Coons et al. 2009). A potential concern that also warrants consideration, is whether by changing the mode of questionnaire delivery, measurement error is introduced into the responses (Marcano Belisario et al. 2015). That is, if completing the questionnaire on a mobile smart device in some way influences how a child responds to the items and introduces bias. The available evidence suggests that using apps for self-administered questionnaires does not affect data equivalence if the validity settings and target population for the paper-based version of the measure are unchanged (Marcano Belisario et al. 2015). However, Marcano-Belisario and co-authors (2015) were unable to determine the effects of: setting; data entry formats for response scales (e.g. drop-down boxes, faces scales); technical specifications (e.g. screen size); and patient characteristics. Therefore, equivalence testing to determine agreement between scores delivered by an electronic questionnaire and the original paper-based questionnaire has been recommended (Coons et al. 2009).

3.5.2. Aims and objectives for Study 3

The aim of this study was to develop and test a web-based version of the CEDAM for use on tablet devices with paediatric dental patients aged 9 to 16 years in a secondary/tertiary care clinical setting. Specific objectives were:

- To involve paediatric dental patients in the development of a web-based version of the CEDAM
- To determine measurement agreement between the web-based version (eCEDAM) and original paper version (CEDAM)
- To determine if, compared to the paper version, the web-based version had an influence on data quality (time to complete, proportion of missing items, and proportion of completed questionnaires)
- To assess acceptability and participant preference for the mode of delivery

Chapter Four

4. Study 1: Examining sociodemographic factors, quality of life and mental health characteristics in paediatric dental patients with dental fear and anxiety

4.1. Introduction

As presented in the previous chapters, the use of standardised questionnaires for DFA self-report by paediatric dental patients has potential to improve patient care, reduce occupational stressors for dental professionals, and promote good communication (Buchanan 2012; Jones and Huggins 2014). DFA measures can support dental professionals to identify children with DFA, or those who have DFA about a specific dental situation or procedure (e.g. local anaesthetic, dental extraction). For dental professionals, there are occupational stressors in completing a dental visit in the time allocated to it within a clinical session (Jones and Huggins 2014). Therefore, DFA assessment could inform time-management planning of an appropriate appointment length or appointment time in the working day (Jones and Huggins 2014; Alshammasi et al. 2018). DFA assessment can also promote good communication between children, families, and dental professionals, whereby patients perceive that their dental professional is concerned about their DFA, and that they are being involved in treatment planning decisions (Jones and Huggins 2014). There is evidence that the use of standardised DFA questionnaires reduces state DFA in patients (Dailey et al. 2002).

A limitation of existing DFA measures for children is their narrow conceptual focus. To date, questionnaires have assessed children's DFA towards dental stimuli and situations that represent specific moments of a dental experience (Porritt et al. 2013). The use of multiple measures that address related constructs has been suggested to improve DFA assessment by providing a more complete picture of DFA in children (Schuurs and Hoogstraten 1993). Assessment of HRQoL and child mental health during DFA assessment in children are potentially clinically useful approaches, by determining the impacts of DFA on children's daily lives, and to identify a patient group who may require additional support (Aartman et al. 1999; Locker et al. 2001a; Stevens 2010). However, dental professionals do not value standardised DFA questionnaires within DFA assessment in children (Alshammasi et al. 2018). The use of multiple measures would further increase the response burden for patients and dental professionals, and potentially introduce additional barriers to their use in clinical practice. Further research to determine the clinical utility of incorporating multiple assessment measures during DFA assessment in children is required.

4.2. Aims and objectives

The aim of this study was to determine a profile of paediatric dental patients aged 11 to 16 years referred for management of DFA in a secondary/tertiary care clinical setting who would be potential users of a new DFA assessment questionnaire in clinical practice.

The specific objectives were:

- To assess the sociodemographic characteristics in paediatric dental patients with DFA to identify a patient profile for DFA assessment
- To assess the child mental health characteristics in paediatric dental patients with DFA
- To evaluate the relationship of DFA on daily living in paediatric dental patients using a generic HRQoL measure for children

4.3. Ethical approval

Ethical approval for the research was granted by the NRES Committee York and Humber: Leeds West REC (13/YH/0163).

4.4. Method

A cross-sectional study design was employed whereby 100 children completed a questionnaire containing sociodemographic items and measures to assess DFA, HRQoL, and child mental health characteristics. The sociodemographic data was collected to determine a profile of children referred to a paediatric dentistry unit within an NHS dental teaching hospital for DFA management. This is a patients group whose dental care could be improved with the use of standardised DFA, are who would be potential users of a new DFA questionnaire in clinical practice. The sample size was based on a study that similarly sought to determine a profile for adult patients with DFA who were referred to a sedation clinic at a secondary and tertiary care setting in London (Boyle et al. 2010).

4.4.1. Materials

The questionnaires used in the study included items to record sociodemographic data (age, sex, postcode, and ethnicity) and four previously validated measures: MCDAS; Revised Children's Anxiety and Depression Scale; Strengths and Difficulties Questionnaire; and Child Health Utility 9D (see Appendix 1). These measures are described in detail in the next section. The questionnaire booklet contained nine additional items that were used to inform the development of a CBT resource for DFA in children (Porritt et al. 2016). Classification of ethnic group was based on the categories (White, Mixed/Multiple ethnic groups, Asian/Asian British, Black/African/Caribbean/Black British, Other ethnic group) used by the Office of National Statistics (ONS) for measuring equality during national surveys (Office of National Statistics 2016).

4.4.1.1. Modified Child Dental Anxiety Questionnaire

The MCDAS is an eight-item, valid, self-report trait measure for DFA in children aged 8 to 15 years (Wong et al., 1998). As it has reasonable psychometric properties, and is easy to administer to children, it has been suggested for use as the current gold standard for DFA assessment in children (Porritt et al. 2013; Porritt et al. 2018). It comprises seven items to assess how relaxed/worried children are to typical situations (e.g. having an injection in the gum), and one item that assesses overall DFA. The item ‘having your teeth scraped and polished’ was changed to ‘having your teeth cleaned and polished’ for the purposes of the study, following evidence that children lack an understanding about the procedure for a scale and polish (Buchanan 2005). The MCDAS has a five-point severity response scale (1=relaxed/not worried to 5=very worried). To assess severity of DFA the scores for each item are summed to provides a total score (minimum score=8; maximum score=40). Higher scores correspond to higher DFA severity. Its psychometric properties have been previously presented. The mean MCDAS score for a sample of U.K. school children (n=277) aged 8 to 15 years was 18.2 (SD=7.14) (Wong et al. 1998). To date, a threshold score for problematic/high levels of DFA has not been reported. However, an identified limitation of the MCDAS is that it appears to generate a high number of incomplete questionnaires (Wong et al. 1998). One possible explanation is that children may lack an understanding of some of the included dental situations (e.g. scale and polish, inhalation sedation) (Buchanan and Niven 2002). No method has been described for management of missing data.

4.4.1.2. Revised Children’s Anxiety and Depression Scale

The Revised Children’s Anxiety and Depression Scale (RCADS) is a 47 item self-report, multidimensional measure for anxiety and depression in children aged 8 to 18 years (Chorpita et al. 2000; Chorpita et al. 2005). It comprises five subscales for anxiety disorders (generalised anxiety disorder, obsessive compulsive disorder, panic disorder, separation anxiety disorder, and social phobia) and one subscale for major depression disorder. The content of the measure was derived from the Spence Children’s Anxiety Scale, but also includes items relating to depression (Spence 1998; Chorpita et al. 2000). The RCDAS assesses current (day the questionnaire completed) symptoms of anxiety and depression disorders using the DSM-IV diagnostic criteria (American Psychiatric Association 1994; Chorpita et al. 2000). Exploratory factor analysis confirmed a six-factor solution consistent with the DSM-IV criteria

(Chorpita et al. 2000). Internal consistency of the measure has been found to be satisfactory overall (α=0.96), and for the individual scales in both non-clinical and clinical samples of children (α=0.71 to 0.85 and α=0.78 to 0.88, respectively) (Chorpita et al. 2000; Chorpita et al. 2005; De Ross et al. 2012). It has a four-point response format based on frequency of symptoms (0=never; 1=sometimes; 2=often; 3=always). The sum of the five anxiety subscales provides a total score for anxiety; whilst the sum of all six subscales gives a total score for internalising problems (anxiety disorders and depressive disorders). Measures with missing data do not need to be excluded from analysis if there were less than two items missing per subscale (Chorpita et al. 2015). A total score for a subscale with missing items is then determined pro rata (i.e. sum of completed scale items divided by number of items completed and multiplied by total number of scale items) using the scores available for completed items (Chorpita et al. 2015). The raw scores for subscales or totals are converted to an overall standardised score based on United States of America (USA) schooling grade and sex. The school-grades are reduced to 5 groupings as follows: grades 3rd and 4th; 5th and 6th; 7th and 8th, 9th and 10th, and 11th and 12th (ages 9 to 10; 11 to 12; 13 to 14; 15 to 16; and 17 to 18, respectively). Standardised scores of 65 and 70 (two standard deviations above the mean score for a child of the U.S.A. school grade and sex) is defined as the borderline clinical and clinical threshold value for each of the subscales and the total scores, respectively (Chorpita et al. 2015). Normative scores are not available for U.K. children. A limitation of the RCDAS is its length, as it is reported to take approximately 25 minutes to complete (Wolpert et al. 2015).

4.4.1.3. Strengths and Difficulties Questionnaire

The Strengths and Difficulties Questionnaire (SDQ) is a measure of social, emotional, and behavioural function in children aged 4 to 17 years (Goodman 2001). It is available as a self-report version for children aged 11 to 16 years old. The SDQ comprises 25 items (negative and positive statements) divided between five scales (emotional problems, conduct problems, hyperactivity, peer relationship problems and prosocial behaviour). Measure users consider their experience of the items over the previous six months. For each item there is a three-point response scale for agreement (0=not true, 1=somewhat true, 2=certainly true). For positive statements the response scale is scored in reverse (2=not true, 1=somewhat true, 0=certainly true). The total scores for each subscale (0=minimum/lowest score, 10=maximum/highest score), excluding prosocial behaviour, are summed to give a total difficulties score (0=minimum/lowest score, 40=maximum/highest score). Measures with missing data do not need to be excluded from analysis if there is a minimum of three items completed for each subscale (Youth in Mind 2006). A total score for a subscale with missing items is determined pro-rata using the scores available for completed items i.e. sum of scores for completed items divided by number of items completed and multiplied by number of items in subscale (Youth in Mind 2006). SDQ scores can be categorised into four bands for social, emotional and behavioural function: close to average (previously termed 'normal' band); slightly raised/lowered (previously termed borderline band); high/low (previously termed 'abnormal' band); and very high/low (previously termed 'abnormal' band) (Youth in Mind 2016). The threshold scores for each banding were based on a community sample whereby 80%, 10%, 5% and 5% of U.K. children are classified into each band respectively (Youth in Mind 2016). Generally, the SDQ has good psychometric properties (Wolpert et al. 2015). The internal consistency for the measure is reported as satisfactory (α= 0.80 to 0.82) (Goodman and Scott 1999; Goodman 2001). However, individual subscales have been found to have low internal consistency (α=0.41 to 0.67) (Goodman 2001). The SDQ has demonstrated excellent completion rates with only 0.4% of data reported missing in a large national survey (Goodman et al. 2010).

4.4.1.4. Child Health Utility 9D

The Child Health Utility 9D (CHU-9D) is a self-report, preference-based measure of HRQoL for children. It was originally developed for children aged between 7 and 11 years, although it has been shown to be valid for children aged 11 to 17 years also (Stevens 2010; Ratcliffe et al. 2012). Its dimensions, response scales, content and formatting were developed through qualitative interviews with children (Stevens 2010). The measure contains one question and a corresponding response scale for each of nine dimensions of HRQoL (worry, sad, pain, tired, annoyed, schoolwork/homework, sleep, daily routine, participation in activities). The response scale comprises five rating levels representing increasing impact within each dimension (e.g. I=I don't feel worried today, 2=I feel a little bit worried today, 3=I feel a bit worried today, 4=I feel quite worried today, 5=I feel very worried today). Children are asked to rate their response based on how they feel that day. The CHU-9D has demonstrated good psychometric properties and completion rates (Stevens 2011; Stevens and Ratcliffe 2012).

As the CHU-9D is a preference-based measure it can be used to generate preference weights (utility values) for each health state described by the descriptive system. Preference weights are calculated from the measure by an algorithm that applies a special tariff to each response (Fuber and Segal 2015). This algorithm is derived from a valuation process whereby all the possible unique health states described by a measure are ranked between 0 (death) and 1 (perfect health) (Fuber and Segal 2015). The preference weights were derived from interviews with the U.K. adult general population using the standard gamble technique (Stevens 2012). The CHU-9D generates utility values between 0.33 and 1 (Fuber and Segal 2015). It is not possible to calculate utility values for completed measures with any missing data (University of Sheffield 2019). Higher utility values reflect better HRQoL. Mean utility scores from 0.85 to 0.88 have been reported for different non-clinical and clinical populations of children aged 6 to 17 years (Stevens and Ratcliffe 2012; Canaway and Frew 2013; Foster Page et al. 2014). Utility scores can then be used to calculate Quality-Adjusted Life Years (QALY) for children for use in cost utility analysis of different heath care interventions.

4.4.2. Participants

Participants comprised paediatric patients attending an assessment appointment at a paediatric dentistry unit within an NHS dental teaching hospital in Sheffield between February 2014 and January 2015. The patients had been referred from a primary or secondary dental care provider for DFA management. Only children newly referred for DFA management were included, as dental treatment could have influenced DFA severity. Participants were identified by a single researcher (A.M.) screening all incoming consecutive paediatric referral letters to the dental hospital. Patients were approached if they were aged between 11 and 16 years, and the referral letter mentioned either: the presence of DFA; or requested dental treatment with inhalation sedation or general anaesthesia for DFA. Only children with DFA were included to increase the likelihood that the patient profile would be relevant to other children with DFA (Porritt et al. 2016). Children were excluded from participating in the study if there was a consideration in the referral letter that would have prevented them from being able to complete the questionnaire (e.g. severe communication difficulties). The age range was selected as the SDQ self-report measure is validated for children aged 11 to 17 years only (Goodman 2001). The potential participants were sent an information pack in the post to their given address containing: a questionnaire; a participant information sheet; a parent/carer information sheet; and a request to bring in the questionnaire with them to their first attendance in the dental hospital, should they agree to participate in the study. Subsequently, all first appointments were then arranged as per the local waiting list and booking arrangements. In the event a potential participant did not bring their completed questionnaire on their first visit, writing materials and a further questionnaire were provided for them whilst they waited for their appointment in a dental hospital waiting room. As it is possible that there could be a difference in DFA in participants who completed the questionnaire at home and those that completed it whilst at the dental hospital, the location the questionnaire was completed was recorded. Consent was implied by completion of the questionnaire. Further written consent was not obtained. Consecutive recruitment continued until 100 participants had completed a questionnaire.

4.4.3. Analysis

Data for the sociodemographic and measure variables were transferred manually to a Microsoft Excel Office 365 database (Washington, Microsoft Corporation). Postcode data was used to identify a deprivation quintile for each participant using the GeoConvert function on the U.K. Data Service Census Support website (U.K. Data Service Census Support 2019). Postcode data determines the Lower Super Output Area (LSOA), a small area/neighbourhood with a population of approximately 1500, based on the 2011 U.K. census (Department for Communities and Local Government 2015). Within the Index of Multiple Deprivation 2015 each LSOA is ranked for relative deprivation from 1 (most deprived) to 32,844 (least deprived) (Department for Communities and Local Government 2015). Subsequently, deprivation quintiles (five equal groups) were determined from the most deprived 20% LSOA, to the least deprived 20% LSOA. Scoring syntaxes for IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation) were used to generate the scores for RCADS, SDQ and CHU-9D (Youth in Mind 2006; UCLA Child First Program 2019; University of Sheffield 2019). Missing data for each of the included measures was managed as per the corresponding user guides (Youth in Mind 2006; Chorpita et al. 2015; University of Sheffield 2019) To date, no method has been described for management of MCDAS missing data. Therefore, when a single item was missing the median/mean scores for the specific item were imputed, dependent on the distribution of the data for the item. When multiple items were missing the participant was excluded. All data was subsequently analysed using IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation) and GraphPad Prism version 7 for Windows (California, GraphPad Software). Simple descriptive analysis was conducted for the sociodemographic variables (age, sex, ethnicity, and postcode). Internal consistency was calculated for the MCDAS, RCADS, SDQ and CHU-9D using Cronbach's alpha. Terwee and co-authors (2007) have proposed within their quality criteria for health status questionnaires, that a minimum standard for internal consistency is when Cronbach's alpha is between 0.70 and 0.95. For each measure normality tests were conducted for each subscale and scale. (Kim 2013). Subsequently, both parametric and non-parametric techniques to compare groups and analyse correlation were used dependent on the underlying distribution and variables. Analysis conducted to compare groups included: DFA and demographic variables; DFA and location questionnaire completed; and sex differences between child mental health variables. To determine the relationship between DFA and child mental health variables scatterplots were generated, and correlation analysis determined. To interpret the strength of correlation, Cohen (1992) recommends a correlation coefficient of 0.10 to 0.29 represents a small correlation, 0.30 to 0.49 represents a medium correlation, and 0.50 to 1.0 represents a large correlation. A p-value of 0.05 was selected as the level of significance for all tests.

4.5. Results

Overall, 207 children were invited to participate in the study, with 108 children recruited between February 2014 and January 2015. Nearly a quarter of children (23%; n=48) who were sent questionnaires were not brought to their assessment appointment, and therefore did not participate in the study. Of the 159 children who were invited to participate and then attended an assessment appointment, 32% (n=51) declined to take part. Participants were not asked to provide a reason for their decision not to participate. The response rate was 68%. Subsequently, seven participants were excluded from the analysis as multiple MCDAS items were missing from the completed questionnaires (range=2-6), and one participant was excluded from the analysis as missing items from the CHU-9D (n=4). A flow diagram of participant recruitment is presented in Figure 1. More participants completed the questionnaire whilst waiting for their assessment appointment in the dental hospital than at home (59% and 41%, respectively).

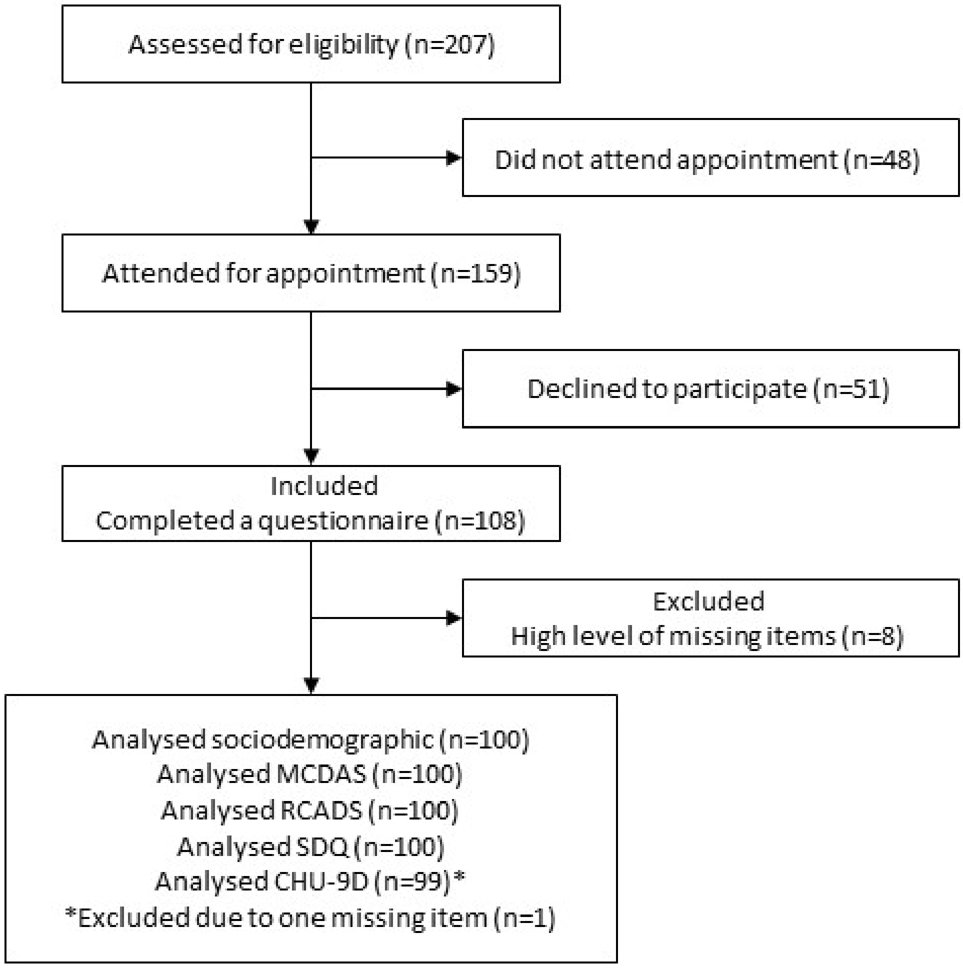

Figure 1. Flow chart of progress of participant recruitment and inclusion through the study

4.5.1. Sociodemographic variables

The age of the participants ranged from 11 to 16 years (mean=13.1, S.D.=1.4). There were more female than male participants (61% and 39%, respectively). Overall, 50% (n=49) of children lived in areas identified as the most deprived areas of England (quintile 1). Most (85%) participants identified their ethnicity as 'White'. Detailed participant demographic data is presented in Table 1. There were no significant differences between participants and those that declined to participate for age (Chi-squared test for independence, χ^2=0.5; p=0.48), sex (Chi-squared test for independence [Yates Continuity Correction applied], χ^2=0.0; p=1.0), or deprivation quintile (Chi-squared test for independence, χ^2=6.1; p=0.19).

Table 1. Data for participants who completed the questionnaire and were included in the analysis

Variable	**Category**	Median/Frequency
Age (n=100)	Mean	13.1 years (S.D.=1.4)
	Median* *Age did not follow a normal distribution	13.0 years* (inter-quartile range=2.0 years)
	Range	Minimum=11 years Maximum=16 years
Sex (n=100)		
	Male	39%
	Female	61%
Deprivation Quintile (n=98)		
	1 (Most deprived)	50.0% (n=49)
	2	10.2% (n=10)
	3	18.4% (n=18)
	4	9.2% (n=9)
	5 (Least deprived)	12.2% (n=12)
Ethnicity (n=100)		
	White	85%
	Mixed/multiple ethnic groups	8%
	Asian/Asian British	2%
	Black/African/Caribbean/Black British	3%
	Other ethnic groups	2%

4.5.2. Reliability

Internal consistency, calculated using Cronbach's alpha, was found to be satisfactory for MCDAS (total score), RCDAS (generalised anxiety disorder subscale, major depressive disorder subscale, obsessive compulsive disorder subscale, panic disorder subscale, separation anxiety disorder subscale, social phobia subscale) and SDQ (total difficulties scale, emotional symptoms subscale and hyperactivity subscale). However, internal consistency was found to be low for the CHU9D, and SDQ conduct and peer problems subscales. Detailed internal consistency data for each measure is presented in Table 2.

Table 2. Internal consistency for MCDAS, RCADS, SDQ and CHU-9D scales and subscales

Measure	Scale/Subscale	Number of items	Internal Consistency
Modified Child Dental Anxiety Questionnaire (n=100)		n=8	α=0.85
Revised Children's Anxiety and Depression Scale (n=100)	Total anxiety	n=37	α=0.96
	Total internalising	n=47	α=0.96
	Generalised anxiety disorder	n=6	α=0.87
	Major depressive disorder	n=10	α=0.88
	Obsessive compulsive disorder	n=6	α=0.82
	Panic disorder	n=9	α=0.89
	Separation anxiety disorder	n=7	α=0.75
	Social phobia	n=9	α=0.88
Strengths and Difficulties Questionnaire (n=100)	Total difficulties	n=40	α=0.84
	Conduct problems s	n=10	α=0.61
	Emotional problems	n=10	α=0.77
	Peer problems	n=10	α=0.65
	Hyperactivity	n=10	α=0.76
Child Health Utility 9D (n=99)	Health Related Quality of Life	n=9	α=0.68

4.5.3. Dental fear and anxiety severity

The eight items in the MCDAS were scored from one to five. Overall, 10% (n=10) participants had one missing item. Each missing item was replaced by the item median (scores for each item were not normally distributed). No participants were excluded from the analysis. The total DFA score was calculated manually for each participant. The minimum total score was 8 and the maximum total score 40. Higher MCDAS scores are associated with higher DFA severity. Normality tests identified that: total DFA score per participant; location questionnaire completed (home or clinic); sex (male or female); and deprivation quintile (one to five), were normally distributed (Kim 2013). The total DFA scores for participants per year of age (11, 12, 13, 14, 15 years) were not normally distributed (Kim 2013). Detailed skewness and kurtosis data are presented in Appendix 2. MCDAS total scores ranged from 9 to 40. The overall mean total DFA score was 25.5 (S.D.=7.4; 95% confidence interval for the mean=24.1-27.0). The mean total DFA scores according to location, sex, and deprivation quintiles, and the median total DFA score and interquartile range (I.Q.R.) for participant age, are presented in Table 3. Independent t-tests were conducted to compare the total DFA scores according to location questionnaire completed ($t=1.6$, $p=0.12$) and sex ($t=-0.9$, $p=0.38$), with no significant differences between the respective groups identified. A one-way ANOVA test was used to compare the relationship between total DFA score and deprivation. Levene's test was carried out and equal variances between groups could be assumed. There was no significant difference in mean total DFA score (one-way ANOVA, [$F(4,93)=0.7$, $p=0.38$]) between deprivation quintiles. A Kruskal-Wallis test found no statistically significant differences in total DFA scores between age groups (Chi-squared=0.5, $p=1.0$). These findings suggest that DFA in the study population was not associated with location questionnaire completed or participant age, sex, or deprivation.

Table 3. Total DFA (MCDAS) score data according to location questionnaire completed, sex, deprivation, and age

Category	Frequency % (n)	Mean MCDAS score*	S.D.	95% C.I. for mean
Minimum MCDAS score=8, Maximum MCDAS score=40				
Location questionnaire completed	**n=100**			
Home (Before appointment)	41	26.9	7.3	24.6-29.2
Clinic (During appointment)	59	24.6	7.4	22.6-26.5
Sex	**n=100**			
Male participants	39	24.7	8.2	22.0-27.4
Female participants	61	26.1	6.9	24.3-27.8
Deprivation	**n=98**			
Quintile 1 (Most deprived)	50.0% (n=49)	25.0	7.6	22.8-27.2
Quintile 2	10.2% (n=10)	28.1	5.0	24.5-31.6
Quintile 3	18.4% (n=18)	24.8	8.3	20.7-29.0
Quintile 4	9.2% (n=9)	24.0	7.4	18.3-29.7
Quintile 5 (Least deprived)	12.2% (n=12)	27.4	7.8	22.4-32.3
Age (years)		(Median)	(I.Q.R.)	
11	14% (n=14)	24.5	21.4-27.8	
12	25% (n=25)	24.0	17.0-35.0	
13	19% (n=19)	26.0	21.0-30.4	
14	20% (n=20)	26.0	19.1-28.8	
15	22% (n=22)	26.0	21.5-29.4	

4.5.4. Anxiety and depression

For the RCADS, raw scores were calculated for generalised anxiety disorder, major depressive disorder, obsessive compulsive disorder, panic disorder, separation anxiety disorder, and social phobia subscales, and the total score for anxiety scale (sum of five anxiety disorder subscales) and total score for internalising scale (sum of five anxiety disorder subscales and major depressive disorder subscale). The raw scores were converted to a standardised score (based on sex and USA school grade) using a syntax programme for IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation). Overall, 12 participants had missing items of data (range 1 to 6 items). The missing items were imputed by calculating a pro rata score based on the other completed items for the corresponding subscale (Chorpita et al. 2015). No participants were excluded.

Normality tests were conducted for each subscale and scales for all participants in the total sample, and according to sex. All subscales and scales, except the major depressive disorder subscale (total, male and female participants) and total internalising scale (female participants), were not normally distributed (Kim, 2013). Detailed skewness and kurtosis data are presented in Appendix 2. Standardised scores of 65 and 70 have been suggested as the borderline clinical threshold and clinical threshold values, respectively, for each of the subscales and scales based on a representative sample of Hawaiian children aged 6 to 18 years (UCLA Child First Program 2019). For the purposes of analysis the borderline clinical and clinical threshold scores (e.g. scores in clinical range) were combined (Thompson et al. 2021).

The median for the total anxiety scale and total internalising scale scores for the total sample were 40.2 (IQR=34.7-55.3) and 40.5 (IQR=34.1-56.9), respectively. The descriptive statistics for each subscale are presented in Table 4. All median/mean scores were below the borderline clinical/clinical threshold. The proportion of total participants with a score above the borderline clinical/clinical threshold was 7% for generalised anxiety disorder, 14% for major depressive disorder, 8% for obsessive compulsive disorder, 16% for panic disorder, 17%

for separation anxiety disorder, 8% for social phobia, 11% total anxiety scale, and 13% for the total internalising scale. There was a significant difference between the proportion of participants identified with a score in the normal range and a score in the borderline/clinical range compared to expected normative scores (normal range=94%; borderline/clinical range=6%) for major depressive disorder, panic disorder, separation anxiety, total anxiety scale, and total internalising scale (Chi-squared test, goodness of hit, $p \leq 0.05$). A Mann-Whitney U Test to compare total anxiety scale scores for male and female participants identified a significant difference between scores for male participants (median=36.6, n=39) and female participants (median=45.0, n=61) (U=833.0, z=-2.5, p=0.01, r=-0.3). Similarly, a significant difference was found for total internalising scale scores for male participants (median=36.7, n=39) and female participants (median=46.5, n=61) (Mann-Whitney U test, U=793.0, z=-2.8, p=0.01, r=0.3). Therefore, female participants had higher total anxiety and internalising scale scores than male participants (Table 5). There were no differences for proportions of children with a score in the clinical category for the total anxiety scale between male and female participants (8% vs 13%, Fisher's Exact Test, p=0.52), or for total internalising scale between male and female participants (10% vs 15%, Chi-squared Test, p=0.73).

Table 4. Descriptive statistics for Revised Child Anxiety and Depression Scale for all participants compared to expected population norms

Scale/Subscale	**n**	**Mean (S.D.)**	**Median (I.Q.R.)**	**Non-clinical* (%)**	**Borderline/ Clinical* (%)**	**χ^2 *P* value****
Generalised anxiety disorder	100	43.8 (12.8)	42.1 (33.4-52.1)	93	7	*p*=0.67
Major depressive disorder	100	49.6 (14.8)	46.8 (36.4-60.8)	86	14	***p*=0.00**
Obsessive compulsive disorder	100	45.9 (13.1)	42.7 (35.2-53.2)	92	8	*p*=0.40
Panic disorder	100	53.4 (15.3)	48.2 (42.9-57.1)	84	16	***p*=0.00**
Separation anxiety disorder	100	52.3 (13.6)	48.0 (42.7-57.4)	83	17	***p*=0.00**
Social phobia	100	43.1 (12.3)	40.3 (33.0-51.5)	92	8	*p*=0.40
Total anxiety	100	46.3 (14.9)	40.2 (34.7-55.3)	89	11	***p*=0.04**
Total internalising	100	46.8 (15.7)	40.5 (34.1-56.9)	87	13	***p*=0.00**

* Borderline/clinical range is a standardised score≥65 (6% of non-clinical population) (Wolpert et al. 2015).

**Statistical significance defined at 5% levels for values in bold.

Table 5. Comparison of proportion of participants in borderline/clinical group for total anxiety scale score and total internalising scale score for sex

	n	Mean (S.D.)	Median (I.Q.R.)	Non-clinical* % (n)	Borderline/ Clinical* % (n)	*P* value**
Total anxiety scale score						
Sex						*p*=0.52
Male	39	42.0 (12.7)	36.6 (33.5-47.8)	92 (36)	8 (3)	
Female	61	49.1 (15.7)	45.0 (36.7-59.4)	87 (53)	13 (8)	
Total internalising scale						
Sex						*p*=0.73
Male	39	41.8 (13.3)	36.7 (33.1-47.2)	90 (35)	10 (4)	
Female	61	50.0 (16.3)	46.5 (36.8-61.4)	85 (52)	15 (9)	

*Borderline/clinical range is a standardised score≥65 (6% of non-clinical population) (Wolpert et al. 2015).

**Statistical significance defined at 5% levels for values in bold.

4.5.5. Social, emotional, and behavioural function

Scores were calculated for conduct problems, emotional problems, hyperactivity, peer problems and total difficulties SDQ subscales using a statistical syntax for IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation) (Youth in Mind 2006). Overall, two participants each had one missing item of data. The missing item was imputed by calculating a pro rata score based on the other completed items for the corresponding subscale (Youth in Mind 2006). No participants were excluded. All subscales were normally distributed, except conduct and peer problems subscales for the total sample; conduct, emotional and peer problems for male participants; and peer problems for female participants (Kim 2013). Detailed skewness and kurtosis data for each subscale is presented in Appendix 2. SDQ scores can be categorised into four bands: close to average; slightly raised; high; and very high (Youth in Mind 2016). The threshold scores for each banding were based on a community sample whereby 80%, 10%, 5% and 5% of U.K. children are classified into each band respectively (Youth in Mind 2016). For the purposes of statistical analysis the slightly raised, high, and very high groups are often combined (Thompson et al. 2021).

The mean scores for conduct problems, emotional problems, hyperactivity, and peer problems scores were classified as 'close to average' (previously termed 'normal') (Youth in Mind 2016). However, the proportion of participants with scores classified within the 'close to average category' were: 61% for total difficulties score; 72% for conduct problems; 58% for emotional problems; 67% for hyperactivity and 57% for peer problems (Table 6). There was a significant difference between the proportion of participants identified with a 'close to average' score and a 'clinical' score (slightly raised, high and very high) compared to a U.K. survey normative scores (80%=normal, 20%=abnormal) for the total difficulty scale and all four subscales (Chi-squared test, goodness of hit, $p \leq 0.05$) (Youth in Mind 2016). The mean total difficulties score was 13.4 (S.D.=7.0) for the total sample, 11.4 (S.D.=6.9) for male participants, and 14.6 (S.D.=6.8) for female participants. There was a significant difference between the total difficulties scores for male and female participants (Independent t-test, t=-2.3, $p=0.03$, Cohen's d=-0.3). That is, female participants had higher total difficulties scores than male participants (Table 7). Chi-squared tests for independence (Yates Continuity Correction Applied) identified no significance differences between the proportion of male and female participants with total difficulties score in the clinical range ($p>0.05$).

Table 6. Descriptive statistics for Strengths and Difficulties Questionnaire for all participants compared to expected population norms

Scale/Subscale	n	Mean (S.D.)	Median (I.Q.R.)	Close to average* (%)	Clinical** (%)	χ^2 *P* value***
Total difficulties	100	13.4 (7.0)	13.0 (8.0-18.0)	61	39	***p*=0.00**
Conduct problems	100	2.5 (1.9)	2.0 (1.0-4.0)	72	28	***p*=0.05**
Emotional problems	100	4.0 (2.7)	4.0. (2.0-6.0)	58	42	***p*=0.00**
Hyperactivity	100	4.4 (2.7)	4.0 (2.0-6.0)	67	33	***p*=0.00**
Peer problems	100	2.4 (2.2)	2.0 (1.0-3.0)	57	43	***p*=0.00**

*'Close to average' (previously termed 'normal') category contains 80% of children (Youth in Mind 2016).

**'Clinical' ('Slightly raised', 'High' and 'Very high' categories) contains 20% of children (Youth in Mind 2016).

**Statistical significance defined at 5% levels for values in bold.

Table 7. Comparison of proportion of participants in clinical group for Total Difficulties Score for sex

	n	Mean (S.D.)	Median (I.Q.R.)	Non-clinical* % (n)	Clinical* % (n)	χ^2 *P* value**
Sex						*p*=0.26
Male	39	11.4 (6.9)	9.0 (6.0-17.0)	69 (27)	31 (12)	
Female	61	14.6 (6.8)	14.00 (10.0-19.5)	56 (34)	44 (27)	

*'Close to average' (previously termed 'normal') category contains 80% of children (Youth in Mind 2016).

**'Clinical' ('Slightly raised', 'High' and 'Very high' categories) contains 20% of children (Youth in Mind 2016).

**Statistical significance defined at 5% levels for values in bold.

4.5.6. Relationship between DFA and child mental health

The relationship between DFA (measured by MCDAS total score) and child mental health (measured by RCADS total anxiety score and total internalising score, and SDQ total difficulties score) was analysed. Spearman's rank correlation coefficient was calculated for the relationship between MCDAS total DFA score and RCADS total anxiety and total internalising score. Pearson product-moment correlation coefficient was calculated for the relationship between MCDAS total DFA score and SDQ total difficulties score. A medium positive correlation was found between DFA and each child mental health variable (Table 8). This suggests that higher levels of DFA are associated with higher levels of anxiety disorders, anxiety and depressive disorders, and social, emotional, and behavioural problems. Scatter plots demonstrating a positive correlation are presented in Figures 2, 3, and 4.

Table 8. Correlations between measures of DFA and child mental health

Scale/Subscale	**MCDAS total score**	**p-value**
n=100	**Correlation coefficient**	
1. MCDAS total score	-	
2. RCADS total anxiety score	r_s=0.4	p=0.001
3. RCADS total score	r_s=0.3	p=0.0009
4. SDQ total difficulties score	r=0.3	p=0.01

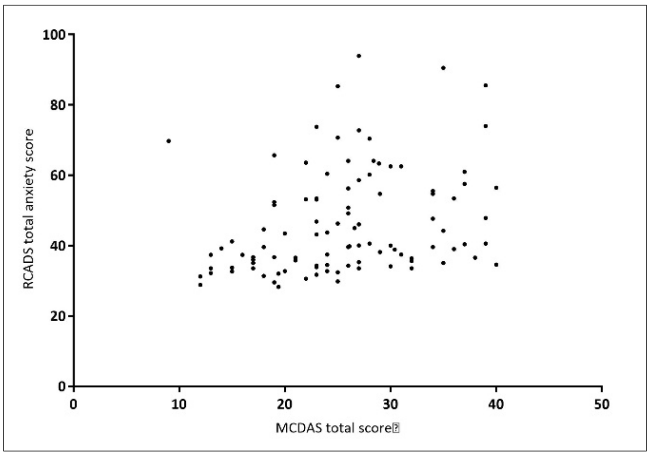

Figure 2. **Relationship between MCDAS total score and RCADS total anxiety score**

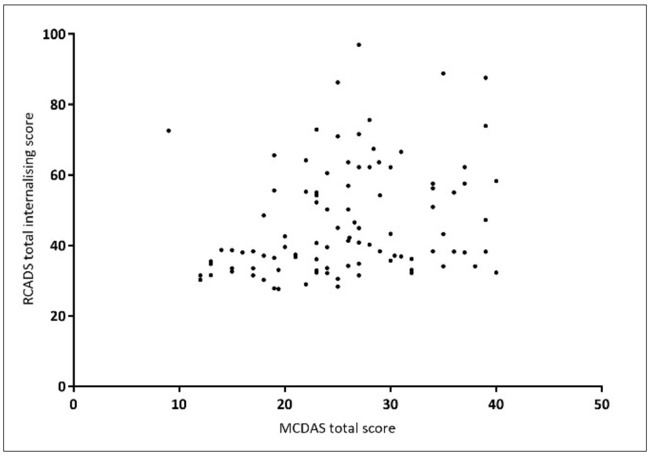

Figure 3. **Relationship between MCDAS total score and RCADS total internalising score**

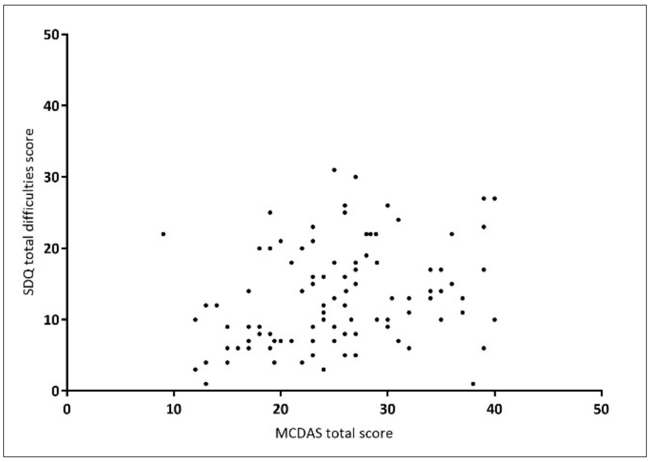

Figure 4. **Relationship between MCDAS total score and SDQ total difficulties score**

4.5.7. Daily living

Utility values were calculated from the nine items in the CHU-9D using a statistical syntax for IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation) (University of Sheffield 2019). Overall, one participant had one missing item from the CHU-9D scale and was excluded from the analysis. The utility scores ranged from 0.52 to 1.0, where 1.0 is perfect health. The CHU-9D generates utility values between 0.33 and 1 (Fuber and Segal 2015). The utility values were not normally distributed (Kim 2013). Detailed skewness and kurtosis data are presented in Appendix 2. The median utility value was 0.9 (IQR=0.8-0.9). The ceiling effect was minimal with 4% (n=4) having the maximum score of 1.0. Spearman's rank correlation coefficient was used to determine the relationship between DFA (measured by MCDAS total score) and HRQoL (measured by CHU-9D utility score). A medium negative correlation was found between DFA and utility scores (r_s=-0.4, p=0.0001). This suggests that higher DFA score are associated with lower utility scores (i.e. lower HRQoL). A scatterplot to demonstrate the negative correlation is presented in Figure 5.

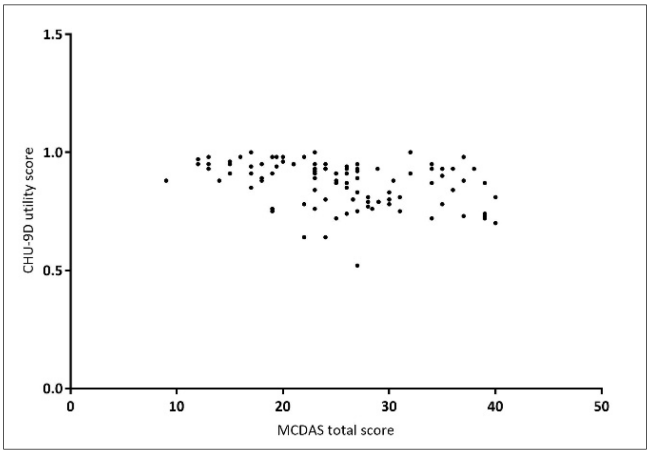

Figure 5. Relationship between MCDAS total score and CHU-9D utility score

4.6. Discussion

This study sought to further understanding of DFA in children by determining a profile of patients who would be potential users of a new DFA assessment questionnaire in clinical practice, and by evaluating the clinical utility of incorporating measures for HRQoL and child mental health screening within DFA assessment. Existing measures for DFA assessment in children have been criticised for having a narrow conceptual focus and under-representing the construct (Armfield 2010b; Porritt et al. 2013). Incorporating multiple measures has potential to increase conceptual coverage and clinical information by assessing related impacts (Schuurs and Hoogstraten 1993; Locker et al. 1996; Seligman et al. 2017).

The first objective in this study was to identify a sociodemographic profile of patients with clinical indications for DFA assessment in clinical practice. That is, children whose patient care could potentially be improved by standardised DFA assessment questionnaires. This study was carried out in a paediatric dentistry unit within a dental hospital. Children included in the patient profile had been referred to the unit from primary and secondary dental care for specialist DFA management. It is important to acknowledge that this referred patient group may not be representative of children with DFA generally. Potential differences included: referred patients may have previously experienced failed management of their DFA; require treatment for a dental problem (e.g. dental caries, traumatic dental injury); and have waited longer to receive dental treatment due to the referral (Marshman et al. 2016). Additionally, management of DFA in children within a secondary/tertiary dental clinical setting is associated with impacts for families (Marshman et al. 2016; Seligman et al. 2017). For example, parents/carers may need to take leave from their employment to bring a child to a dental hospital appointment. Consequently, factors related to the referral could increase the fear and anxiety a child with DFA experiences. That is, a child feels stress to overcome their DFA, or under pressure to please their parents, to minimise the implications on their families (Silverman and Ollendick 2005). To date, there has been a paucity of research into the impact of a hospital referral on DFA in children.

The sociodemographic profile identified that children referred for DFA management were more likely to identify themselves as being of White ethnicity and female sex; and be living in areas of high social deprivation. The ethnicity and sex profiles are not surprising. The ethnicity data mirrors that from the 2011 U.K. Census, whereby 81% of the Sheffield population identified as White British (Sheffield City Council 2021). This suggests the referral pattern corresponds to the local population. A female preponderance for DFA has also been previously identified in U.K. children, whereby female children aged 12 and 15 years were also more likely to report high DFA severity than male children in the 2013 CDHS (Health and Social Care Information Centre 2015a). There are several possible explanations for this sex difference. The first consideration is that anxiety disorders (e.g. DFA) tend to stabilise during adolescence (Hofstra et al. 2000). This tendency is greatest in females compared to males (Hofstra et al. 2000). Additionally, female children have a greater propensity for the temperamental quality of negative emotionality, which is considered a risk factor for DFA (Klingberg and Broberg 1998). Differences may also be influenced by gender socialisation and parental reinforcement, that shape gender-specific expectations of emotional expression (Chaplin et al. 2005; McLean and Hope 2010). Socialisation describes the process by which an individual learns to behave in a manner consistent with societal standards (e.g. develop gender-linked behaviours and roles) (Keenan and Shaw 1997). As expressing fears and anxieties is more consistent with the female gender role, there is evidence that parents accept, or even encourage, fear and anxiety in female children (Birnbaum and Croll 1984; Ginsburg and Silverman 2000). In contrast, male children are expected, within traditional masculine roles, to demonstrate self-confidence and overcome their fears and anxieties (Ginsburg and Silverman 2000; Chaplin et al. 2005). Ginsburg and Silverman (2000) have demonstrated that children reporting high levels of masculine traits also reported experiencing less fearfulness when compared to less 'masculine' peers. However, it is possible that children who perceive themselves in a traditional masculine role may be less willing to express their DFA, as it may suggest to others that they are weak (McLean and Hope 2010). Within traditional feminine roles, it is more acceptable to express emotions without negative consequences (McLean and Hope 2010). Therefore, it is not known whether the sex difference reported reflects a true difference in DFA experience; or is a result of differing social pressures and expectations for male and female children.

Overall, half of the participants in the profile were living in local areas associated with high social deprivation. This is not in keeping with recent national survey data, which found no relationship between DFA and being in receipt of free school meals (taken as a proxy measure for social deprivation) (Health and Social Care Information Centre 2015a). Admittedly, the patient profile differed from children with DFA who participated in the CDHS 2013 as this was a convenience clinical sample rather than a representative population sample. An important consideration is that the participants had all been referred with DFA and needing dental treatment. It is likely that children with DFA, but whom did not need dental treatment, would not be referred to secondary/tertiary care. Therefore, it is possible that socio-economic background of participants was influenced by dental caries status. As potential support, children from socially deprived backgrounds are generally more likely to have extensive dental caries and be referred to paediatric dentistry services than children from affluent backgrounds (Harris et al. 2008; Health and Social Care Information Centre 2015c; Knapp et al. 2020; Public Health England 2020). Whilst, DFA is not associated with caries experience, per se, it is associated with untreated and extensive dental caries in children (Julihn et al. 2006; Nuttall et al. 2008; Grisolia et al. 2021). Correspondingly, the most common reason children with DFA are referred to paediatric dentistry is for treatment of dental caries (Harris et al. 2008). Without recording caries diagnosis, it is not possible to identify whether the participants had concomitant high caries experience.

As all the participants in the patient profile had been referred for specialist DFA management, it would be a reasonable assumption that this patient group had high levels of DFA. An unexpected finding was that MCDAS total scores ranged from 9 to 40 (minimum score=8; maximum score=40), suggesting not all participants had high DFA severity. To date, a threshold value that represents clinically significant DFA, or categorises DFA scores into different severities, has not been determined for MCDAS scores. There is an argument against using threshold values for dimensional conditions like DFA, as they create artificial distinctions between what is considered health and disease (Youngstrom 2013). However, from a practical perspective, threshold values can aid interpretation of scores, and enable clinical prioritisation of health service resources. Although the MCDAS does not have a threshold score, a total score of 26 has been suggested for the $MCDAS_f$ for children aged 8 to 12 years, based on referral to paediatric dentistry services as the reference standard for ROC curve analysis (Howard and Freeman 2007). Applying this threshold (MCDAS≥26) to the patient profile, only half (n=50, 50%) of the participants would have been identified as having high DFA severity and requiring specialist DFA management. There are several potential explanations for this finding that warrant discussion. Firstly, there is a possibility that the participants did not have high levels of DFA, and that dental professionals refer patients without severe DFA for DFA management in secondary/tertiary care settings. Correspondingly, a recent study of children referred for dental extractions under general anaesthesia in Greater Manchester found that 35% (n=30) of children were able to complete dental treatment with local anaesthesia alone (Shepherd and Ali 2015). However, the referrals are not necessarily inappropriate, and may provide further evidence that dental professionals can identify when a child with DFA could be managed in primary dental care, or when additional support for dental treatment is needed in a specialist setting (Holmes and Girdler 2005). It is also feasible that participants had a high intensity state DFA response towards a challenging dental treatment (e.g. a difficult dental extraction) which prompted the referral, but in normal dental situations would have low baseline levels of DFA. As the MCDAS is a trait DFA measure it is likely those individuals would not have high DFA scores. The second possibility is whether the participants did have high levels of DFA but were not correctly identified as such. A final consideration is the conceptual limitations of the MCDAS as a DFA measure. The MCDAS comprises seven items relating to specific dental stimuli and procedures that may have threat potential for children with DFA (Wong et al. 1998). An eighth item relates to DFA generally (Wong et al. 1998). A short list of dental stimuli may provide an incomplete picture of DFA, potentially resulting in an underestimation, whereby, children do not identify with the items as demonstrating their DFA experience, and, accordingly, do not self-report their DFA (Schuurs and Hoogstraten 1993). It should also be acknowledged that caution is required to use and interpret the $MCDAS_f$ cut-off score, as it has only been validated for children aged 8 to 12 years, whereas the patient profile included children aged 11 to 16 years (Howard and Freeman 2007).

An important finding was that correlations were identified between MCDAS total scores and RCADS and SDQ total scores (RCADS total anxiety and total internalising scales; SDQ total difficulties scale). This indicates a relationship between DFA and psychological difficulties in children. Versloot and co-authors (2008) similarly found a correlation between CFSS-DS scores and SDQ total scores in children. Furthermore, there was an increased prevalence of emotional, social, and behavioural problems in children with DFA compared to the prevalence expected in community samples. Overall, 13% and 40% of the study participants reported symptoms that reached the borderline clinical/clinical threshold for RCADS total scores for anxiety and depression, and SDQ total score for social, emotional, and behavioural difficulties, respectively. For the RCADS, this is compared to approximately 6% of children in the reference non-clinical population from the USA (Chorpita et al. 2000; Chorpita et al. 2005; Wolpert et al. 2015). In U.K. children aged 11 to 16 years, 8% have an emotional disorder, which suggests the representative USA sample have similar levels to those in the U.K. child population generally (Health and Social Care Information Centre 2018). The SDQ threshold values are more representative as they are based on a large sample of U.K. children aged 5 to 16 years (Goodman and Goodman 2011). However, the threshold values for both measures are broadly appropriate for use with the study population.

A key finding was that anxiety and depression symptoms identified by the RCADS measure were common in paediatric dental patients with DFA. Stenebrand and co-authors (2013) reported that children aged 15 years with high levels of DFA are more likely to have an anxiety and depression symptoms than peers without DFA, although levels of depression did not reach a clinical threshold. Similarly, in a young adult population, Locker and co-authors (2001a) identified an increased prevalence of anxiety and depressive disorders in individuals with high levels of DFA. With consideration of cognitive and behavioural theories for anxiety disorders, there is a theoretical basis for a relationship between anxiety and DFA (Williams and Garland 2002). That is, it is possible that children with anxiety disorder symptoms are at increased vulnerability for the development and maintenance of DFA (Locker et al. 2001a). Whereby, if a child is already experiencing cognitions, affective, physiological, and behavioural symptoms of general anxiety, then they may be more likely to perceive a dental situation in a negative manner; or have negative cognitions and avoidant behaviours that act to maintain their DFA overtime (Williams and Garland 2002; Armfield and Heaton 2013).

In the study population, approximately 30%-40% of children reported symptoms of conduct, emotional, hyperactivity and peer problems with the SDQ measure. This was compared to 20% of U.K. children in the validation study (Goodman and Goodman 2011; Youth in Mind 2016). Versloot and co-authors (2008) identified that SDQ score is associated with uncooperative behaviour during local anaesthetic delivery for dental treatment. Identifying children who are at risk of treatment difficulties has clinical relevance. For example, shortening appointment times or use of distraction media for a child with hyperactivity problems who may have attention difficulties. However, since completion of the study, further research has questioned the suitability of the SDQ measure for children younger than 13 years (Patalay et al. 2018). Black and co-authors (2021) identified that the reading age of individual items varied from 5 to 18 years. Questionnaires with inappropriate reading ability requirements place high cognitive demands on children which act to decrease the reliability of the responses (Omrani et al. 2019).

The relationship between dental treatment acceptance and SDQ score may partly explain the discrepancy between the high proportion of participants in the study population who were identified by the referrer as needing specialist management (e.g. to accept treatment), but where the child themselves did not self-report high levels of DFA. As previously presented, DFA and BMPs in children are terms frequently used with synonymous meaning, although not all children with DFA have behaviours that disrupt dental treatment (Freeman 2007). Therefore, there is an argument that the term BMPs is ambiguous and inappropriate in the context of DFA in children. Behaviours to avoid/escape a perceived threatening situation are central to theories of fear and anxiety (Barlow 2002). By considering unhelpful behavioural coping responses in DFA as 'bad' behaviour on the part of the child, the dental profession is failing to acknowledge the fundamental role of fear and anxiety in these behaviours, and essentially placing blame on the child for a failure to complete dental treatment (British Society of Paediatric Dentistry 2011). If services are to be child-centred, then perhaps it is finally time to abandon the term BMPs for children with DFA (British Society of Paediatric Dentistry 2011; Gilchrist et al. 2013).

Unexpectedly, one in six of participants reached the RCADS clinical thresholds for panic disorder and separation anxiety disorder. This is compared to only 1% of U.K. children (Health and Social Care Information Centre 2018). As the RCADS asks children to rate the severity of their symptoms, it is possible that children's responses were influenced by the context for completing the questionnaires (DFA assessment prior to an appointment) and related to DFA rather than a concomitant emotional disorder. That is, children reported symptoms of a panic attack (e.g. 'heart beats fast', 'I feel shaky') and separation anxiety (e.g. 'I worry about being away from my parents') as part of their DFA experience. It has been suggested that the context primes children to recognise associated words when completing questionnaires (Black et al. 2021). To date, separation anxiety has not been identified as a factor for DFA in children. Separation anxiety describes fear and anxiety about separation from specific attachment figures (e.g. parents/caregivers) (World Health Organisation 2018). As the participants were new referrals to a paediatric dentistry unit, this may demonstrate their uncertainty about whether parents/caregivers would be able to accompany them. For the clinical utility of the RCADS this is interesting. It suggests the RCADS may tap into the construct of DFA in children and capture parts of the construct not previously considered in existing DFA measures. Additionally, MCDAS scores were correlated with RCADS total scores suggesting concurrent validity. However, further research would be needed to evaluate the construct validity of the RCADS as a DFA measure for children. As the symptoms of panic attacks and separation anxiety relate to thoughts and physical symptoms, it is possible that they would be captured in a measure based on a theoretical model that considers the DFA construct to be composed of cognitive, affective, behavioural, and physiological response systems.

Importantly, most paediatric dental patients with DFA in the study did not have symptoms of anxiety or depressive disorders. Moreover, symptoms of generalised anxiety disorder, obsessive compulsive disorder, and social phobia did not reach the borderline/clinical threshold in children with DFA compared to the representative population suggesting not all anxiety disorders are associated with DFA (UCLA Child First Program 2019). A potentially important confounding factor is that emotional disorders are more prevalent in children from low-income backgrounds (Health and Social Care Information Centre 2018). Although, half of the participants in the study were living in areas with high social deprivation, household income specifically was not recorded as part of the study (Health and Social Care Information Centre 2018). The prevalence of emotional problems in paediatric dental patients without DFA is also not known. It is possible that the presence of a dental condition associated with negative self-appraisal and body dissatisfaction (e.g. visible dental caries) could influence symptoms of emotional disorders (Eisenberg et al. 2003). The role of dental conditions on body dissatisfaction of children requires further attention. However, the findings of this study do suggest there are a group of paediatric dental patients with DFA and concomitant anxiety and depression difficulties. Further research is needed to understand how emotional difficulties influences the development and maintenance of DFA in children.

A limitation of this study was that the participants represented a convenience sample. Although participants were referred with DFA, not all children self-reported high levels of DFA. As the study sought to evaluate the use of multiple measures in a population where there is a clear clinical indication for DFA assessment this is acceptable for the aims and objectives. Having a control group of patients referred without DFA would have allowed adjustment for confounding factors and facilitated further statistical analysis. A potential approach to identify a control group would have been to dichotomise the sample population into low DFA and high DFA groupings based on the threshold value validated for the $MCDAS_f$.

Admittedly, each group would have had a small sample size for the further analysis. Without a control group it should be acknowledged that it is more difficult to interpret the findings. For example, the CHU-9D did demonstrate construct validity for DFA. There is also clinical relevance to assess the impacts on DFA on children's daily living to prioritise patients for treatment. However, the median utility value of 0.9 is consistent with that reported for similar community samples (Ratcliffe et al. 2012). Having a utility score for paediatric dental patients without DFA would have been useful to determine if the CHU-9D was sensitive enough to detect DFA impacts (Foster Page et al. 2014).

The aim of this study was to evaluate the assessment of HRQoL and child mental health in paediatric dental patients referred for management of DFA. Importantly, the measures used (CHU-9D, RCADS, SDQ) all demonstrated concurrent validity with MCDAS scores for DFA suggesting construct validity. The question is whether the clinical utility of using multiple measures justifies the response burden to children and dental professionals. A further potential barrier is whether the child mental health screening instruments would be acceptable to paediatric dental patients and their families. During the study, a small number of parents declined to participate as they objected to items within the RCADS measure. Unfortunately, this was not explored further, as due to the ethical requirements for the study, individuals who did not participate were not asked to provide a reason. The evidence suggests that for dental professionals to use standardised DFA assessment questionnaires in clinical practice, they would need to be brief, straightforward and require no additional scoring (Alshammasi et al. 2018). The RCADS is a lengthy questionnaire, taking approximately 25 minutes to completes, and all three measures are not straightforward to score or interpret, which may limit their application in clinical practice (Wolpert et al. 2015). The measures did provide an insight into additional problems and difficulties which children may be experiencing which could be of relevance to the dental team The findings also suggest that our understanding of children's experience of DFA is still limited and requires further consideration.

4.7. Conclusions

1. Children referred with DFA had a female preponderance, were most likely to be of White ethnicity, and lived in areas identified with high levels of deprivation.
2. Multiple factors may influence children's self-reporting of DFA severity in a clinical setting.
3. Most children with DFA do not have additional psychological difficulties and reported levels of impacts on daily living consistent with those reported in community samples.
4. DFA may share clinical features of panic disorder and separation anxiety in children.
5. The high response burden for children and dental professionals of completing multiple measures may limit the potential usage in clinical practice.

4.8. Publications arising from this chapter

Marshman Z, Morgan A, Porritt J, Gupta E, Baker S, Creswell C, Newton T, Stevens K, Williams C, Prasad S, Kirby J, and Rodd, H. (2016) 'Protocol for a feasibility study of a self-help cognitive behavioural therapy resource for the reduction of dental anxiety in young people', *Pilot and Feasibility Studies*, 2(1), 13. doi: http://doi.org/10.1186/s40814-016-0054-2

Morgan, A.G., Porritt, J.M., Baker, S.R., Rodd, H.D., Marshman, Z. (2015) 'Dental anxiety, health-related quality of life, and psychological problems in young people', *Int J Paed Dent*, 25(s1), pp. 14.

Chapter Five

5. Study 2: Exploring children's experiences of dental fear and anxiety

5.1. Introduction

As presented, the construct validity of existing DFA assessment measures for children is questionable. The most widely used measures have all been identified as having weak and narrow conceptual foundations and are at risk of under-representing the DFA construct (Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013; American Educational Research Association et al. 2014). If a measure does not provide full coverage of the construct, then not all children with DFA who complete the measure may be identified as having DFA (Locker et al. 1996). Consequently, different measures may identify different target populations (Locker et al. 1996). That is, they may only identify children with certain DFA features, and not identify others. Consequently, comparing findings between research studies and surveys is difficult, with potential implications for scientific progress (Locker et al. 1996).

The development of paediatric DFA measures with a theoretical framework based on a CBT model has been proposed to improve the construct validity of DFA assessment (Porritt et al. 2013). The theory considers that fear and anxiety responses are a construct composed of cognitive, behavioural, affective, and physiological systems (Lang 1968; Barlow 2002). At the time of the study, a single existing DFA measure was based on these four responses systems (Armfield 2010a). The IDAF-4C^{+} (core fear and anxiety module) has been used with children and found to be valid and reliable (Carrillo-Diaz et al. 2013; Baharuddin et al. 2018; Buldur and Armfield 2018). However, the measure was developed for adult DFA assessment, using a top-down approach that was based on the scientific literature and expert opinion (Armfield

2010a). The questionnaire items, wording and response scales may therefore not be relevant to children's experiences of DFA, or appropriate for use with children (Stevens 2010). That is not to dispute previous findings that support the validity of IDAF-4C+ in children. The question isn't whether it is valid or not, but rather what it is, or isn't assessing within the construct of DFA in children. There is evidence that the validity and relevance of assessment measures could be improved if children were involved in the development process (Porritt et al. 2013). Qualitative interviews with children using the CBT model as the theoretical framework to consider how each response system contributes to children's overall DFA experience, and how they may be interacting to maintain DFA over time, could inform the content of a new DFA measure.

5.2. Aims and objectives

The aim of this study was to explore the DFA experiences of paediatric dental patients with DFA aged 11 to 16 years. Specific objectives were:

- To conduct qualitative interviews with paediatric dental patients with DFA
- To utilise a CBT assessment model as a theoretical framework to inform the qualitative interviews and understanding of children's multidimensional DFA experiences

5.3. Ethical approval

Ethical approval for the research was granted by the NRES Committee York and Humber: Leeds West REC (13/YH/0163).

5.4. Study design

The Consolidated Criteria for Reporting Qualitative Research checklist was used to inform reporting of the study design and findings (Tong et al. 2007).

5.4.1. Theoretical framework

The theoretical framework used for DFA assessment within this study is the Five Areas™ CBT model (Figure 6) (Williams and Garland 2002). This is based on a conventional CBT model but uses simplified language to replace the psychotherapeutic CBT terminology. Broadly, CBT is a structured assessment and treatment approach that considers the interactive relationship of thoughts, behaviours, and physiological symptoms to the emotional state of an individual (Kendall 1985; Williams and Garland 2002). Within Five Area™, situational factors are the fifth system/area. These are the external factors that surround a child and influence their fear and anxiety (e.g. dental treatment requirements) (Williams and Garland 2002). Individual situation factors have a role in the experience of DFA, but it is the child's perception of the situation, rather than objectively what it is, that is crucial (Townend et al. 2000; ten Berge et al. 2002a).

The four internal response systems (thoughts, feelings, physical symptoms, behaviours) are highly interactive and together result in the experience of fear and anxiety (Craske and Craig 1984). However, the response systems are also capable of differential responding (Craske and Craig 1984). Concordance between systems is increased in situations associated with strong emotional responding and high levels of fear and anxiety, and disconcordance increased in situations associated with weak emotional responding and low levels of fear and anxiety (Craske and Craig 1984). Therefore, measures of DFA should consider each response system to provide a complete picture of the DFA construct (Schuurs and Hoogstraten 1993; Armfield 2010b).

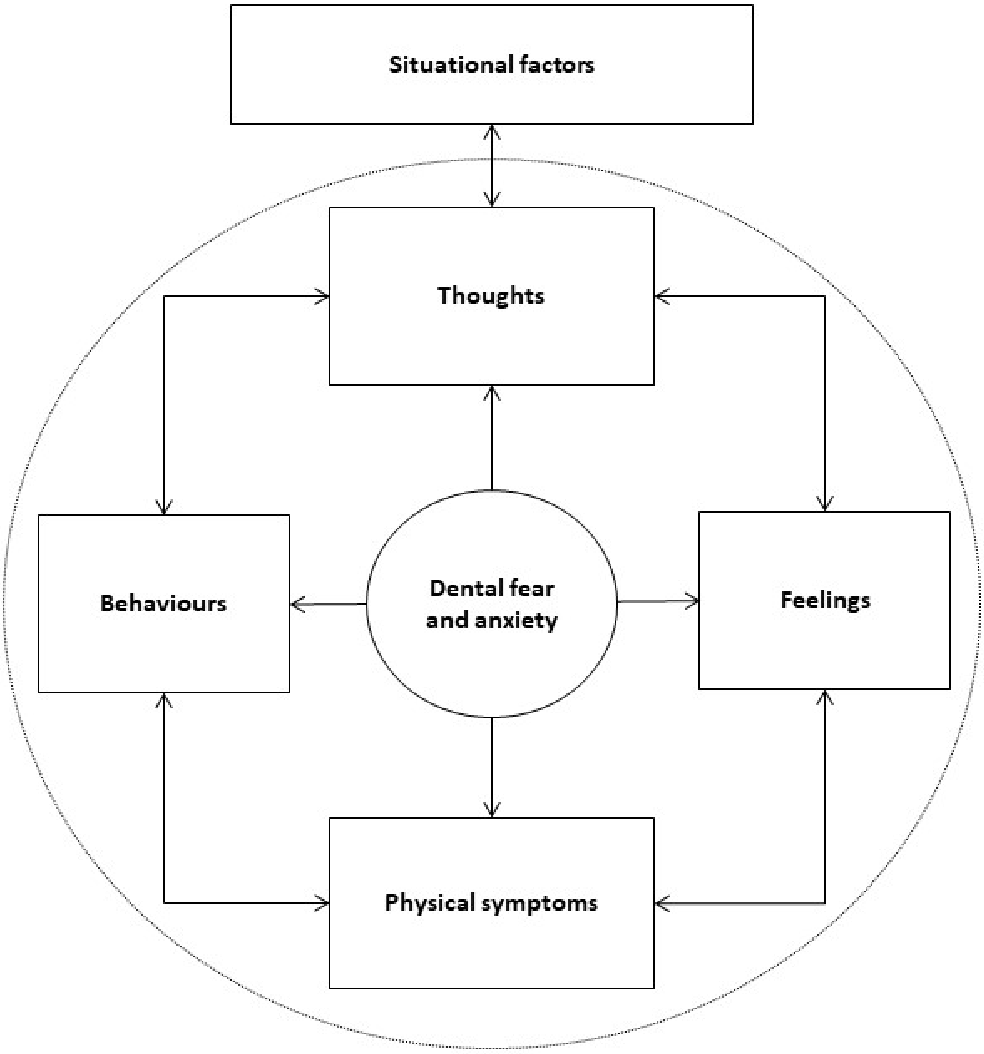

Figure 6. Five Areas™ assessment framework for DFA in children

Adapted from Williams and Garland (2002)

5.4.2. Participants

Participants comprised children aged 11 to 16 years with DFA. The age range was selected to recruit participants with sufficient cognitive development and maturity to be able to reflect on their experiences of DFA within a CBT assessment framework (James et al. 2015). Participants were recruited from two general dental practices, the community dental service and a paediatric dentistry unit within an NHS dental teaching hospital in Sheffield. Potential participants were identified and initially approached by the dental professional providing their management. The nature of the study was described verbally, and parents/carers and children were also given information sheets about the study. Permission was sought for a researcher (A.M.) to make contact again by telephone in two weeks to enquire whether they had chosen to take part. Participants were purposively sampled to provide a range and diversity of DFA experiences. The key participant demographic characteristics used for sampling included: sex; age; dental care setting (e.g. primary dental care, community dental service, dental hospital); different socio-economic backgrounds and ethnicity. Classification of ethnic group was based on the categories used by the Office of National Statistics (ONS) for measuring equality during national surveys, as previously described (see section 4.4.1) (Office of National Statistics 2016). A sampling matrix was used to monitor the recruitment of participants against key background characteristics. Children with additional communication needs were excluded (e.g. children whom interpreting services were required).

5.4.3. Data collection

Face-to-face, semi-structured interviews were conducted with participants. The option was given of the interview being held in the participant's home or in a non-clinical room in the School of Clinical Dentistry in Sheffield. Children could also be interviewed with their parent present or independently, depending on the preferences of the child and/or parent. The first interview was carried out by one female researcher (Z.M.), with a second female researcher present (A.M). All subsequent interviews were conducted by the second researcher (A.M.). The researcher (A.M.) is a Specialist in Paediatric Dentistry and completed a NatCen learning course on Depth Interviewing Skills before the study commenced. After the initial three interviews were completed, both researchers met to undertake a debriefing process. A topic guide was included to support the interviews, informed by the Five Areas™ Model of CBT (Williams and Garland 2002). The topic guide is presented in Appendix 3. The specific content of the topic guide was reviewed and modified by two expert panels: the first contained five psychologists and psychiatrists with extensive experience of anxiety, DFA and child-centred research; and the second panel comprised two patient representatives from Healthwatch England. During the interviews, the topic guide was used to prompt discussions, and the participants were freely able to discuss and explore other views and ideas that developed. As the interviews were conducted on a conversational basis, parents/carers, when present, were able to make contributions to the discussions. These additional comments were not included in the analysis but did act to aid interpretation. In addition, participants were asked further questions about the possible content and format of a proposed low-intensity CBT intervention for DFA that was currently under development. These finding are not included or analysed within this study. At the end of each interview the participant was given a £10 gift voucher as gratitude for their contribution. The interviews were recorded using a digital sound recorder (Digital Voice Recorder WS-813, Olympus U.K. and Ireland) and then transcribed verbatim. A.M. transcribed the interviews for participants 1, 2, 3, 4, 5, 6, 12, and 13; E.G. transcribed the interviews for participants 7, 9, and 10; and Dictate2us (Manchester, U.K.) a professional transcribing service, was used to transcribe the interviews for participants 8 and 11. Participant identifiable information was not included within the interviews prior to transcription (e.g. by the use of pseudonyms), and all identifying information was removed from the completed transcripts to maintain the anonymity of participants, dental professionals, and dental services. Field notes, written immediately after each interview, were used to provide context to the audio-files. As this was a qualitative exploration a formal sample size calculation was not appropriate, with recruitment continuing until no new ideas emerged and saturation of information was achieved.

5.4.4. Data analysis

Recruitment of participants, data collection and analysis were conducted concurrently. Thematic analysis was conducted using a hybrid of deductive (a priori based on the topic guide) and inductive approaches (Fereday and Muir-Cochrane 2006). Four researchers (A.M., Z.M., J.P., and H.R.) completed the initial familiarisation stage with the first five transcripts. Each researcher independently read and reviewed the transcripts to identify important and repeating ideas that emerged from the data. Any disagreements in interpretation were resolved through discussion. The data were organised into themes and subthemes underpinned by the Five Areas™ Model (Williams and Garland 2002). Subsequently, each section of the transcripts was systematically reviewed, labelled, and indexed onto a Microsoft Excel Office 365 database (Washington, Microsoft Corporation). Finally, a thematic framework was developed whereby evidence to support the subthemes was traced to the original text from each participant (Smith and Freeman 2010). Following analysis of the first five transcripts, further interviews were conducted. For each subsequent transcript additional discussions were carried out to fully elucidate and refine each identified theme and subtheme, until a stage was reached where no new ideas emerged, and data saturation was accomplished. Data for the sociodemographic variables were transferred manually to a Microsoft Excel Office 365 database (Washington, Microsoft Corporation), and simple descriptive analysis was conducted. To determine deprivation quintile, postcode data were converted to a deprivation rank using the GeoConvert function on the U.K. Data Service Census Support website, based on postcode data from the 2011 Census and the English Indices of Multiple Deprivation 2015, as previously described in Section 4.4.1. (Bryan et al. 2014; U.K. Data Service Census Support 2019).

5.5. Results

5.5.1. Sociodemographic variables

Overall, 17 families were approached, with interviews conducted with 13 children between January and April 2014. The families of the four children who declined to participate were not asked to provide an explanation for their decision. The age of the participants ranged from 11 to 15 years. Ten participants were female. Overall, 31% (n=4) of children lived in LSOA identified as the most deprived of England (quintile 1). Most (92%, n=12) participants identified their ethnicity as 'White'. Although all participants were recruited from dental services within Sheffield, the participants came from a wide geographic area including Derbyshire, Lincolnshire, Nottinghamshire, and South Yorkshire. All interviews, except two (participants 1 and 3) were held in the participant's own home. Only one participant

(participant 7) chose to be interviewed without a parent present, with nine conducted with the mother present, two with their mother and father present, and one interview with a paternal grandmother present. The characteristics of the participants are detailed in Table 9. Participants 2, 3, 4, 5, 7, 8, 10, 11, 12 were not previously known to the researcher (A.M.) conducting the interviews. For participants 1, 6, 9, and 13, researcher A.M. had provided clinical support and supervision within the role as a consultant in paediatric dentistry to the dental professional providing their treatment, prior to interview.

Table 9. Sociodemographic characteristics of participants in Study 2

Interview	Recruitment location	Sex	Age (years)	Deprivation quintile*	Ethnicity
1	Dental Hospital	Female	11	5	White
2	General Dental Practice	Female	15	1	White
3	Dental Hospital	Female	11	1	White
4	Dental Hospital	Male	14	2	White
5	Dental Hospital	Female	12	3	White
6	Dental Hospital	Male	13	3	White
7	Dental Hospital	Female	11	4	White
8	General Dental Practice	Female	14	1	White
9	Dental Hospital	Female	12	4	White
10	Salaried Dental Service	Female	13	4	White
11	Dental Hospital	Female	14	4	White
12	Salaried Dental Service	Male	14	1	White
13	Dental Hospital	Female	13	4	Other ethnic groups

*1=most deprived; 5=least deprived

5.5.2. Themes and subthemes

As previously described, the topic guide employed during the face-to-face interviews and the thematic analysis, were guided by the Five Areas™ model of CBT (Williams and Garland 2002). Correspondingly, the five domains according to the Five Areas™ are thoughts, feelings, physical symptoms, and behaviours. The detailed results are described for each domain with quotes to illustrate each theme presented and summarised in Figure 7.

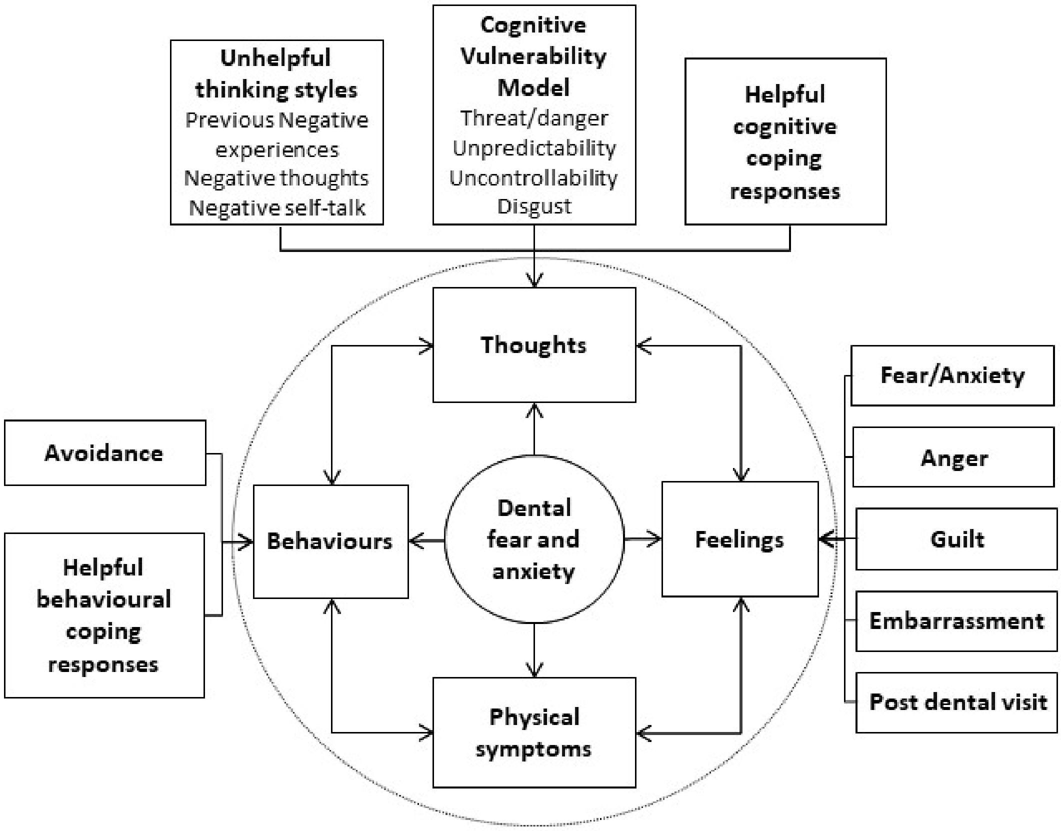

Figure 7. Conceptual diagram for children's experiences of DFA based on the Five Areas™ model of CBT

(Williams and Garland 2002)

5.5.3. Thoughts

Within this domain, three main themes were identified: unhelpful thinking styles; the Cognitive Vulnerability Model; and cognitive coping responses.

5.5.3.1. Unhelpful thinking styles

Previous negative experiences

Children discussed negative dental experiences that they considered as being important to their DFA. Previous studies have demonstrated that children with DFA often report painful or traumatic dental experiences, and that they can act as conditioning events in the development of DFA (Townend et al. 2000; ten Berge et al. 2002a; De Jongh et al. 2003). However, there is evidence that it is what children think about a what happened that is crucial for DFA, rather than the objectively whether something was painful or traumatic (Townend et al. 2000; ten Berge et al. 2002a).

'I think it is really what has already happened. I was always worried they were going to hit one of my nerves. It happened.' (Participant 6)

'And they had to like bring people to hold me down and stuff. And I was like wrestling with them and stuff. I was like terrified because they take out the needle like really slowly like that. And it's scary and stuff.' (Participant 13).

The negative experiences described had individual differences. However, the dental team played a significant role for some participants, whereby it was perceived they had acted dishonestly, or not listened to requests for help.

'I think I told them I said my tooth was a bit wobbly and they said they were just going to like do a check-up and they pulled it out.' (Participant 2)

'When I had my needle I said when I put my hand up can you stop, he said yeah, and then when I put my hand up they didn't, they went on.' (Participant 3)

Thoughts about previous negative dental experiences were persistent and relived by participants over time.

*'Yeah, and then for about a year after I had it done it's kind of, it's still the same memories was going around in my head, the same day every night.' (*Participant 9*).*

Negative thoughts

Participants discussed what they thought would happen at a dental visit and made catastrophic inferences about worst possible outcomes. They discussed receiving bad news; experiencing unimaginably painful treatment; or that a clinical error could occur and cause them harm.

'What I feel always in my head is like what if I go and say I got like gum cancer. And I don't even know what that is. I just know it's gum cancer.' (Participant 8)

'What if they do something wrong? They slip, and then I swallow something and it chokes and I die.' (Participant 13)

Participants also believed that dental professionals had made negative judgements about them; that they did not want to provide dental treatment to them, thought badly of them, or would fail to believe them when they told the truth about sugar consumption.

'People just get sick of me because I'm taking ages because I don't want it.' (Participant 3)

'So if I've got unhealthy teeth they might think she constantly eats junk food. She must be like a really lazy person and things like that'. (Participant 6)

'Like they're just going to say, always eat sweets, like always gets sugar. But I don't.' (Participant 7)

Negative self-talk

Self-talk describes one's internal dialogue accessible to conscious awareness (Prins 1985). There is evidence that increased levels of anxiety is associated with increased negative self-talk in children (Lodge et al. 1998). Participants made statements, that were suggestive of negative beliefs and self-talk. Generally, participants placed blame on themselves for their DFA.

'Because my teeth are not the best.' (Participant 7)

'Because I'm scared of everything. If you ask any of my friends, I am scared of everything.' (Participant 6)

'Everyone around me just gets on with it, it is just me sometimes.' (Female participant, age 11 years)

5.5.3.2. Cognitive Vulnerability Model

Corresponding to the model, children described perceptions of danger, unpredictability, uncontrollability, and disgust related to their experiences of DFA.

Threat/Danger

Study participants described threatening stimuli in terms of their sensory experience of it. That is, what they heard, saw, or felt. These included noises produced by dental equipment, the sight of sharp instruments, feeling unable to breathe with materials placed in, or over, their mouths, and hearing distress in other patients.

'No, I don't like that drill. Because it's proper loud.' (Participant 8)

'When the tooth was being pulled out you could hear it like cracking. Which is not very nice.' (Participant 4)

'I couldn't quite breathe, but then they told me to breathe through my nose and I did.' (Participant 3)

'When I first go in, I go through the waiting room area, and I go in and I see like little kids screaming and stuff from like the pain.' (Participant 13)

Unpredictability

Participants found dental visits to be unpredictable. This was seen as a failure in communication by dental professionals.

'It made you slightly more scared because he wasn't talking about what he was doing. He only said like in a couple of, like, I am going to do this, and didn't like say how he was going to do it.' (Participant 1)

Generally participants wanted the dental team to tell them what was going to happen during a dental visit and did not want anything kept hidden from them. It was also described as important to have this information explained in an age-appropriate manner, whereby children did not feel patronised.

'Well tell me like exactly what they would do, cause I don't like surprises.' (Participant 12)

'She was just annoying me. Talking to me like I was five.' (Participant 10)

Conflicting views were expressed about how much information should be provided. Some participants described when they knew what to expect (i.e. predictability increased) they were better able to manage their DFA responses, whilst others did want more information provided, but then found it unhelpful for their DFA.

'Yeah. Because did you know her, when you are sitting down it's like it's going to take for ages and it really hurts, but when people tell you what time it's going to be you feel better like it isn't going to take that long so just hold it in.' (Participant 13)

Interviewer: "Some people have said they like to see everything beforehand, and have it explained to them how everything works."

Participant: "I do, but then I just get upset and don't want it." (Participant 3)

Once participants had been given information about what to expect, they then became intolerant to any unexpected changes, such as provision of different dental treatment.

'They did one (injection) and then I was like really relieved and happy it was done, and then they were like, "Why don't we do 3 more", and I was like "err".' (Participant 4)

Participants also described seeking information about dental treatment from sources other than the dental team. Although parents were identified as a possible source of information, participants placed importance on the dental experiences of peers.

'Because I didn't know what to expect so like, stories I've heard have been horrible, well it wasn't just one person it was like loads of different people. They were like it's the biggest needle you'll ever see, so.' (Participant 4)

Uncontrollability

Variation was reported of perceptions of control during dental visits. Some children described the perception of having no control during dental treatment.

'No, cause they are like in my mouth, and I can't talk.' (Participant 7)

However, participants also sought control by negotiating and bargaining with the dental team, or by withholding their consent for dental treatment.

'I said I was going to pull it, but they wouldn't let me.' (Participant 3)

'Understand that they're not the ones in control. You are. You can say yes, and you can say no.' (Participant 11)

Disgust

The role of the emotion disgust in DFA is not well understood (Armfield et al. 2008). Children did discuss aspects of dental care they found unpleasant.

'As soon as we open the door, she was, "Oh god. That's smell." We got off the bus and we cross the road she will say, "I can smell it already".' (Mother of Participant 8)

'Because when my friends have teeth out they say it was really horrible and there was blood every-where, and it was really gross and stuff.' (Participant 4)

5.5.3.3. Helpful cognitive coping responses

Participants described efforts to distract themselves from their DFA using both cognitive self-control and music through earphones.

'I just shut my eyes and like, and not to be stupid, just pretend that you're in a happy place. On beach with the sea trickling along.' (Participant 5)

'There's not really a lot of thought behind it. The more you think about it, the worse it gets.' (Participant 11)

'Listening to music helped cause then I couldn't hear it. Well I like Taylor Swift, so I was listening to her. Like sort of took my mind off it.' (Participant 4)

Participants also utilised their previous learning experiences to manage their DFA-related cognitions.

'Yeah, it has affected my life a lot. Because once I've gone through it once I can go through it again and again. I just get bored with it and say right it's not that bad I can just go through it.' (Participant 13)

5.5.4. Feelings

Within this domain, five subthemes were identified: fear/anxiety; anger; guilt, embarrassment; and post dental visit.

5.5.4.1. Fear/anxiety

Participants used many emotive words to illustrate their fear and anxiety feeling states during their accounts. These included: dread; flustered; nervous; petrified; panicked; trapped; terrified; uncomfortable; and worried.

'But when I was a little kid and I walked though I saw like little kids going like AHHHHH like that and I was terrified. Petrified of it.' (Participant 13)

They described screaming as a response to their fear during their dental treatment.

'I screamed when they did it and stuff.' (Participant 4)

'*I screamed the place down.'* (Participant 3)

Participants also spoke about attempts to hide their fear and anxiety responses out of concern for others.

'Cause if I feel worried, I'm scared my Mum will feel worried.' (Participant 6)

5.5.4.2. Anger

Participants described feeling angry in relation to their DFA. This strong feeling response was identified before dental appointments, during dental visits, and post dental experiences.

'I get mardi before. Like angry and snappy.' (Participant 8)

'Last time I nearly hit somebody...on purpose. I got really annoyed like when people mess around with you like this, pulling your face and like opening your mouth and stuff, it gets really annoying so I was like stop it! You want to hit them and stuff.' (Participant 13)

'Like I'm just going to punch someone.' (Participant 7)

'Angry...Because they didn't listen. They lied. I wanted to shout at them, "So why didn't you listen.' (Participant 3)

5.5.4.3. Guilt

Participants discussed that other people made statement that left them feeling guilty for refusing treatment, but that it did not then influence their actions.

'Yeah, there were teachers saying stuff like if you don't get it done then people are going to have to pay thousands of pounds and making me feel really guilty. But I just kind of ignored them.' (Participant 4)

5.5.4.4. Embarrassment

Some participants were embarrassed by their DFA, comparing themselves unfavourably to their peers, or because of what they perceived others thought of them.

'I was a bit embarrassed…Other children kind of made me feel a bit, because I could see children that were younger than me having like the same thing as me done, and they were like having it done and I was just there crying, so I felt a bit that the children were more putting me down.' (Participant 9)

'Participant: They might like be annoying and start shouting.

Interviewer: And what might the dentist say if they shout at you?

Participant: Stop being silly.' (Participant 7)

5.5.4.5. Post dental visit

After dental visits, participants experienced a range of emotional responses. They described feeling exhausted and drained by the high emotional intensity of their experience.

'I feel like a ragdoll that's been messed with.' (Participant 8)

If the visit had been successful, participants used words such as proud and relieved, to explain their emotions. They discussed happiness at being perceived favourably by others.

'I opened my mouth to let him do it for the first time in a long time, and then he did this and all he was having was a look and obviously I felt over the moon. I was so proud of myself that Dad was happy with me as well.' (Participant 9)

However, some participants immediately began to think about their next dental experience, which resulted in the feeling of dread.

'Once they said it was out I was like happier after, but I was like dreading the next time.' (Participant 4)

Participants also described their relief from anxiety when dental treatment had been avoided. They described feeling pleased and having a weight lifted from them.

'Interviewer: When your Mum said you didn't have to go, she was going to cancel your appointment, what did it feel like then?

Participant: Just like a weight lifted off your shoulder' (Participant 12)

5.5.5. Physical symptoms

During an episode of DFA participants experienced physical symptoms that are characteristically associated with increased muscle tension and arousal of the autonomic nervous system. Symptoms described included: '*sweating and shaking*'; '*clammy palms*';

‘*having butterflies*’; ‘*stomach-aches*’; ‘*feeling sick*’; and ‘*becoming pale*’. Other somatic manifestations were sleep disturbances, and symptoms of temporomandibular dysfunction, including tooth clenching and mandibular pain.

5.5.6. Behaviours

Within this domain, themes of avoidance and helpful behavioural coping responses were identified.

5.5.6.1. Avoidance

Strategies were employed by participants to avoid dental care. Avoidance attempts were made at every stage of a dental experience. Before appointments, children spoke of efforts to cajole their parents/carers into cancelling dental appointments. This included attempts to deceive their parents/carers by claiming to be feeling unwell, or by down-playing dental problems.

‘Interviewer: “Have you ever made excuses not to go to the dentist?”

Participant: “Tried to. Like I’m poorly and I can’t go. I feel ill”.’ (Participant 2)

Participants also tried to delay sitting in the dental chair, by asking siblings to take their turn first.

‘Normally if it is all three of us going I’d say like I’m going last, because like I don’t go first, cause I want to see what happens to my brother and sister first. Because if like what happened before, if that like happens to them then I’m like oh I’m leaving now.’ (Participant 6)

Once in the dental chair, participants identified that dental care could be avoided by refusing to open their mouths

‘They can’t force your mouth open or anything, so I thought to myself, ‘Well if I keep it shut they can’t really do anything’.’ (Participant 9)

'Every other time I did the injection I'd like open my mouth and I'd close it again' (Participant 4)

Participants also discussed being discourteous, with the aim it might stop dental treatment being provided.

'She's like, if you want me to help you, then I'm like I don't want you to help like I don't want to be here, just things like that.' (Participant 8)

5.5.6.2. Helpful behavioural coping responses

Participants discussed needing reassurance from others. This included parents/carers, friends, teachers, and the dental team. Talking about fears and anxieties, reassuring smiles, and physical contact (e.g. holding a hand) were all identified as helpful.

'If I'm in school I tell my friends and my teachers.' (Participant 3)

'She [parent] looks at me and she smiles and she sticks her tongue out.' (Participant 6)

'They let me hold her [dental nurse] hand and stuff.' (Participant 4)

However, some participants found parental anxiety an additional burden to their DFA.

'Participant's Mother: For some children they want to have their Mum to hold their hand, but my anxiety did definitely have an effect on [participant] as well.

Interviewer: So what made the difference when your Mum wasn't in the room?

Participant: There was not so much negativity surrounding it.' (Participant 11)

5.6. Discussion

This study is the first to ask children directly about their DFA experiences. Scales and questionnaires have been frequently used in scientific studies to identify children with DFA. Correspondingly, research into DFA in children is dependent on the validity of measures, and how the DFA construct is operationalised by them (Armfield 2010b). That is, measures, by the nature of their use in research, define the concept of DFA in children (Armfield 2010b). To date, most published research into DFA has used questionnaires that have a been developed for adults, or from the adult perspective on DFA in children (Marshman et al. 2007). Therefore, our understanding of DFA in children has been influenced from the standpoint of adults. As it is children who are experiencing DFA, it is important to ascertain their expert knowledge and perspective on their own experiences, otherwise what is being assessed by measures may be an incomplete, or even inaccurate, representation of the DFA construct in children (Larsson et al. 2018).

The study was based on semi-structured interviews with 13 children. A purposive sampling approach was used to identify participants who would provide a range and diversity of DFA experiences. During the study it was challenging to recruit participants from certain population groups, most notably, children from ethnic minorities, who are under-represented in the study population. A further barrier to the inclusion of some children from ethnic minority backgrounds was the exclusion of children and families where interpreting services were required. It should be acknowledged that the inclusion of children in qualitative research where English is not the preferred language, and the research team lacks the necessary language skills, has multiple complexities (Plumridge et al. 2012; Premji et al. 2020). However, there is evidence that patients from ethnic minority groups have multiple barriers to obtaining dental care (e.g. communication barriers) which could have influenced a different DFA experience from the children in the study population (El-Yousfi et al. 2020).

Participants in the study discussed their multidimensional DFA experiences. The findings support the use of the Five Areas™ CBT model as a theoretical framework for DFA in children. In this study a hybrid of deductive and inductive approaches was used for data analysis. Children described their DFA related thoughts, feelings, physical symptoms, and behaviours. Situational factors, which is the fifth area of the Five Areas™ CBT model, was included in the topic guide, and participants discussed the people and situations they perceived had contributed to their DFA (e.g. negative dental experiences) during their accounts. As the evidence suggests it is what children *think* about dental situations, not what objectively happened, that is crucial for DFA development and maintenance, these factors were included within the 'Thoughts' domain during the analysis (Townend et al. 2000; ten Berge et al. 2002a).

The thoughts, feelings, physical symptoms, and behaviours experienced by children with DFA corresponded well with the CBT principles on the characteristic changes in these areas associated with anxiety (Williams and Garland 2002). In brief, children experienced unhelpful and negative thoughts, which focused on previous negative dental experiences, catastrophic thoughts about future dental care, and their beliefs that their dentist thought badly of them, and that they were to blame for their DFA. Children's behavioural coping strategies were also largely unhelpful for their DFA, with efforts to avoid dental care predominating. These included: convincing their parents/carers to cancel appointments; refusing to open their mouths; and employing rudeness to prompt the dental team to abandon the visit. Children also experienced distressing emotional and physical responses due to DFA, with feelings of fear, anxiety, anger, guilt, and embarrassment, and symptoms consistent with arousal of the autonomic nervous system.

Although the Five Areas™ CBT model considers responses that are maintaining anxiety, components of another theoretical model for DFA acquisition, the Cognitive Vulnerability Model (CVM) (see section 2.5.3.2.), were also identified during the data analysis (Armfield et al. 2008). Key to the CVM is a theorised schema for vulnerability. This contains perceptions that a dental stimulus or situation is dangerous, unpredictable, uncontrollable, and disgusting. Activation of the vulnerability schema then drives further cognitive, affective, behavioural, and physiological DFA responses. Although the CVM has been proposed as a model for DFA acquisition, there is some evidence to support its use as an exploratory model for the maintenance of established DFA (Edmunds and Buchanan 2012). That is, negative thoughts associated with DFA maintenance include CVM components. Correspondingly, participants described thoughts relating to danger/threat, unpredictability, uncontrollability, and disgustingness within their dental experiences.

Participants particularly identified the unpredictability of dental visits being of high importance. Interestingly, in adult studies, unpredictability has not been found to have a significant relationship with DFA maintenance (Armfield et al. 2008; Edmunds and Buchanan 2012). A proposed explanation is that as familiarity with the dental setting increases, and dental visits become more predictable, perceptions about unpredictability are less relevant for DFA (Edmunds and Buchanan 2012). Similarly, there is evidence that it is possible to promote dental habituation in children through frequent exposure to the dental environment, whereby there is a reduction in DFA during successive dental visits (De Menezes Abreu et al. 2011; Carrillo-Diaz et al. 2012; Ramos-Jorge et al. 2013). However, it is possible that DFA creates barriers to habituation in children, with evidence that children with DFA are less likely to have dental treatments (e.g. prevention) that provide learning opportunities and decrease the unpredictability of dental experiences (Seligman et al. 2017).

Children's perceptions of unpredictability in their dental experiences could also be considered a failure of communication, whereby dental professionals did not adequately explain and prepare children for dental treatments. A further difference between adults and children with DFA that may influence the predictability and communication factors of dental visits, is the legal need for dental professionals to obtain valid consent from adult patients prior to dental treatment (General Dental Council 2013). This includes a requirement for the patient to be informed about their treatment. For children, the legal duty is for consent to be obtained from the individual bearing parental responsibility for the child, with no requirement to obtain child assent (General Dental Council 2013; Herbst 2020). The available evidence suggests that dental professionals generally allocate little time to discussing specifics of a dental visit with children and that children do not have knowledge of what is going to happen to them (Watson 2010; Rodd et al. 2014). To date, there is little ethical guidance about obtaining child assent and mutual agreement for dental treatment, and whether this has an influence on DFA in children (ten Berg 2008).

There is evidence that young adults with high levels of DFA are generally more likely to perceive their interpersonal interactions with the dental team negatively, and that DFA reduces the establishment of trust in dentist-patient relationships (Jaakkola et al. 2014). Correspondingly, dental professionals who did not communicate effectively before dental procedures were seen as being dishonest, with participants reporting their dentist had lied to them. For dental professionals, there are potentially significant occupational stressors to complete dental treatment for children with DFA within the allocated appointment times (Jones and Huggins 2014). Dentists have also been found to give less direction, and are more likely to use force, when children with DFA resist treatment (ten Berge 2001). This suggests that the relationship between dentists and children with DFA may be more susceptible to breaking down with anxiety contributing to both parties. The use of novel communication tools to support dental teams in their interpersonal exchanges have shown potential to reduce state DFA before appointments in children (Jones 2015; Yee et al. 2017).

Children also reported experiencing DFA about being reprimanded by a dental professional about their oral health. There is evidence that experiencing shame during dental interactions has a negative influence on the development of trust between dental teams and patients and is likely to be counter-productive to relationships (Yuan et al. 2020). Therefore, the responsibility is on dental professionals to avoid embarrassing or shaming children during their dental visits. Although not specifically explored, it is possible that children with DFA are at increased susceptibility to perceive caries prevention advice provided by dental professionals as criticism. Recently, the use of motivational interviewing, with a focus on an empathetic communication style, to change how dental teams traditionally talk to parents/carers about oral health, has been found to be successful in changing behaviours and reducing dental caries in children (Pine et al. 2020). There is considerable scope to develop skills congruent with motivational interviewing of dental teams to promote positive communication and interactions with children with DFA (Jones and Huggins 2014).

Out of the CVM components, there has been the least research into the role of disgust perceptions in DFA in children. Conceptually, the function of the emotion disgust is the avoidance of contamination (Cisler et al. 2009). Studies of small animal phobias in children have suggested that disgust promotes fear and anxiety, and that if a child is experiencing fear and anxiety, they are more likely to find a stimulus disgusting (De Jong et al. 1997; Muris et al. 2012). There is also evidence to suggest disgust contributes to blood injury injection phobia (Koch et al. 2002). Interestingly, the perception that dental treatment would be disgusting was based on what participants had been told by peers. Correspondingly, there is evidence that disgust can be learnt from others (De Jong et al. 1997). However, further research is needed to explore dental stimuli that may have disgust potential, and the contribution of disgust-specific thoughts, feelings, physical symptoms, and behaviours in DFA in children.

Avoidance of dental care was the key behavioural change associated with DFA in the study. In adults, dental avoidance can manifest as irregular dental attendance or missed appointment (Cohen et al. 2000). However, children are not independently able to decide to attend dental appointments, and avoidance of dental care included study participants refusing to open their mouths and employing rudeness to force the dental appointment to be abandoned by the dental team. There is evidence that children with DFA are more likely to refuse dental treatment (Humphris and Zhou 2014). In Berggren and Meynert's (1984) biopsychosocial model, avoidance of dental care, as a maladaptive coping response, is central to the maintenance of DFA in adults. In this model DFA is associated with avoidance of dental care, deterioration of oral health, and the experience of shame and embarrassment. Similarly, Buchanan (2017) has described that the way a child responds when faced with an adverse dental situation is important in the maintenance of DFA in children. That is, if a child employs a maladaptive coping response (e.g. an action that stops dental treatment) they do not have the opportunity for further learning which can exacerbate the DFA experienced (Zimmer-

Gembeck and Skinner 2016). Moreover, if a maladaptive coping response is successful, at least in the short term, in achieving a reduction in DFA (e.g. if treatment is abandoned), it is more likely to be used as coping responses again and act to maintain the DFA experienced in the longer term (Heszen-Niejodek 1997). However, a further consideration is that avoidance behaviours, such as patient rudeness, may be challenging for the dental team to deal with. This is supported by evidence that dentists are more likely to use 'put-downs' during their communication with children with DFA, than children without DFA, and children who demonstrated BMPs are more likely to have their treatment abandoned by dental professionals (Klingberg et al. 1994b; ten Berge 2001). Correspondingly, dental professionals may have a role in facilitating and reinforcing children's unhelpful behavioural responses. Traditionally, dental professionals have employed behavioural management strategies (e.g. rapport-building, voice-control, modelling) to support children to employ helpful coping strategies within dental situations (Newton et al. 2012). However, the use of CBT to help individuals identify and address unhelpful thinking styles and behaviours that make a situation worse for them offers an evidence-based approach for the treatment of DFA (Newton et al. 2012; Buchanan 2017; Marshman and Williams 2017). In recent years, a CBT self-help guide for children with accompanying resources for dental teams has been developed with good initial results based on the findings from this study (Porritt et al. 2016).

5.7. Conclusions

1. Children describe DFA as a multidimensional experience.
2. The Five Areas™ CBT model is applicable as a theoretical framework for DFA assessment that considers children's thoughts, feelings, physical symptoms, and behaviours associated with DFA.
3. Perceptions of vulnerability are important in children with DFA
4. Children with DFA have a high need for age-appropriate preparatory information
5. The communication skills of dental teams can influence children's DFA experience

5.8. Publications arising from the chapter

Marshman Z, Morgan A, Porritt J, Gupta E, Baker S, Creswell C, Newton T, Stevens K, Williams C, Prasad S, Kirby J, and Rodd, H. (2016) 'Protocol for a feasibility study of a self-help cognitive behavioural therapy resource for the reduction of dental anxiety in young people', *Pilot and Feasibility Studies*, 2(1), 13. doi: http://doi.org/10.1186/s40814-016-0054-2

Morgan AG, Rodd HD, Porritt JM, Baker SR, Creswell C, Newton T, Williams C, Marshman Z. (2017), 'Children's experiences of dental anxiety', *Int J Paediatr Dent*, 27(2), pp. 87-97. doi: 10.1111/ipd.12238.

Porritt, J., Rodd, H., Morgan, A., Williams, C., Gupta, E., Kirby, J., Creswell, C., Newton, T., Stevens, K., Baker, S., Prasad, S. and Marshman, Z. (2017) 'Development and testing of a cognitive behavioral therapy resource for children's dental anxiety', *JDR Clin Trans Res,* 2(1), pp. 23-37. doi: 10.1177/2380084416673798.

Porritt J, Morgan A, Rodd H, Gupta E, Gilchrist F, Baker S, Newton T, Creswell C, Williams C, Marshman Z. (2018) 'Development and evaluation of the children's experiences of dental anxiety measure', *Int J Paediatr Dent*, 28(2), pp. 140-151. doi: 10.1111/ipd.12315.

Noble F, Kettle J, Hulin J, Morgan A, Rodd H, Marshman Z. (2020) '"I Would Rather Be Having My Leg Cut off Than a Little Needle": A Supplementary Qualitative Analysis of Dentally Anxious Children's Experiences of Needle Fear', *Dent J (Basel)*, 8(2), pp. 50. doi: 10.3390/dj8020050.

Porritt JM, Morgan A, Rodd H, Gilchrist F, Baker SR, Newton T, Marshman Z. (2021) 'A Short Form of the Children's Experiences of Dental Anxiety Measure (CEDAM): Validation and Evaluation of the CEDAM-8', *Dent J (Basel),* 9(6), pp. 71. doi: 10.3390/dj9060071.

Chapter Six

6. Study 3: Development and testing of a web-based version of the Children's Experience of Dental Anxiety Measure for clinical assessment of paediatric dental patients aged 9 to 16 years. Phase 1: Web app design and development

6.1. Introduction

Following Study 2, a new measure for DFA was developed based on a CBT assessment model (Porritt et al. 2018). The CEDAM is a self-report questionnaire comprising 14 items and a three-point response scale. It was developed by a team of researchers that included consultants in dental public health, consultants in paediatric dentistry, clinical psychologists, and health psychologists. The CEDAM items were generated from the qualitative interviews with paediatric dental patients presented in the previous chapter (Morgan et al. 2017). Items were mapped onto four response systems/areas of the theoretical model (thoughts, behaviours, feelings, and physical symptoms). Situational factors, the fifth area of the model, was not included in the measure. A total score for the CEDAM is calculated by summing the scores for each item. It is then converted to a transformed interval score using a manual conversion table (scores=14-42, higher scores reflecting higher levels of DFA) (Porritt et al. 2018). To determine the appropriateness of the items, wording, and response scales, children were also involved in cognitive pretesting and piloting of the measure. The CEDAM has demonstrated a unidimensional scale on Rasch analysis, satisfactory internal consistency (Cronbach's alpha=0.88), construct validity (Known-group analysis), concurrent validity (with MCDAS total score), and test-retest reliability after two weeks (Porritt et al. 2018). Recently, an eight item, short form of the CEDAM has also been developed (CEDAM-8) (Porritt et al. 2021).

The minimum requirement to demonstrated comparability between electronic and paper-based measures is for cognitive and usability testing to be conducted with users. Cognitive testing explores how respondents perceive and interpret questions (Drennan 2003), whilst usability testing evaluates the subjective perception of the interaction with a system (users perception of ability to complete the electronic assessment) (Borsci et al. 2009). Usability broadly includes performance (e.g. navigation, learnability) and satisfaction with the product (content, interface, design) (Coons et al. 2009). Jones (2010) has suggested usability in DFA assessment considers the extent to which a measure engages a child, how simple it is for a child to complete, and how simple it is to score. Usability testing can be achieved through heuristic evaluations, qualitative approaches (e.g. 'think-aloud' technique, structured interviews) or the use of questionnaires. During a 'think aloud' exercise, participants are encouraged to verbalise their thoughts, feelings and actions whilst completing the measure under consideration (Drennan 2003; Jaspers 2009). It has been demonstrated that children identify more usability problems with this approach then when asked the same questions during a structured interview or in a questionnaire (Donker and Markopoulos 2002).

To date, there is very little guidance in the literature about the practical issues of how to migrate a paper measure onto an electronic platform. Schick-Makaroff and co-authors (2015) highlighted some of the challenges of developing an electronic measure during a pilot study to evaluate the feasibility of using tablets for electronic patient reported outcomes in outpatient renal dialysis clinics. Issues identified included: logistics (e.g. use of NHS Hospital Trust intranet, Wi-Fi availability, printing); data security (e.g. password-protection of tablet, encryption of data during wireless transfer to central database on local area network, protocols for data transfer and deletion of data from the tablet, storage of tablet when not in use); and cross-infection concerns (e.g. disposable screen covers, use of stylus). Other features considered as important in the development of electronic measures for adults have included: navigation control (e.g. back button so participants can check answers, avoidance of scrolling for next questions); incorporating a questionnaire completion tracker bar (so users can monitor progress); and to minimise the need for scrolling of the tablet screen (Morley et al. 2014). Additionally, Turner-Bowker and co-authors (2011) identified, during usability testing of a measure for adult headache sufferers, that a preference was given for an off-white background and a single item per page layout, although no preference was reported for tablet screen orientation.

6.2. Aims and objectives

The aim of this research was to design and develop a web-based version of the CEDAM for use on tablet devices with paediatric dental patients aged 9 to 16 years in a secondary/tertiary care clinical setting. A specific objective was as follows:

- To involve paediatric dental patients in the design and development of the new web-based version of the CEDAM

6.3. Ethical approval

Ethical approval for the research was granted by the NRES Committee York and Humber: Sheffield REC (16/YH/0038).

6.4. Method

The Consolidated Criteria for Reporting Qualitative Research checklist was used to inform reporting of the design and findings of this study. (Tong et al. 2007).

6.4.1. Development of the prototype web-app

Initially, a prototype version of the eCEDAM was built using Google Forms (Google Apps). The purpose was to test the acceptance of the questionnaire in a digital format. Face-to-face interviews were then completed with children on an iterative cycle basis, whereby suggested modifications were made to the prototype eCEDAM prior to further interviews (first development stage). To address the difficulties of making changes to the software coding, this was accomplished with screenshots of the iterative prototype on Microsoft PowerPoint (Microsoft Corporation, Washington) (Appendix 5). Following completion of the interviews, a Research Software Engineer from the University of Sheffield (A.T.) wrote the software for the eCEDAM web application (web-app). A web app is a small computer programme that runs in the internet browser of a mobile smart device. For this study, the web-app was developed with a standard iPad (Apple Inc.) as the user interface. Images for the web-app were provided by Five Areas Ltd (Clydebank, Scotland). This company had previously produced CBT resources for children as part of a trial involving the same research team (Porritt et al. 2016). All children in the images had neutral expressions to avoid influencing children's responses to the items. Subsequently, further face-to-face interviews were conducted with children to test the completed eCEDAM web application for system flaws (second development stage).

6.4.2. Participants

During the design and development of the prototype eCEDAM web-app, cognitive and usability testing was conducted via face-to-face interviews with children and their parents/carers. Participants comprised children aged 9 to 16 years attending an appointment in a paediatric dentistry unit within an NHS dental teaching hospital in Sheffield. The age range was selected to recruit participants consistent with the age range reported during validation of the CEDAM (Porritt et al. 2018). For the first development stage (participants 1 to 7) children were included if they had a clinical diagnosis of DFA, based on a subjective report of DFA by the dental professional responsible for their management. Children with additional communication needs were excluded (e.g. children whom interpreting services were required). Children were initially approached, based on the identification of a patient who met the inclusion criteria for the study by the responsible dental professional. The presence of DFA was then confirmed verbally by participant self-report by a researcher (A.M.). For the second development stage (participants 8 to 13) children who took part in study phase 2 were approached. Parents/carers and children were given information sheets about the study, and permission was sought for a researcher (A.M.) to make contact again by telephone in two weeks from the initial contact to enquire whether they had chosen to take part. Participants were purposively sampled to provide a range of experiences and views. Criteria used for sampling included: age; sex; ethnicity and living in varying areas of deprivation. Classification of ethnic group was based on the categories used by the ONS for measuring equality during national surveys, as previously described (see section 4.4.1) (Office of National Statistics 2016). A sampling matrix was used to monitor the recruitment of participants against key background characteristics. For the interviews, participants were given the option of it being held in their home or in a room in the School of Clinical Dentistry in Sheffield. Children were also given the option of being interviewed with their parent present or independently. Written parental consent and child assent were obtained by a single researcher (A.M.) at the time of the interview. At the end of each interview the participant was given a £10 gift voucher as gratitude for their contribution.

6.4.3. Data collection and analysis

During both interview stages the study participants were encouraged to verbalise their thoughts, emotions and actions as they completed the web-app (Jaspers 2009). A topic guide was included to support the 'think-aloud' interviews, comprising questions about the web-application design and interface guided by the available literature on the development of electronic measures in healthcare, although the participants were freely able to discuss and explore other views and ideas that developed (Appendix 4) (Turner-Bowker et al. 2011; Morley et al. 2014; Schick-Makaroff and Molzahn 2015). To determine the severity of DFA for each participant, the total score for the paper version of CEDAM was used. However, those involved in the first development stage did not always complete the CEDAM in its entirety, so to assess DFA, those participants also completed the $MCDAS_f$ (Howard and Freeman 2007). The $MCDAS_f$ comprises seven questions to assess worry about specific dental situations and procedures (e.g. having a filling) and one global question about dental anxiety generally

(Wong et al. 1998). Children rate themselves on a five-point Likert scale (1=relaxed/not worried; 5=very worried) with a corresponding faces scale. Total scores range from 8 to 40, with higher scores reflecting higher DFA levels.

The interviews were recorded using a digital sound recorder (TX650 Series, Sony Europe Limited) and then transcribed verbatim. All interviews were transcribed by Dictate2us (Manchester, U.K.), a professional transcribing service. Participant identifiable information was not included within the interviews prior to transcription (e.g. use of pseudonyms during the interview), and all identifying information was removed from the completed transcripts to maintain the anonymity of participants, dental professionals, and dental services. Thematic analysis was conducted using a hybrid of deductive (a priori based on the topic guide) and inductive approaches (Fereday and Muir-Cochrane 2006). As the interviews were completed on an iterative cycle basis, one researcher (A.M) read and reviewed each of the transcripts for the first seven participants prior to the next interview to inform modifications to the prototype eCEDAM. Therefore, data collection and analysis were conducted concurrently until data saturation occurred and no further cognitive or usability concerns emerged. However, four researchers (A.M., Z.M., J.P., and H.R.) independently read and reviewed all transcript after all the interviews were conducted. Each researcher independently identified important and repeating ideas that emerged from the data and organise the data into themes. Subsequently, each section of the transcripts was systematically reviewed, labelled, and indexed onto a Microsoft Excel Office 365 database (Washington, Microsoft Corporation). Finally, a thematic framework was developed whereby evidence to support the themes was traced to the original text from each participant (Smith and Freeman 2010). Simple descriptive analysis was conducted for the participant sociodemographic variables. To determine deprivation quintile, postcode data were converted to a deprivation rank using the GeoConvert function on the U.K. Data Service Census Support website, based on postcode data from the 2011 Census and the English Indices of Multiple Deprivation 2015, as previously described in Chapter 4 (Bryan et al. 2014; U.K. Data Service Census Support 2019).

During testing of the measure (Chapter 7) it was proposed to use the System Usability Scale (Appendix 7) as a quantitative assessment of the usability of the eCEDAM web-app. The questionnaire comprises 10 items (alternating positive and negative statements) with a five point agreement response scale (strongly agree to strongly disagree) (Bangor et al. 2009). An adjective rating scale has been added to the questionnaire to aid interpretation of the scores (worst imaginable usability to best available usability) (Bangor et al. 2009). At the time of the study, the measure has not been formally validated for use by children, although it has been previously employed in research studies involving children (Froisland et al. 2012; Ni et al. 2014). To confirm its initial validity for use in this study, children who participated in the cognitive and usability interviews were asked to complete the measure. Unfortunately, it was identified that participants had considerable difficulty understanding the alternating statements. Therefore, the measure was not used further in this study.

6.5. Results

6.5.1. Sociodemographic variables

Overall, 24 families were approached. Face-to-face, semi-structured interviews were conducted with 13 children between April 2016 and January 2017 by a single female researcher (A.M.). As previously described, the researcher A.M. is a Specialist in Paediatric Dentistry and completed a NatCen learning course on Depth Interviewing Skills prior to commencement of the study. The age of the participants ranged from 9 to 15 years. Eight participants were female. Overall, 69% (n=9) of children lived in LSOA identified as the most deprived of England (quintile 1). Three participants identified themselves as having additional needs (autism, learning disability, and dyslexia). For the first development stage, the mean MCDASf score of participants (n=6) was 22.0 (S.D.=9.1). For the second development stage, the mean CEDAM score of participants was 21.65 (S.D.=2.65). The characteristics of the participants are detailed in Table 1. All interviews, except one (participant 8) were held in the participant's own home with a parent present. Participants 1 to 7 were not previously known to the researcher (A.M.) conducting the interviews. However, participants 8 to 13 had taken part in phase 2 of the study which was also conducted by researcher A.M.

Table 10. Characteristics of children purposively sampled for cognitive and usability testing of the eCEDAM

Interview	Sex	Age (years)	Dental anxiety score	Deprivation quintile*	Self-reported disability	Ethnicity
Participants involved in the first development stage						
			$MCDAS_f$ (range=8-40)			
1	Female	10	40	1	No	White
2	Female	9	20	1	No	Other ethnic group**
3	Female	9	16	4	No	White
4	Male	11	16	1	Autism	White
5	Male	15	19	1	Dyslexia	White
6	Female	13	Unable to complete	2	Learning difficulties	White
7	Male	13	21	1	No	White
Participants involved in second development stage						
			CEDAM converted score (range=14-42)			
8	Female	15	20.12	1	No	White
9	Female	9	23.4	1	No	Asian/Asian British
10	Male	12	20.12	1	No	White
11	Female	9	25.78	1	No	White
12	Female	9	18.49	4	No	White
13	Male	10	21.99	2	No	Other/Arabic

*Quintile 1=most deprived; Quintile 5=least deprived

**Ethnic group recorded as 'other ethnic group not listed'

6.5.2. Themes and subthemes

During the think-aloud cognitive and usability exercise, participants made suggestions to improve the eCEDAM user experience. The refinements made to the prototype eCEDAM are presented in Table 11. There were no issues identified with how items were interpreted by participants. Additionally, participants were also able to discuss and explore other views and ideas that developed. From these discussions two themes were identified: use of DFA assessment measures with children; and use of tablet computers for DFA assessment.

Table 11. Refinements made to eCEDAM during web-app development

- Audio-recording for each item
- Background and font colour based on recommendations of British Dyslexia Association
- Cartoon images of dental situations
- Touch-screen button colour change when response to item selected
- Images, information, and welcome message from responsible dental professional
- Inclusion of a logo
- Increase in size of text and touch-screen buttons
- Incorporation of a progression bar
- Personalisation of first screen
- Questionnaire completed in portrait or landscape layout
- Single item per screen (versus scrolling)
- Tools to help children from inadvertently missing an item

6.5.2.1. Use of DFA assessment measures with children

Communication with the dental team

Generally, DFA questionnaires were identified as a useful tool for communication with the dental team, particularly if children found it challenging to express their emotional experience verbally. It was suggested that their use would result in the dental team acting differently and helping children with their DFA.

'Yeah, because if you've got any concerns about what they're doing you could tell them about it before they actually go in.' (Participant 10)

'Yeah, because then they'll talk it through you and then they'll make you, like, calmer and then they won't, like, make you really worried as you were before.' (Participant 4)

Contribution to children's DFA experience

Participants described a range of thoughts and feelings associated with completing DFA questionnaires. Children made catastrophic interferences that the dental team would make negative judgements about them based on their answers to the items, or that they would choose the 'wrong' answer, and the dental team would not know they were experiencing DFA.

'Because it will say I don't like dentist, they might tell I'm just being silly.' (Participant 11)

'Like saying if you don't want to go to the dentist that could be embarrassing.' (Participant 10)

'*If you press the wrong answer then you press next, obviously the dentist will think you're okay for going.'* (Participant 1)

Participants also described embarrassment about being singled out to complete a DFA questionnaire in the dental surgery waiting room.

'Yeah. Because it's less noticeable [using a mobile phone] and they're not sure what you're doing. You could be texting anyone. Whereas on a tablet, they could probably see it from further away. They could see that like, and the dental questions.' (Participant 7)

'Depending on how old they are. I think like in between 14, not 14, maybe 16 and 18. I think they would probably laugh at like people who are 11 and 12 doing it.' (Participant 12)

However, children also described how completing a DFA questionnaire could have positive impacts on their DFA experience by facilitating helpful cognitive coping responses.

'Take your mind off what is going to happen as well.' (Participant 7)

'[Help children to know] It's okay to be scared.' (Participant 11)

Assessment anxiety

The provision of a score meant DFA questionnaires were perceived unfavourably as a type of test for children. As described previously, children were concerned about choosing the 'correct' response.

'No, that's not okay because it really feels like it was a test, like in results papers it tells you what score you got to them.' (Participant 13)

'And if you did do it, instead of like say your score is, because people might be like, "I thought this wasn't a test?' (Participant 12)

'If it's saying you're not worried then that might, like that might make you feel more better.' (Participant 9)

Respecting children's autonomy

Participants discussed the importance of respecting children's right to choose whether they want to complete a DFA questionnaire. It was felt that it should not be a requirement for dental visits. Additionally, if children chose to complete it, their right to confidentiality should be maintained. For example, only their dentist should have access to their responses.

'Yeah, but you can't force, like, children to do it. If they don't want to do it, they don't have to do it. (Participant 3)

'I'd want to keep it private.' (Participant 11)

6.5.2.2. Use of tablet computers for DFA questionnaires

Participants discussed confidence in completing the CEDAM on a tablet computer and felt other children would feel similarly.

'I would say most children would know what to do.' (Participant 4)

'On iPad because kids, they mostly go on iPads, and they don't really read books. Well I don't, so I go on that that's why.' (Participant 9)

Additionally, interactive features on tablet computers, such as the use of 'Read Aloud' functions to provide audio for the written text, were identified as being useful.

'I'd like to listen to a few and read a few because you might get bored of reading.' (Participant 12)

'Like writing next to it like 'Click this button to hear' it will speak.' (Participant 13).

Source of information for children with DFA

Children also suggested the web-app could be a possible source of information to decrease the unpredictability associated with dental care. Suggestions were given for images of the dental team and dental treatments be included, and advice provided on what actions children should take in certain dental situations.

'Because, like, if they see photos of what happens at the dentist, they could just, instead of, like, doing the questions they could actually have a think about the pictures as well and see if they still feel really upset about going to the dentist or if they're still going to be still happy going to the dentist.' (Participant 1)

'Yeah, because if they don't really understand, they can look at it and then it might show them what it's about.' (Participant 9)

'And, like say someone said stopped the dentist, I don't know if this was one of the questions, but maybe a question saying, if you were having teeth pulled out whilst being awake, what would you say to the dentist if you wanted him to stop?' (Participant 12)

However, some children described that more information might increase unhelpful behavioural responses (e.g. avoidance).

'If I didn't like the picture of the dentist, I'd say I'm not going to that dentist at all.' (Participant 11)

How to increase the children's interest in the eCEDAM

Children also reported that completing the eCEDAM web-app was not as entertaining as anticipated. However, differing views were given on whether this was important. Some children felt that it should be more like a game, identifying that such a change could incentivise other children to want to complete it.

‘Because it is a quiz and then it tells you how you feel and that’s not even need to be fun.’ (Participant 13)

‘Because it’s just like boring. Yeah. So, if you put a game, they might like they might want to do it again for the game.’ (Participant 9)

6.6. Discussion

This study included children with DFA directly in the design and development of a prototype eCEDAM web-app for DFA assessment. In recent years there has been a significant increase in commercially available mobile health (mHealth) apps for children (Grist et al. 2017). The focus of mHealth apps vary, but many include self-report questionnaires (Grist et al. 2017). To date, scientific literature to document changes made to original survey questionnaires during their adaption for use on mobile technology is lacking (Marcano Belisario et al. 2015).

The aim of the study was to develop an electronic version of the CEDAM for the purpose of DFA assessment only. Contrastingly, children perceived that the web-app should also be an information resource for children with DFA. Generally, provision of information can reduce the unpredictability of a dental situation for children (Buchanan 2017). However, studies on the provision of procedural dental information using written leaflets, verbal explanations, or video recordings have not demonstrated a reduction in state DFA in children (Olumide et al. 2009; Wright et al. 2010; Al-Namankany et al. 2014). A possible explanation is that there are individual differences in the extent of information children prefer about potentially DFA-prompting dental situations or procedures (Buchanan 2017). To explain these differences Miller (1987) has suggested the concepts of monitoring and blunting coping styles. This theory describe whether individuals respond to stressful situations by attending to information (‘Monitors’) about potential stressors, or by avoiding information (‘Blunters’), and preferring to have their attention focused elsewhere (Buchanan 2017). Individuals with a high monitoring coping style seek out information to reduce unpredictability about health care, but are also less satisfied with the information provided by health professionals than those with a low monitoring/blunting preference (Miller 2015). However, if health professionals are able to tailor information provision to matches an individual's informational coping style, it can lead to reduced patient anxiety (Miller 1995). With relevance to paediatric dentistry, there is some evidence to support dental information-specific monitoring and blunting coping styles in children, although there is little research into the informational coping styles of children with DFA (Williams and Jones 2012). Following participant feedback, the prototype eCEDAM included a final page with a photograph and welcome message from the dental professional who would be responsible for their dental care during their visit, and who would be discussing their questionnaire responses (Appendix 6). However, further research is needed to determine informational coping styles of children with DFA, and how it relates to children's DFA experiences.

Within the U.K., dental professionals do not routinely use DFA assessment questionnaires in clinical practice (Alshammasi et al. 2018). A possible barrier is a concern that questionnaire items and terminology (e.g. items about local anaesthetic) may precipitate a DFA response in children (Alshammasi et al. 2018). That is by assessing DFA, dental professionals will cause DFA (Alshammasi et al. 2018). To date, the available evidence has suggested that DFA assessment reduces state DFA in patients (Dailey et al. 2002). Although, participants were generally supportive of the use of DFA questionnaires, the findings from this study would suggest that DFA assessment may contribute, for some children, to DFA, with children having unhelpful thoughts and feelings about completing the measure (Williams and Garland 2002). Corresponding to characteristic changes in thinking associated with anxiety, children made catastrophic inferences: that they could make a mistake with their responses and there would be detrimental consequences; or that dental professionals, upon receiving their responses, would make negative judgements about them (Williams and Garland 2002). Participants also described feeling singled out and embarrassed about completing a DFA when they could be observed by others. In adults with DFA, embarrassment about poor oral health has been suggested to contribute to avoidance of dental care (Berggren and Meynert 1984). Contrastingly, study participants were more concerned about stigma associated with DFA and social ridicule from peers, than their oral condition. Previous research has identified children similarly were embarrassed for their peers to know they used mental health apps (Kenny et al. 2016). Interestingly, tablet computers and the ability to send responses electronically to the dental team provide the possibility for children to complete DFA assessment measures at home prior to dental visits. However, stigma about DFA is a study limitation. Although potential participants were taken to a clinical area to discuss the study, they were initially approached in a waiting area where they could have been observed by others. It is possible that this could have influenced children's decision-making on whether to participate in the research. However, as part of the study ethical requirements, children who declined to participate were not asked to provide an explanation. Therefore, it is not known if embarrassment about being identified for the study did influence participant recruitment. Further research is needed into children's DFA related social evaluative fears and anxieties, and how social stigma associated with DFA can be addressed on an individual and population level. Additional consideration is also required on how children are approached to participate in research.

An interesting idea discussed by participants was the gamification of the eCEDAM web-app. The aim of gamification is to use game-like features (e.g. competition elements) to promote engagement with a routine and repetitive task (Lumsden et al. 2016). In a previous study, a game used to provide information to children and families before a dental general anaesthetic was found not to influence preoperative anxiety (Huntington et al. 2018). Conversely, there is evidence of successful employment within cognitive testing (Lumsden et al. 2016). A challenge with DFA assessment was that children did perceive it as boring. Therefore, there is scope to incorporate electronic DFA assessment within game play for the purposes of patient engagement, rather than an attempt to reduce anxiety. As previously described, children have individual differences in informational coping (Buchanan 2017). However, when a situation is uncontrollable, information is unlikely to reduce DFA, and blunting/distraction may be a more helpful coping response (Miller 1980). Corresponding, a game may have additional advantages as a distraction for children with DFA prior to their appointment. Moreover, gamification has been suggested as an approach to reduce assessment anxiety (Lumsden et al. 2016). That said, the implications for gamification on the psychometric properties of DFA assessment measures is unknown. It would also be challenging to develop a game that children would find engaging without involvement of commercial partners.

6.7. Conclusions

1. Children expressed confidence in the use of tablet computers to complete DFA assessment measures.
2. Suggestions were made to the design of the eCEDAM that would support children with reading difficulties and promote children's autonomy over completion of DFA assessment measures.
3. DFA assessment was associated with social evaluative fears and anxieties and contributed to the DFA experience of some children.
4. Gamification of the eCEDAM has potential to promote patient engagement, reduce assessment anxiety, and provide a distraction for DFA. A potential barrier is the identification of an appropriate game.

6.8. Publications arising from the chapter

Morgan AG, Bower C, Wilson V, Towers A, Stokes CW, Porritt JM, Rodd HD, Marshman Z. (2016) 'Usability testing of a web application to assess childhood dental anxiety' [Poster], *Int J Paediatr Dent*; 26 (Suppl. 1): p11.

Chapter Seven

7. Study 3: Development and testing of a web-based version of the Children's Experience of Dental Anxiety Measure for clinical assessment of paediatric dental patients aged 9 to 16 years. Phase 2: Evaluation of measurement agreement of the CEDAM and eCEDAM

7.1. Introduction

The available evidence would support data equivalence between questionnaires completed by paper or electronic administration (Gwaltney et al. 2008; Marcano Belisario et al. 2015). Given that the CEDAM and eCEDAM present identical text for the introduction, items, and responses, it is likely that the scores generated by the two questionnaires would not differ. However, a recent systematic review supporting data equivalence between apps and paper questionnaires included only one study involving children and did not separate studies with adult and child participants during the analysis (Marcano Belisario et al. 2015). Therefore, it is not known whether children would respond differently to the eCEDAM compared to the CEDAM. That is, in some way measurement error is introduced (Marcano Belisario et al. 2015). For example, a single questionnaire item on the eCEDAM is presented on each page, whereas multiple questionnaire items are included on a single page of paper for the CEDAM, or if the functionality of the tablet computer (e.g. use of audio-recordings) influenced responses (Gwaltney et al. 2008).

7.2. Aims and objectives

The aim of this research was to test a web-based version of the CEDAM for use on a tablet device with paediatric dental patients aged 9 to 16 years in a secondary/tertiary care clinical setting. Specific objectives were as follows:

- To determine measurement agreement between the new web-based version (eCEDAM) and original paper version (CEDAM)
- To determine if, compared to the paper version, the web-based version had an influence on data quality (time to complete, proportion of missing items, and proportion of completed questionnaires)
- To assess acceptability and participant preference for the mode of delivery

7.3. Ethical approval

Ethical approval for the research was granted by the NRES Committee York and Humber: Sheffield REC (16/YH/0038).

7.4. Method

To determine agreement between the CEDAM and eCEDAM a cross-over study design was employed, whereby 100 consecutive children were given both versions to complete in a randomly assigned order. The wording of the questionnaires was identical. The primary research question was to evaluate agreement between CEDAM and eCEDAM total scores. This was achieved using a Bland and Altman plot (Bland and Altman 1986). Correspondingly, a sample size of 100 was determined (Bland 2014).

7.4.1. Participants

Participants comprised children aged 9 to 16 years with DFA attending an assessment or follow-up appointment in paediatric dentistry unit within an NHS dental teaching hospital in Sheffield. As previously, the age range was selected to recruit participants consistent with the validation study for the CEDAM (Porritt et al. 2018). Children were included if they had a clinical diagnosis of DFA and were able to complete both versions of the questionnaire. Children with additional communication needs were excluded (e.g. children whom interpreting services were required). Children were initially approached, based on a subjective report of DFA by the dental professional responsible for their management during their appointment. The presence of DFA was then confirmed verbally by participant self-report by a researcher (A.M.). Parents/carers and children were given information sheets about the study, and parental consent and child assent obtained by a single researcher (A.M.). Participants then completed the study during the same dental attendance.

7.4.2. Data collection

Participants completed both paper and electronic questionnaires. Allocation to the paper or electronic group for the first measure to be completed was determined by block randomisation. Participants opened consecutively numbered sealed opaque envelopes containing their allocation following recruitment. Between completing their first and second questionnaires, participants had a timed 15 minute resting period, with the options of having a drink and contributing to an art project during the break.

Participants completed the eCEDAM on the internet browser of a standard iPad (Apple Inc.). To meet local standards for the prevention and control of cross-infection an iPad cover designed for healthcare environments (Tough-PAC, Innervision Technology Ltd) was used and cleaned daily (Charles Clifford Dental Hospital 2015). A researcher (A.M.) was available to give support with any difficulties that arose (e.g. internet connectivity issues), to record the time taken to complete both versions of the questionnaire and the resting period, and to provide the participants with refreshments and art materials. Time was measured using a digital stopwatch (HS-80TW, Casio Electronics Company Limited). For the duration of the study, the application website was hosted on a secure University of Sheffield server and accessed over an encrypted connection. Participant responses were sent from the tablet to the website using the secure University of Sheffield Wi-Fi network. A file transfer daily protocol was implemented which included back-up of data from the website. No patient data was stored on the iPad. When not in use the tablet was stored in a locked office within the University of Sheffield. Participant demographic data (age, sex, postcode, and ethnicity) were recorded. Classification of ethnic group was based on the categories used by the ONS for measuring equality during national surveys, as previously described (Office of National Statistics 2016). Participants also completed a questionnaire on the acceptability of both CEDAM versions (based on the five-item response format used for the paediatric version of the NHS Friends and Family Test), and their preference for future CEDAM use (CEDAM, eCEDAM, no preference) (Picker Institute Europe 2015). Each participant was given a £10 gift voucher as gratitude for their contribution following completion of both measures and the questionnaire booklet.

7.4.3. Data analysis

Data from both measures was transferred manually to a Microsoft Excel Office 365 database (Washington, Microsoft Corporation). Raw scores for the CEDAM/eCEDAM were converted to an interval scale (Porritt et al. 2018). The data were subsequently analysed using IBM SPSS Statistics for Windows version 23 (New York, IBM Corporation). Simple descriptive analysis was conducted for the sociodemographic variables, CEDAM/eCEDAM total scores, data completeness, time to complete and acceptability. Postcode data were converted to a deprivation quintile using the GeoConvert function on the U.K. Data Service Census Support website, as previously described in Section 4.4.3. (Bryan et al. 2014). Internal consistency is a measure of reliability and determines the extent items in a questionnaire are correlated (e.g. measures the same construct) (Terwee et al. 2007). Internal consistency was calculated using Cronbach's alpha. Terwee and co-authors (2007) have proposed within their quality criteria for health status questionnaires, that a minimum standard for internal consistency, is when Cronbach's alpha is between 0.70 and 0.95. Concurrent validity was assessed by determining the correlation coefficient between CEDAM ('gold standard') and eCEDAM. A minimum standard for concurrent validity is a correlation of at least 0.70 with the 'gold standard' (Terwee et al. 2007). A scatter plot was performed using GraphPad Prism version 7 for Windows (California, GraphPad Software). In contrast to correlation which measures association, agreement determines how much the eCEDAM score is likely to differ from the CEDAM score, and whether this difference is clinically important e.g. whether the eCEDAM can replace the CEDAM (Bland and Altman 1986). To assess agreement the Bland-Altman method was used to plot the difference in score between the CEDAM and eCEDAM (CEDAM score minus eCEDAM) score against the means score for the two questionnaires (Bland and Altman 1986). From the mean difference, the limits of agreement are calculated, whereby 95% of the differences are between these limits. Bland and Altman (1986) recommended that the difference between the levels of agreement that is clinically relevant is based on clinical judgement. However, Gwaltney (2008) recommended that the difference should not exceed the minimal clinically important difference for a measure. The minimal clinically important change in score for the CEDAM is 3.9 points (Porritt et al. 2021). Therefore, a difference between levels of agreement less than 3.9 points is not clinically relevant, and the eCEDAM could be recommended as an alternative to the CEDAM. A Bland-Altman plot was performed using GraphPad Prism version 7 for Windows (California, GraphPad Software).

7.5. Results

7.5.1. Sociodemographic variables

Overall, 108 children were invited to participate in the study, with 100 children recruited between July 2016 and January 2017. The recruitment rate was 92%. Seven children were unable to complete the eCEDAM following Wi-Fi connectivity difficulties and were excluded from the analysis. The age of the participants ranged from 9 to 16 years (median=11.0 years, I.Q.R.=3.0). There were more female than male participants (60% and 40%, respectively). Overall, 40% (n=36) of children lived in LSOA identified as the most deprived of England (quintile 1). Most (71%, n=66) of participants identified their ethnicity as 'White'. Detailed participant demographic data are presented in Table 12.

Table 12: Data for participants who completed the agreement study (phase 2)

Variable	Category	Median/Frequency
Age (n=93)	Mean	11.1 years (S.D.=2.1)
	Median*	11.0 years* (I.Q.R.=3.0 years)
	Range	Minimum=9 years Maximum=16 years
Sex (n=93)		
	Male	39.8% (n=37)
	Female	60.2% (n=56)
Deprivation Quintile (n=89)		
	1 (Most deprived)	40.4% (n=36)
	2	21.3% (n=19)
	3	19.1% (n=17)
	4	6.7% (n=6)
	5 (Least deprived)	12.4% (n=11)
Ethnicity (n=93)		
	White	71.0% (n=66)
	Mixed/multiple ethnic groups	7.5% (n=7)
	Asian/Asian British	10.8% (n=10)
	Black/African/Caribbean/Black British	8.6% (n=8)
	Other ethnic groups	2.2% (n=2)

*Age did not follow a normal distribution

7.5.2. Reliability

Internal consistency, calculated using Cronbach's alpha, was found to be satisfactory for both the CEDAM (α=0.91) and eCEDAM (α=0.92).

7.5.3. DFA scores

The 14 items in the CEDAM/eCEDAM were scored from one to three. Therefore, the minimum total score was 14 and the maximum total score 42. The raw total scores are then converted to an interval scale (Porritt et al. 2018). The total scores for both the CEDAM (skewness=1.9, S.E=0.3; kurtosis=7.1, S.E=0.50) and eCEDAM (skewness=2.3, S.E=0.25; kurtosis=8.8, S.E=0.50) were not normally distributed (Kim 2013). The median score for both the CEDAM and eCEDAM was 20.1 (interquartile range=5.1 and 4.8, respectively). There were no significant differences between the total CEDAM and eCEDAM scores (Wilcoxon Signed Rank Test, z=-1.6, p>0.05).

7.5.4. Correlation

The correlation coefficient between the CEDAM and eCEDAM was found to be satisfactory (Spearman's Rank Correlation Coefficient, r_s=0.94, p≤0.05). A scatterplot for correlation of CEDAM/eCEDAM total score is presented in Figure 8.

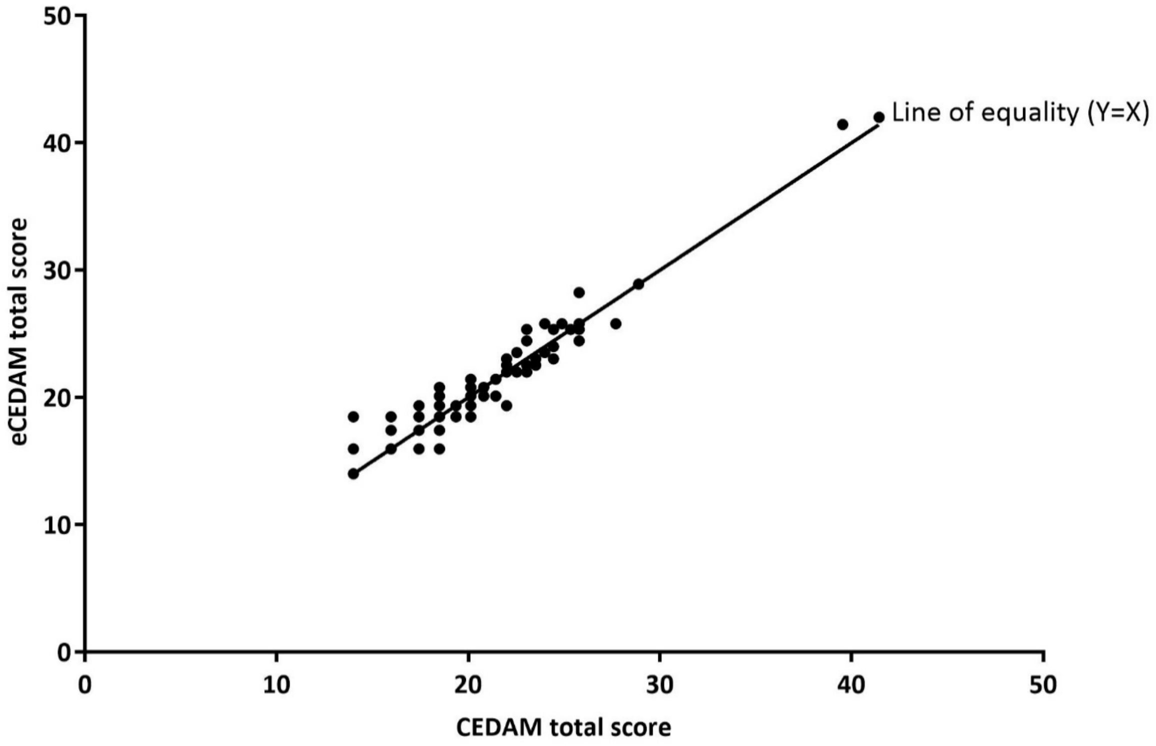

Figure 8. Scatterplot to show correlation between CEDAM and eCEDAM total score

7.5.5. Agreement between CEDAM and eCEDAM

The Bland Altman plot is presented in Figure 9. The difference between the total scores for the CEDAM and eCEDAM did approximate to the normal distribution (skewness=0.4, S.E=0.25; kurtosis=0.7, S.E=0.50) (Kim 2013). The mean difference between total scores was 0.20 (S.D.=1.23). The 95% confidence interval for the mean difference was -1.2 to 0.59. The limits of agreement are -2.2 (95% confidence interval=-2.6 to -1.8) to 2.6 (95% confidence interval=2.2 to 3.0). Therefore, the eCEDAM score may be 2.2 below or 2.6 above the score obtained for CEDAM. Based on the minimal important different score of 3.8 for the CEDAM, the agreement between the CEDAM and eCEDAM was satisfactory (Porritt et al. 2021).

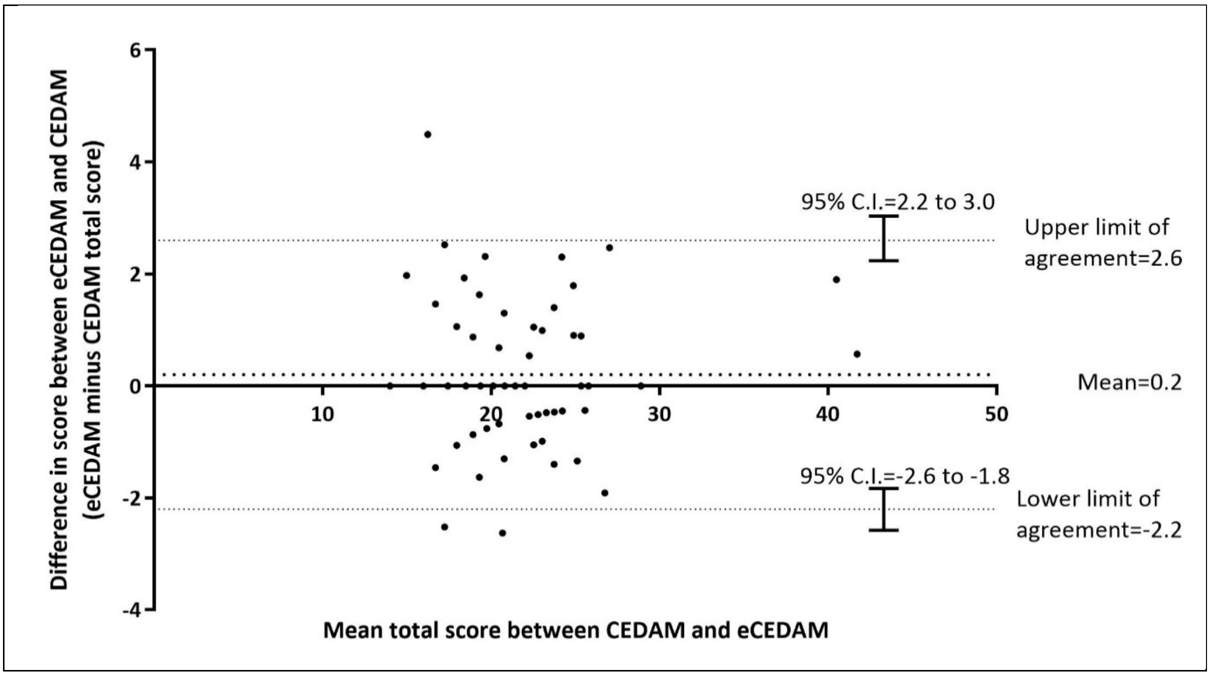

Figure 9. Bland-Altman Plot

To show the difference between scores (eCEDAM total score minus CEDAM total score) against the mean of the scores for CEDAM and eCEDAM.

7.5.6. Data completeness, time to complete and acceptability

For both questionnaires (CEDAM and eCEDAM) there were no missing items. The time taken to complete both the CEDAM (skewness=1.12, S.E.=0.27; kurtosis=1.89, S.E.=0.53) and eCEDAM (skewness=1.14, S.E.=0.26; kurtosis=3.60, S.E.=0.51) were not normally distributed. The median time for the CEDAM was 144 seconds (n=82, I.Q.R.=100 seconds), and for the eCEDAM was 159 seconds (n=86, I.Q.R.=77 seconds). There were no significant differences between the time taken to complete CEDAM and eCEDAM (Wilcoxon Signed Rank Test, z=-7.53, p>0.05). However, there was a significant difference between the time taken to complete the first and second questionnaires (CEDAM or eCEDAM), with the second questionnaire completed more quickly (Wilcoxon Signed Rank Test, z=-4.67, p≤0.05). Acceptability was determined for both the CEDAM and eCEDAM (Table 13). Most participants found both the CEDAM and eCEDAM acceptable (82%; N=75 and 90%; n=83, respectively). Overall, nearly half of participants (57%, n=52) did not express a preference for one mode of delivery (e.g. paper or tablet computer). For those that did select a preference, 58% (n=23) preferred the tablet computer. There was no significant association between preference and sex (Chi-squared Test, z=2.92, p>0.05).

Table 13. Acceptability of the CEDAM and pCEDAM (based on the five-item response format used for the paediatric version of the NHS Friends and Family Test)

I think the paper questionnaire where I write my answers is good for children and young people		
I think that the questionnaire on a tablet computer is good for children and young people		
	CEDAM (n=92)	**eCEDAM (n=92)**
I agree a lot	52.2% (n=48)	66.3% (n=61)
I agree a bit	29.3 %(n=27)	23.9% (n=22)
I disagree a bit	10.9% (n=10)	4.3% (n=4)
I disagree a lot	1.1% (n=1)	2.2% (n=2)
I don't know	6.5% (n=6)	3.3% (n=3)

7.6. Discussion

An important key finding was that agreement between total scores on the CEDAM and eCEDAM were satisfactory. That is, the scores derived from the eCEDAM were equivalent to the scores derived from the CEDAM. This provides evidence that the psychometric properties of the measure were not changed when it was adapted for use as a web-app on a tablet computer (Coons et al. 2009). To test agreement a crossover study design was employed, whereby consecutive children were given both versions to complete in a randomly assigned order. A potential study limitation was the relatively short duration of the washout period between administration of the paper and web-app versions, and the possibility of carry-over effect where participant's memory of their previous responses contributed to data agreement (Marcano Belisario et al. 2015). The use of a crossover design has been recommended for assessing data equivalence between different delivery modes for the same questionnaire (Gwaltney et al. 2008; Marcano Belisario et al. 2015). This approach also addressed participant allocation difficulties identified in a previous study, whereby children who were not allocated to the computerised intervention group for a DFA communication tool declined to be allocated into the control group (Jones 2014). To date, there is little available evidence to inform the duration of the washout period for DFA assessment in children. A short duration was chosen, whereby children completed the study during the same dental attendance, as it was possible children's DFA severity/score could have changed between attendances (e.g. following dental treatment). Moreover, activities were organised for participants (e.g. colouring, having a drink) during the washout interval, although the effectiveness of using this approach on the reduction of recall bias is not known (Salaffi et al. 2009). As possible evidence of a carryover effect, participants did complete their second questionnaire faster than they had completed the first questionnaire. However, as previous described, children suggested the CEDAM/eCEDAM was not as entertaining as expected, and it also possible that for their second questionnaire children simply completed it faster to reach the end of the study.

Most children with DFA rated the acceptability of the CEDAM and eCEDAM highly and agreed with the statement that they are good for children and young people. As previous described, dental professionals do not support the use of DFA assessment questionnaires in clinical practice partly due to the perception they may precipitate DFA in children (Alshammasi et al. 2018). The overall findings from Study 3 do suggest that some children experience anxiety specifically related to DFA assessment questionnaires, although most children would support their use. It should also be acknowledged that the CEDAM/eCEDAM are based on children's DFA responses, whereas DFA questionnaires have typically included a list of dental situations and procedures that may provoke DFA. Therefore, it is not possible to generalise the high acceptability of the CEDAM/eCEDAM to other measures. Most children also did not report a preference for paper or mobile smart device for mode of delivery. This is surprising, as a previous study to develop a computerised DFA assessment tool found that children overwhelming preferred using electronic questionnaires compared to paper questionnaires. The findings presented in Chapter 6 may provide an explanation, whereby children suggested the eCEDAM web-app was boring. As most children found both the CEDAM/eCEDAM acceptable, and did not report a preference, it would suggest both could be utilised during DFA assessment, offering dental professionals increased choice.

A further barrier to the use of standardised DFA assessment questionnaires reported by dental professionals is the perception that they are time-consuming to complete. During this study, children on average took between two and three minutes to complete the CEDAM/eCEDAM, and overall 95% of questionnaires were completed within five minutes. A possible consideration is the presence of a researcher to assist children whilst completed the questionnaires. It is possible that in clinical practice that high level of support would not be available. During the study a stopwatch was used to record the time taken to complete each measure, to standardise the data collection for the paper and web-app versions. However, further analysis of the log data for the tablet computer may have provided information about children's cognitive processes during DFA assessment (Marcano Belisario et al. 2015). For example, how long did children spend on each item; or did they change their responses, and how many times? It may also have yielded details about how design of the web-app influenced children's completion of the questionnaire e.g. did they utilise the audio-recording feature.

7.7. Conclusions

1. Completion of the CEDAM as a web-app on a tablet computer does not affect data equivalence compared to the original paper version
2. There were no differences in time taken to complete the measure, or data completeness of responses between CEDAM and eCEDAM
3. Most children completed the CEDAM/eCEDAM within 5 minutes

4. High acceptability was reported by children with DFA for the CEDAM and eCEDAM
5. Children did not have a preference for completion of the questionnaire on paper or on a tablet computer

Chapter 8

8. Discussion

Assessment of DFA in children is of paramount importance in, not only, the development of individual care plans, but also the development of services for this group. This thesis has established through the first two studies presented in Chapters 4 and 5, that both qualitative and quantitative differences exist between adults and children in all dimensions of the theoretical model of DFA outlined in Chapter 2. Having understood these differences, a trait measurement of dental anxiety in children was developed (the CEDAM), and an electronic version tested.”

Chapters 4 to 7 have each included a discussion section pertinent to the specific study described in that chapter. This further chapter therefore provides an overarching discussion which considers the body of work as a whole.

8.1. Summary of research aims and objectives

During the literature review in Chapter 2, the potential of DFA assessment using standardised questionnaires to improve patient care in clinical practice was described. However, conceptual limitations and barriers to clinical utilisation of existing measures for DFA assessment in children were identified. The aim of the research described in this thesis was to further the understanding of DFA assessment in paediatric dental patients. The research was conducted as three separate studies. In Study 1, the sociodemographic, quality of life, and child mental health characteristics of children referred for DFA management to a secondary/tertiary NHS clinical setting were described; whilst in Study 2, a qualitative approach was employed to explore children’s DFA experiences using a CBT model as a theoretical framework for DFA. The findings of these two studies were used to inform the development of a new child centred DFA assessment measure (Porritt et al. 2018; Porritt et al. 2021). In Study 3, a web app version of the CEDAM was designed and developed with children for use on a mobile smart device. A mixed-method approach was used that included cognitive, usability, and equivalence testing to compare data agreement between the web-app and paper versions of the measure.

8.2. Key research findings and study limitations

8.2.1. Study 1: Sociodemographic, quality of life, and child mental health characteristics of paediatric dental patients with DFA

The first objective of this cross-sectional study was to determine a profile of children who would potentially be users of a new DFA assessment questionnaire in clinical practice. The key findings were that children referred to a secondary/tertiary NHS dental teaching hospital with DFA had a female preponderance, were most likely to be of White ethnicity, which corresponded to ethnicity data for the local population, and lived in areas identified with high levels of deprivation. As described in Section 4.6, the study population of children referred for DFA management may not be representative of children with DFA generally. An additional consideration is whether the socio-economic background of participants was influenced by dental caries status, as it is unlikely children with DFA would be referred to secondary/tertiary care without needing dental treatment. However, caries status was not recorded, and it is not possible to identify whether participants had a high caries experience. It should also be acknowledged, that nearly a quarter of children invited to participate in the study were not brought to their first appointment and were excluded from the study. As generally, 10% of patients referred to the Paediatric Dentistry service in Sheffield are not brought to their first appointments, this suggests that the frequency of missed appointments was higher in potential study participants than children referred for paediatric dentistry treatment overall (Marsden 2020). It is possible that children who are not brought to their first appointment had different DFA characteristics than children who were brought to their appointment. For example, did DFA contribute to their failed attendance? Although beyond the scope of this study, it is possible that some of the children would have been re-referred and an opportunity provided to assess their DFA and follow-up their outcomes.

Children were included in the study if they had been referred for specialist DFA management, so a reasonable assumption was that the patient group would have high levels of DFA. However, an unexpected finding in Study 1 was that when DFA was assessed using the MCDAS as a standardised measure, this was not what was identified, and children in the study population had a range of DFA severities. In Section 4.6., the conceptual limitations of the MCDAS as a measure were discussed as an explanation. However, the findings were similar in Study 3, when the CEDAM was used as the DFA assessment questionnaire. Notably, the CEDAM was developed to improve the theoretical foundations for DFA assessment in children. Additionally, in Section 4.6., the possibility that children without DFA were being referred was considered. However, to address this concern, potential participants for Study 3 also confirmed verbally during study recruitment that they self-identified with DFA. There are several possible explanations for this inconsistency between patient and clinical reporting of DFA and standardised DFA assessment, identified during the studies. Firstly, In Study 2 and Study 3 (Sections 5.5.4.4 and 6.5.2.1), children with DFA described being embarrassed about having DFA and were concerned about how it was perceived by others. They also described fears and anxieties about their questionnaire responses. A consideration is whether an inclination towards social desirability influenced children's self-reported DFA assessments. That is, participants themselves may have provided an inaccurate DFA assessments to play-down their perceived flaws, or to please their parents or their researcher (Dadds et al. 1998; Silverman and Ollendick 2005; De Leeuw 2011). A potential study limitation was that some participants completed their questionnaires in a waiting area before their appointments, and the lack of privacy may have influenced their responses (De Leeuw 2011). Social desirability bias has potential clinical relevance, as if children have been identified by others as experiencing anxiety, but deny their anxiety themselves, it can indicate an avoidant emotional processing style and result in poorer treatment outcomes (Dadds et al. 1998). However, further research is needed to understand the relationship between DFA and social desirability in children (Dadds et al. 1998). Moreover, as children rely of adults for guidance, further research is also needed into what role parents and dental professionals play in children's stigma-related perceptions of DFA. The importance of state and trait concepts in DFA assessments, and whether children's DFA 'state' influenced how they reported their DFA on the trait measures, also requires consideration. For example, children who took part in Study 3 frequently completed the CEDAM at the end of their dental visit, whereby they were no-longer likely to be experiencing high levels of state DFA. It is possible they self-reported their trait DFA accordingly. That is, the responses that were provided were for their here-and-now. There is some anecdotal evidence for this from the study, whereby participants discussed that their responses would be different if they knew they were to have dental treatment. There is some evidence from the literature that trait anxiety is not associated with DFA, but children who report high state anxiety are more likely to have DFA (Chellappah et al. 1990). However, in Study 1 children completed the MCDAS at home or in the waiting room before their appointment when higher levels of state DFA could be expected. Another potential explanation is that some participants were experiencing DFA only towards a specific dental situation or stimulus. That is, their DFA is contextual, whereby their levels of trait DFA were low generally, which was what was determined by the MCDAS and CEDAM as trait measures, but they had been referred because a specific treatment need exceeded their perceived ability to cope. Correspondingly, an individual may have situation specific DFA, rather than general DFA. Consequently, four DFA diagnostic categories have been suggested for adults. These are: a specific stimulus DFA; generalised DFA, distrust of dental personnel DFA: and fear of having a medical catastrophe (e.g. cardiac arrest) (Weinstein et al. 1987; Locker et al. 1999b). To date, identifying diagnostic features of different DFA typologies in children has received little research interest. However, delineation between general DFA, situation specific DFA, and distrust of dental personnel DFA, may have clinical relevance, as potentially the latter two typologies may be more amenable to DFA treatment, as once their treatment needs have been addressed, they may return to their normal low DFA levels (Armfield and Heaton

2013). This DFA scenario has previously been discussed in the literature regarding DFA assessment, that an individual may have a low score overall, but a high score for a particular measure item (e.g. use of local anaesthesia) (Buchanan 2017). However, dental care is complex, and there are many different potential stressors (e.g. clinical, interpersonal). Therefore, it would be challenging to develop a measure that can account for these complexities and generate a score that places an equal importance on overall score for all items, and a high score on a single item. Further research is needed involving individuals who have DFA towards a specific dental stressor, rather than general DFA, and how this influences children's self-reporting of DFA on assessment questionnaires.

The key findings for the second and third objectives, were that most children with DFA did not have additional psychological difficulties and reported levels of impacts on daily living consistent with those reported in community samples. Due to the response burden of completing multiple measures, questionnaires for child mental health and HRQoL are not recommended for DFA assessment in clinical practice of all children. For the aims and objectives of the study, a cross-sectional design and convenience sample of participants was appropriate. However, as discussed in Section 4.6, the lack of a control group of children not referred with DFA is a significant study limitation. Having a control group of patients would have facilitated further statistical analysis and interpretation. For example, to determine if children with DFA more likely to have additional psychological difficulties or worse HRQoL than children without DFA.

For the participants that did report symptoms of psychological difficulties there were ethical considerations during the study about whether those participants should be referred for further psychological assessment and management. On receipt of advice from a Consultant Clinical Psychologist (C.C), it was deemed that the measures used to assess child mental health were not diagnostic tools and had been completed in the context of DFA. As the participants were receiving appropriate clinical management for DFA no further action was considered necessary.

8.2.2. Study 2: Exploring children's experiences of dental fear and anxiety

The objectives of this qualitative study were to conduct semi-structured interviews with paediatric dental patients with DFA using a CBT model as the theoretical framework for children's DFA experiences. This was the first study to ask children directly about their DFA experiences. The findings supported the use of the Five Areas™ CBT model as a theoretical framework that considers the thoughts, feelings, behaviours, and physical symptoms that are maintaining children's DFA over-time (Williams and Garland 2002; Porritt et al. 2013). Components of another theoretical model for DFA acquisition, the Cognitive Vulnerability Model (CVM) (see section 2.5.3.2.), were also identified (Armfield et al. 2008).

It should be acknowledged that the background characteristics, and potentially DFA experiences, of the participants in Study 2 did differ from the profile of paediatric dental patients with DFA identified in Study 1, whereby Study 2 compared to Study 1: had a higher female preponderance (77% versus 61%); fewer participants who lived in the deprived areas (31% versus 50%); and more participants who described their ethnicity as 'White' (92% versus 85%). As discussed previously (see Section 5.6.), in Study 2 it was challenging to recruit participants from ethnic minorities. Interestingly, in Study 3, 29% of participants described their ethnicity as Mixed/multiple ethnic groups, Asian/Asian British, Black/African/Caribbean/Black British, or Other ethnic groups, with the highest proportion identifying as Asian/Asian British. This suggests an overrepresentation in the study population compared to the profile described in Study 1 where 85% of participants identified their ethnicity as 'White'. A possible explanation for the difference in ethnicity profile in Study 3, was that the participants were aged between 9 and 16 years, compared to between 11 and 16 years in Studies 1 and 2. Evidence from the 2013 CDHS has demonstrated that at age 8 years, children of Indian and Pakistani ethnicity have higher caries prevalence, than other ethnic groups, whilst by age 15 years those differences have reduced (Rouxel and Chandola 2018). To date, little is known about the relationship between ethnicity and DFA for U.K.

children (Seligman et al. 2017). Further research is needed to explore the role of cultural factors in children’s DFA experiences.

The aim of using the Five Areas™ model as a theoretical framework is that it incorporates an integrative conceptualisation of DFA as a multidimensional experience, and provides a structured assessment approach that considers how the cognitive, affective, physiological and behavioural responses to DFA interact to maintain it overtime (Lang 1968; Kendall 1985; Schuurs and Hoogstraten 1993; Armfield 2010b; Porritt et al. 2013; Buchanan 2017). A point to highlight is that not all participants in the study were able to describe their experiences of all response domains. A possible explanation is that the response components are not equally sensitive and can demonstrate disconcordance (Craske and Craig 1984). Additionally, if an individual was forced to proceed with the dental treatment, regardless of their DFA, a decoupling of their responses could have occurred (Craske and Craig 1984). It is also possible that it is a limitation of the study. Children with DFA were asked to reflect on the DFA experiences, but at the time of the interviews they were not in a dental situation. Children discussed at length their negative thoughts and unhelpful behaviours but found it more difficult to describe their feelings and physical symptoms. Therefore, the total collective findings would suggest children had strong, equal responding in each domain, but for individual children unequal responding and even disconcordance was evident. Children’s experiences may also have been influenced by the metacognitive processes required during the qualitative interviews. These concerns are not likely clinically important if the Five Areas™ model is used as part of a CBT intervention for DFA on an individual patient basis. However, it may have relevance for the CEDAM, which was based on the findings of the qualitative interviews.

8.2.3. Study 3: Development and testing of a web-based version of the Children's Experience of Dental Anxiety Measure for clinical assessment of paediatric dental patients aged 9 to 16 years

The key findings of this study were that migrating the CEDAM onto a web-app for use on a mobile smart device did not affect data equivalence. The use of a mobile smart device also did not affect the time taken to complete the measure, or data completeness of responses. Moreover, both the paper and web-app questionnaire versions had high ratings of acceptability by children. However, DFA assessment generally was associated with social evaluative fears and anxieties and contributed to the DFA experience of some children. No preference was identified for paper or electronic completion of DFA questionnaires, which suggests either approach could be used in clinical practice.

A particular challenge encountered during this study was unreliable and inconsistent internet connectivity in the NHS dental teaching hospital. For the duration of the study, the web-app was hosted on a secure University of Sheffield server and accessed on the internet browser of the iPad over the secure wireless network for the University of Sheffield (Eduroam, Géant). However, once the study was commenced it was identified that the Paediatric Dentistry Clinic had significant signal fluctuations. As an alternative, a 4G mobile hotspot provided by a commercial mobile broadband network (Pocket Hotspot, Telefonica U.K. Limited) was used, and the tablet connected to the University of Sheffield Virtual Private Network. This was used successfully during the development stage of the web-app, when participants were interviewed in their own homes, but in the Paediatric Dentistry clinical area the signal again proved unreliable. The problems with connectivity were finally resolved as the University of Sheffield's IT Services were able to extend their network and improve coverage. Unfortunately, during the early stages of the study, 6.5% (n=7) participants were excluded when they were unable to complete the eCEDAM because it was not possible to connect to the internet. This is a potential study limitation, as these participants had a different experience with the eCEDAM compared to those that were included. However, since the study was concluded, a programme to introduce free Wi-Fi for staff and patients in NHS settings in England has been completed (NHS Digital 2021). This suggests internet connectivity should be less of a concern for future research.

Although this study compared paper and mobile smart devices to collect questionnaire data for children, the different environmental impacts of each approach was not considered. As described in Section 2.6.8., mHealth has usability and clinical advantages over traditional pen and paper questionnaires, and on the surface the use of a tablet computer would appear to be more sustainable and better for the environment. However, there is little published data on their comparative life cycles and associated carbon footprint, and what data is available suggests any comparisons are not straightforward (Moodie 2014). For example, considerations include manufacture, electricity usage during lifetime, and recycling and waste potential (Suksuwan et al. 2020). Additionally, tablets produced by different manufacturers may have different life cycles (Suksuwan et al. 2020). Regardless, the environmental impact of DFA assessment could be reduced if patients could complete questionnaires on mobile smart devices that they already own. For this to be possible the eCEDAM would need to be compatible with different types of devices (e.g. smartphones, tablets). It should be acknowledged, that during this research project sustainability was not considered, and two new tablet computers were obtained for the purposes of data collection in the study only. Future research studies involving electronic equipment should describe how it will be reused and recycled as part of the study design.

8.3. Implications for clinical practice

Overall, this study provided evidence that failures in communication can influence DFA in children. It should be acknowledged that DFA in children, and occupational stressors in dental professionals, can promote situations with miscommunication vulnerabilities (Jones and Huggins 2014). For example, dental professionals may perceive they are providing oral health advice, but it is perceived by children with DFA as being reprimanded. Ultimately it is the responsibility of dental professionals to demonstrate effective communication skills. Recently, communication tools have become available to support interpersonal exchanges between dental professionals and children (Jones 2015; Yee et al. 2017; Rodd et al. 2019). However, standardised DFA questionnaires, as used in this study, have potential to aid communication (Jones and Huggins 2014). Moreover, their usage has been suggested to create an expectancy of empathy by patients (Dailey et al. 2002).

The research has demonstrated that DFA assessment using the CEDAM/eCEDAM is not time-consuming and has high patient acceptability. Dental professionals need to be aware when treating children with DFA, that some children experience assessment anxiety and DFA when completing standardised questionnaires. As there are no differences in data equivalence or completeness between the CEDAM or eCEDAM, dental professionals can choose a mode of delivery for the questionnaire that is appropriate for their clinical practice. However, the web-app version has potential to address barriers for utilisation, such as automatic scoring and providing support for clinical interpretation, that have been identified by dental professionals as important concerns (Alshammasi et al. 2018).

8.4. Implications for further research

As discussed in Chapter 2, existing self-report measures lacked a theoretical framework for DFA, had a narrow conceptual focus, and considered the construct of DFA in children from an adult perspective. As measures, by the nature of their use in research, define the concept of DFA in children, it is important that they have construct validity (Armfield 2010b). The research presented in this thesis has progressed conceptual understanding of DFA assessment in paediatric dental patients and contributed to the development of a new child centred DFA assessment measure based on a CBT theoretical framework for DFA. However, the research has highlighted that, conceptually, DFA in paediatric dental patients remain not fully understood.

A key area for future research is further evaluation of the Five Areas™ CBT model as a theoretical framework for DFA in children. Generally, the findings of Study 2, support its use. That is, children with DFA experienced thoughts, feelings, behaviours, and physical symptoms related to DFA. A potential consideration is that it is not known if each of these response domains are equally important in children's overall DFA experiences. During qualitative interviews, children were better able to describe, their thoughts, than their behaviours, feelings, and physical symptoms associated with DFA. The recent development of the CEDAM-8 provides further evidence for cognitions being an important factor in DFA experience. During measure development an item impact analysis was used to identify the items from the CEDAM-14 that are most important to participants (Porritt et al. 2021). Of note, the data set used in the analysis include the results for the paper versions of the CEDAM described in Study 3. The three items with the highest proportion of children reporting an impact were all based on thoughts (Porritt et al. 2021). Interestingly, these items all related to the CVM (see Section 2.5.3.2), and perceptions of threat/danger ('Think it will be painful'), unpredictability ('Worry if I need to have something done'), and uncontrollability ('Control over what happens'). Further research is needed to understand the contribution of each response system to DFA assessment in children. However, the findings from this research suggests that potentially it is DFA-related cognitions that are most important in DFA assessment, and greater research focus should be on children's perceptions of vulnerability in the dental setting.

An additional research consideration is that little is known about the factors that may influence how children self-report DFA on assessment questionnaires. Firstly, the research presented provides tentative evidence that when children with DFA self-report their DFA using trait measures, their responses are heavily influenced by the state DFA they are experiencing at a particular time. This has potential relevance if DFA measures are being used to quantify children's DFA severity as this suggests it may not be a stable characteristic. Secondly, in adults, it is well accepted that different typologies of DFA are possible (Aartman et al. 1999; Locker et al. 1999b; Armfield and Heaton 2013). Further research is needed into different DFA typologies in children and their associated characteristics. As described in

Section 8.2.1., assessing DFA using standardised questionnaires and associated scoring in children who have DFA towards a specific dental stimulus or procedure is problematic, as although their DFA may be less global, it is not necessarily less severe. The last factor for consideration in DFA self-assessment, is the need to explore children's stigma related perceptions of DFA. Participants in this research described being embarrassed about their DFA, and not wanting others to find out. Although not explored during this research, dental professionals may contribute to this stigma, as children with DFA are frequently labelled as being uncooperative (i.e. demonstrated bad behaviour), whereas children who complete dental treatment are rewarded for being brave. The role of dental professionals and parents in children's stigma related DFA perceptions requires further research.

Although most children referred with DFA did not have psychological difficulties, it should be acknowledged that 13% and 40% of the study participants did report symptoms that reached the borderline clinical/clinical threshold for anxiety and depression, and for social, emotional, and behavioural difficulties, respectively. Moreover, this was a cross-sectional study that assessed child mental health characteristics on a single occasion. Therefore, it is not known if participants previously had psychological difficulties or would have psychological problems in the future. Although child mental health assessment of all children with DFA is not recommended from this research, a finding is that there is a patient group of children with both DFA and symptoms of psychological difficulties. The available evidence would suggest that adults with DFA and concomitant psychological disorders are more likely to have negative DFA treatment outcomes and to have DFA that is maintained over time (Aartman et al. 1999; Locker et al. 2001a). However, treatment outcomes for participants reporting symptoms of psychological difficulties were not evaluated. Further research is needed to evaluate the clinical features and treatment outcomes for children with DFA and concomitant psychological difficulties to evaluate if this a priority group for intervention.

8.5. Ethical considerations

The study was conducted in accordance with the Research Ethics Committee and Health Research Authority protocols and was subject to regular progress meetings. No child, parent or staff-related concerns were raised throughout the study. However, as described earlier, clinical advice was sought regarding the most appropriate action to take for those participants who were identified as having symptoms of psychological disorders. In this instance further specialist referral was not advocated. The potential to identify other needs, or indeed safeguarding concerns, should always be considered in studies involving children, and appropriate action should be discussed with the whole team.

Chapter Nine

9. Conclusions and recommendations

The studies within this thesis aimed to further the understanding of DFA assessment in paediatric dental patients. The research objectives were to: (1) assess the sociodemographic, quality of life, and child mental health characteristics of paediatric dental patients with DFA; (2) explore the experiences of DFA in paediatric dental patients to inform the development of a new child-centred measure of DFA; and (3) design and test a web based DFA assessment measure for use on mobile smart devices. To achieve these study goals, a mixed method study design and a child centred approach was used. This chapter provides a summary of the key research findings, study limitation, and recommendations.

9.1. Summary of findings

1. The sociodemographic profile of paediatric dental patients aged 11-16 years, who had been referred to secondary/tertiary care for DFA management, found that children were mostly female, identified as White ethnicity, and lived in areas with high levels of deprivation.
2. Paediatric dental patients referred with DFA reported a range of DFA severity scores following completion of standardised DFA assessment questionnaires.
3. Most paediatric dental patients with DFA did not have additional psychological difficulties. Correspondingly, universal child mental health assessment during DFA assessment is not recommended.
4. Higher DFA scores were associated with poorer HRQoL. However, levels of impacts were consistent with non-clinical populations.

5. The Five Areas™ CBT model is applicable as a theoretical framework for DFA assessment that considers children's thoughts, feelings, physical symptoms, and behaviours associated with DFA.
6. Children expressed confidence in the use of mobile smart devices to complete DFA assessment measures and made suggestions to the design of the eCEDAM that would promote inclusiveness and autonomy.
7. Completion of the eCEDAM did not affect data equivalence, data completeness, time taken to complete the questionnaire, or participant preference for mode of delivery compared to the original paper version.
8. Both the CEDAM/eCEDAM were quick to complete and had high acceptability ratings.
9. DFA assessment did result in assessment anxiety and contribute to DFA in some participants.

9.2. Summary of research limitations

1. The study population of children referred with DFA from primary/secondary care may not be representative of children with DFA generally.
2. Study recruitment factors may have introduced bias. Considerations include: children who were approached for inclusion in the study but were not brought to their first appointment; recruitment of participants from certain population groups to all research stages; exclusion of children with internet connectivity issues during completion of the eCEDAM; and approaching children as potential participants in an open waiting area where there was insufficient privacy.
3. For an evaluation of the sociodemographic, child mental health and HRQoL characteristics in children referred with DFA, a control group of patients without DFA would have allowed further statistical analysis and interpretation of findings.
4. During CEDAM/eCEDAM data agreement testing, participants had a washout period between completing the first and second questionnaires of 15 minutes. This short

duration may have allowed for carry-over effects and children remembering their previous responses.

9.3. Recommendations for clinical practice

1. Dental professionals need to be aware of the importance of effective communication with children with DFA.
2. Dental professionals should consider the use of standardised DFA assessment questionnaires, or use specifically developed communication tools, to support their communication with children with DFA.
3. Both the CEDAM and eCEDAM are suitable for use within clinical practice.

9.4. Recommendations for further research

1. Further evaluation of the Five Areas™ CBT model as a framework for DFA in children is needed to explore disconcordance and parity of importance between responses.
2. Future research should focus on perceptions of vulnerability (threat/danger, unpredictability, uncontrollability, and disgust) in children's DFA-related cognitions.
3. Further research is needed into the factors that influence DFA self-reporting by children. Considerations include a social desirability response bias, different typologies of DFA (e.g. global, specific), and the influence of state DFA.
4. Evaluation of the oral health status and treatment outcomes in children with DFA and concomitant psychological difficulties is recommended. Potentially this is a patient group that should be considered a high priority for early clinical intervention.
5. Further research is needed to determine the impact of DFA on HRQoL for paediatric dental patients.

9.5. Conclusions

Dental fear and anxiety is a multidimensional experience for children. Further research is needed to explore children's self-reporting of DFA using standardised DFA questionnaires. As most children with DFA did not have additional psychological difficulties, child mental health screening during routine DFA assessment in clinical practice is not recommended. The use of a mobile smart device for DFA assessment is acceptable to patients and has potential to address scoring and interpretation clinical utilisation barriers in dental professionals.

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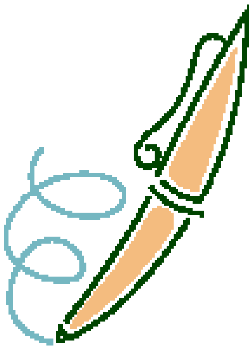
10. Appendices

Appendix 1. **Participant questionnaire booklet for Study 1**

28th November 2013 v4





Questionnaire about going to the dentist and how you feel

Hello

Thanks for agreeing to help us with our study. This study is being done so we understand more about how young people feel about going to the dentist and how they feel about themselves. By answering the questions, you will help us develop ways to make young people feel happier about visiting a dentist.

In this booklet, you will find some sets of questions about you, how you feel about going to the dentist and how you feel about yourself.

We would be very grateful if you could answer all the questions using the instructions. There are no right or wrong answers. Some of the questions may seem to be asking the same thing but each question tells us about something slightly different that we would like to find out.

Section 1 About you

1. Are you: (please tick)

A boy .... ☐

A girl .... ☐

2. How old are you? ....years

3. What is your home postcode | | | | | | |

Please ask your parent or carer if you need help with this

Section 2 How afraid are you of the following things?

For these next questions I would like you to tell me how relaxed or worried you get about going to the dentist and what happens at the dentist.

How do you feel about: (please tick)

	Relaxed/not worried	Very slightly worried	Fairly worried	Worried a lot	Very worried
Going to the dentist generally?					
The night before going to the dentist?					
Sitting in the waiting room?					
Sitting in the dental chair?					
Lying back in the dental chair?					
Having somebody put instruments in your mouth?					
The smell of the dentists room?					
Having your teeth looked at?					
Having your teeth cleaned and polished?					
Having an injection in your gum?					
Having your teeth drilled?					
Having a filling?					
Having a tooth taken out?					
Being put to sleep to have treatment?					
Having a mixture of gas and air which will help you feel comfortable for treatment but cannot put you to sleep?					

If a workbook was available for you to do to help you feel happier about visiting the dentist how interested would you be? (please tick)	
☐	Not interested at all
☐	A little interested
☐	Neither interested nor disinterested
☐	Quite interested
☐	Very interested

Section 3 How do you feel?

These questions ask about how you are **today.** They are not about going to the dentist. For each question, read all the choices and decide which one is most like you today. Then put a tick in the box next to it. Only tick one box for each question.

1. Worried	
☐	I don't feel worried today
☐	I feel a little bit worried today
☐	I feel a bit worried today
☐	I feel quite worried today
☐	I feel very worried today

2. Sad	
☐	I don't feel sad today
☐	I feel a little bit sad today
☐	I feel a bit sad today
☐	I feel quite sad today
☐	I feel very sad today

3. Pain	
☐	I don't have any pain today
☐	I have a little bit of pain today
☐	I have a bit of pain today
☐	I have quite a lot of pain today
☐	I have a lot of pain today

4. Tired	
☐	I don't feel tired today
☐	I feel a little bit tired today
☐	I feel a bit tired today
☐	I feel quite tired today
☐	I feel very tired today

5. Annoyed

- ☐ I don't feel annoyed today
- ☐ I feel a little bit annoyed today
- ☐ I feel a bit annoyed today
- ☐ I feel quite annoyed today
- ☐ I feel very annoyed today

6. School work/homework (such as reading, writing, doing lessons)

- ☐ I have no problems with my schoolwork/homework today
- ☐ I have a few problems with my schoolwork/homework today
- ☐ I have some problems with my schoolwork/homework today
- ☐ I have many problems with my schoolwork/homework today
- ☐ I can't do my schoolwork/homework today

7. Sleep

- ☐ Last night I had no problems sleeping
- ☐ Last night I had a few problems sleeping
- ☐ Last night I had some problems sleeping
- ☐ Last night I had many problems sleeping
- ☐ Last night I couldn't sleep at all

8. Daily routine (things like eating, having a bath/shower, getting dressed)

- ☐ I have no problems with my daily routine today
- ☐ I have a few problems with my daily routine today
- ☐ I have some problems with my daily routine today
- ☐ I have many problems with my daily routine today
- ☐ I can't do my daily routine today

9. Able to join in activities (things like playing out with your friends, doing sports, joining in things)

- ☐ I can join in with any activities today
- ☐ I can join in with most activities today
- ☐ I can join in with some activities today
- ☐ I can join in with a few activities today
- ☐ I can join in with no activities today

Please put a tick in the box that shows how often each of these things happen to you. There are no right or wrong answers.

	Never	Sometimes	Often	Always
I worry about things	☐	☐	☐	☐
I feel sad or empty	☐	☐	☐	☐
When I have a problem, I get a funny feeling in my stomach	☐	☐	☐	☐
I worry when I think I have done poorly at something	☐	☐	☐	☐
I would feel afraid of being on my own at home	☐	☐	☐	☐
Nothing is much fun anymore	☐	☐	☐	☐
I feel scared when I have to take a test	☐	☐	☐	☐
I feel worried when I think someone is angry with me	☐	☐	☐	☐
I worry about being away from my parents	☐	☐	☐	☐
I get bothered by bad or silly thoughts or pictures in my mind	☐	☐	☐	☐
I have trouble sleeping	☐	☐	☐	☐
I worry that I will do badly at my school work	☐	☐	☐	☐
I worry that something awful will happen to someone in my family	☐	☐	☐	☐
I suddenly feel as if I can't breathe when there is no reason for this	☐	☐	☐	☐
I have problems with my appetite	☐	☐	☐	☐

	Never	Sometimes	Often	Always
I have to keep checking that I have done things right (like the switch is off, or the door is closed)	☐	☐	☐	☐
I feel scared if I have to sleep on my own	☐	☐	☐	☐
I have trouble going to school in the mornings because I feel nervous or afraid	☐	☐	☐	☐
I have no energy for things	☐	☐	☐	☐
I worry I might look foolish	☐	☐	☐	☐
I am tired a lot	☐	☐	☐	☐
I worry that bad things will happen to me	☐	☐	☐	☐
I can't seem to get bad or silly thoughts out of my head	☐	☐	☐	☐
When I have a problem, my heart beats really fast	☐	☐	☐	☐
I cannot think clearly	☐	☐	☐	☐
I suddenly start to tremble or shake when there is no reason for this	☐	☐	☐	☐
I worry that something bad will happen to me	☐	☐	☐	☐
When I have a problem, I feel shaky	☐	☐	☐	☐
I feel worthless	☐	☐	☐	☐
I worry about making mistakes	☐	☐	☐	☐
I have to think of special thoughts (like numbers or words) to stop bad things from happening	☐	☐	☐	☐

	Never	Sometimes	Often	Always
I worry what other people think of me	☐	☐	☐	☐
I am afraid of being in crowded places (like shopping centers, the cinema, buses, busy playgrounds)	☐	☐	☐	☐
All of a sudden I feel scared for no reason at all	☐	☐	☐	☐
I worry about what is going to happen	☐	☐	☐	☐
I suddenly become dizzy or faint when there is no reason for this	☐	☐	☐	☐
I think about death	☐	☐	☐	☐
I feel afraid if I have to talk in front of my class	☐	☐	☐	☐
My heart suddenly starts to beat too quickly for no reason	☐	☐	☐	☐
I feel like I don't want to move	☐	☐	☐	☐
I worry that I will suddenly get a scared feeling when there is nothing to be afraid of	☐	☐	☐	☐
I have to do some things over and over again (like washing my hands, cleaning or putting things in a certain order)	☐	☐	☐	☐
I feel afraid I will make a fool of myself in front of people	☐	☐	☐	☐
I have to do some things in just the right way to stop bad things from happening	☐	☐	☐	☐
I worry when I go to bed at night	☐	☐	☐	☐
I would feel scared if I had to stay away from home overnight	☐	☐	☐	☐
I feel restless	☐	☐	☐	☐

Nearly finished!
For each item, please put a tick in the box for Not True, Somewhat True or Certainly True.
It would help if you answered all items as best you can, even if you are not absolutely certain or the question seems daft! Please give your answers on the basis of how things have been over the last **six months**.

	Not True	Somewhat True	Certainly True
I try to be nice to other people. I care about their feelings	☐	☐	☐
I am restless, I cannot stay still for long	☐	☐	☐
I get lots of headaches, stomach-aches or sickness	☐	☐	☐
I usually share with others (food, games, pens, etc.)	☐	☐	☐
I get very angry and often lose my temper	☐	☐	☐
I am usually on my own. I generally play alone or keep to myself	☐	☐	☐
I usually do as I am told	☐	☐	☐
I worry a lot	☐	☐	☐
I am helpful if someone is hurt, upset or feeling ill	☐	☐	☐
I am constantly fidgeting or squirming	☐	☐	☐
I have one good friend or more	☐	☐	☐

	Not True	Somewhat True	Certainly True
I fight a lot. I can make other people do what I want	☐	☐	☐
I am often unhappy, down-hearted or tearful	☐	☐	☐
Other people my age generally like me	☐	☐	☐
I am easily distracted. I find it difficult to concentrate	☐	☐	☐
I am nervous in new situations. I easily lose confidence	☐	☐	☐
I am kind to younger children	☐	☐	☐
I am often accused of lying or cheating	☐	☐	☐
Other children or young people pick on me or bully me	☐	☐	☐
I often volunteer to help others (parents, teachers, children)	☐	☐	☐
I think before I do things	☐	☐	☐
I take things that are not mine from home, school or elsewhere	☐	☐	☐
I get on better with adults than with people my own age	☐	☐	☐
I have many fears, I am easily scared	☐	☐	☐
I finish the work I'm doing, my attention is good	☐	☐	☐

Section 4

Last question! We would like to know whether the way young people think about going to the dentist is affected by their background.
Please tick the box that best describes your ethnic group, you only need to tick **one** box on this page. Ask your parent or carer to help you with this section if you need to.

A: White

☐ English/Welsh/Scottish/Northern Irish/British
☐ Irish
☐ Gypsy or Traveller
☐ Any other White background, write in

B: Mixed/multiple ethnic groups

☐ White and Black Caribbean
☐ White and Black African
☐ White and Asian
☐ Any other Mixed/multiple ethnic background, write in

C: Asian/Asian British

☐ Indian
☐ Pakistani
☐ Bangladeshi
☐ Chinese
☐ Any other Asian Background, write in

D: Black/African/Caribbean/Black British

☐ African
☐ Caribbean
☐ Any other Black/African/Caribbean background, write in

E: Other ethnic group

☐ Arab
☐ Any other ethnic group, write in

Finally, please let us know whether you would be willing for us to contact you again about this project? Yes ☐ No ☐

Thank you very much for your help

Appendix 2. Skewness and Kurtosis data for MCDAS, RCADS, SDQ and CHU-9D scales and subscales

	Skewness		Kurtosis	
Measure	Statistic	Standard error	Statistic	Standard error
Modified Child Dental Anxiety Scale				
Total DFA score	0.1	0.2	-0.6	0.5
Total DFA score for completed at home	-0.3	0.4	0.0	0.7
Total DFA score for completed on dental clinic	0.3	0.3	-0.7	0.6
Total DFA score for male participants	0.4	0.4	-0.9	0.7
Total DFA score for female participants	0.4	0.3	-0.1	0.6
Total DFA score for participants aged 11 years	-0.7	0.6	-0.3	1.2
Total DFA score for participants aged 12 years	0.0	0.5	-1.4	0.9
Total DFA score for participants aged 13 years	0.1	0.5	-0.4	1.0
Total DFA score for participants aged 14 years	-0.2	0.5	-0.5	1.0
Total DFA score for participants aged 15 years	0.4	0.5	-0.3	1.0
Total DFA score for deprivation quintile 1	0.4	0.3	-0.5	0.7
Total DFA score for deprivation quintile 2	-0.3	0.7	-0.6	1.3
Total DFA score for deprivation quintile 3	0.1	0.5	-0.8	1.0
Total DFA score for deprivation quintile 4	0.4	0.7	-0.2	1.4
Total DFA score for deprivation quintile 5	-0.4	0.6	0.3	1.2
Revised Children’s Anxiety and Depression Scale				
Generalised anxiety disorder	0.9	0.2	0.2	0.5
Major depression disorder	0.5	0.2	-0.6	0.5

	Skewness		Kurtosis	
Measure	Statistic	Standard error	Measure	Statistic
Obsessive compulsive disorder	1.6	0.2	2.9	0.5
Panic disorder	1.5	0.2	1.8	0.5
Separation anxiety disorder	1.4	0.2	1.5	0.5
Social phobia	0.9	0.2	0.3	0.5
Total anxiety	1.2	0.2	0.5	0.5
Total internalising	1.0	0.2	0.2	0.5
Generalised anxiety disorder for male participants	1.1	0.4	0.5	0.7
Generalised anxiety disorder for female participants	0.8	0.3	0.2	0.6
Major depressive disorder for male participants	0.7	0.4	-0.7	0.7
Major depressive disorder for female participants	0.3	0.3	-0.8	0.6
Obsessive compulsive disorder for male participants	1.4	0.4	1.2	0.7
Obsessive compulsive disorder for female participants	1.3	0.3	1.9	0.6
Panic disorder for male participants	1.9	0.4	3.5	0.7
Panic disorder for female participants	1.4	0.3	1.2	0.6
Separation anxiety disorder for male participants	1.8	0.4	2.8	0.7
Separation anxiety disorder for female participants	1.2	0.3	1.1	0.6
Social phobia for male participants	1.1	0.4	0.7	0.7
Social phobia for female participants	0.7	0.3	-0.3	0.6
Total anxiety scale for male participants	1.3	0.4	0.8	0.7
Total anxiety scale for female participants	1.1	0.3	0.7	0.6

	Skewness		Kurtosis	
Measure	Statistic	Standard error	Measure	Statistic
Total internalising scale for male participants	1.2	0.4	0.5	0.7
Total internalising scale for female participants	0.9	0.3	0.4	0.6
Strengths and Difficulties Questionnaire				
Conduct problems	0.7	0.2	-0.8	0.5
Emotional problems	0.3	0.2	-1.0	0.5
Hyperactivity	0.4	0.2	1.3	0.5
Peer problems	1.1	0.2	-0.3	0.5
Total difficulties	0.5	0.2	-0.5	0.5
Conduct problems for male participants	0.8	0.4	0.3	0.7
Conduct problems for female participants	0.5	0.3	-0.5	0.6
Emotional problems for male participants	0.9	0.4	-0.4	0.7
Emotional problems for female participants	0.2	0.3	-0.9	0.6
Hyperactivity for male participants	0.5	0.4	-0.4	0.7
Hyperactivity for female participants	0.2	0.3	-1.0	0.6
Peer problems for male participants	1.1	0.4	1.0	0.7
Peer problems for female participants	1.1	0.3	1.5	0.6
Total difficulties for male participants	0.7	0.4	-0.5	0.7
Total difficulties for female participants	0.4	0.3	-0.4	0.6
Child Health Utility 9D				
Utility score	0.9	0.2	0.6	0.5

Appendix 3. Topic Guide for qualitative interviews in Study 2

Development of a guided self-help CBT resource for the reduction of dental anxiety in young people aged 11-16 years

Topic Guides Phase 1

Young person topic guide

Introduction

- Purpose is to talk to young people (and their parents/carers) about their experiences of living with dental anxiety or fear
- Interview
  - Interview will last as long as the they wish, but on average 45 minutes
  - Use of a digital recorder by researcher
  - Not a test, and no right or wrong answers (young person is the expert)
  - Doesn't have to talk about anything they don't want to
  - Participation is voluntary (can change their mind and stop the interview at any point)
  - Preferred term for people their age (teenager? young person?).
- Confidentiality
  - Answers will be private
  - Can choose a name that they would like to be referred to on the tape
  - If they makes an important disclosure that they needs help with, then a dentist looking after them will contact them and their parents/carers to talk through things and arrange for the right help

Background to dental anxiety

- Tell me a bit about yourself (favourite subject at school, free time when not busy with schoolwork)
- Previous dental experience (*prompt for check-up visits, prevention, fillings, extractions, injections*)?
- What makes you feel anxious/worried or afraid about going to see the dentist?
- What is it about going to the dentist makes you feel this way (*prompt for specific procedures/treatments*)?
- When did your feelings about going to the dentist start (*prompt for age or a specific incident*)?
- Why do you think your feelings about going to the dentist started?
- Do your feelings about going to the dentist bother you? How much?
- Apart from going to the dentist, are there any other things that you feel anxious/worried or afraid about?
- Is there anyone else you know who also feels worried or afraid about going to see the dentist (*prompt for Mother, Father, siblings, and friends*)?

Situational influences

- Does anything make you feel better or worse about going to the dentist?
    - *At home beforehand, in the waiting room before your visit, and when you are at the in the dentist's chair?*
    - *Anything about the dentist or the dental nurse?*
    - *Does the person who comes with you affect you?*
- What brings on your anxious/worried or afraid feelings when you are at the dentist?
- Are there any specific things that make you feel anxious/worried or afraid (*prompt for specific procedures/treatments, dental chair and room, noises, smells, tastes and equipment*)
- Does the dentist do anything that would make you feel better or worse?
- When you go to the dentist do you get any kind of reward? E.g. a gift. What do you get? Do you think getting a reward helps make going to the dentist bearable/more positive?
- What do your parents say or do before your appointments, during your appointments and afterwards? How does this affect your dental anxiety?

Thoughts

- What thoughts are running through your mind about going to the dentist? What bad thoughts?
    - *At home beforehand, in the waiting room before your visit, and when you are in the dentist's chair?*
- When you are at the dentist, what do you think might happen?
- Why are you anxious/worried or afraid about this happening?
- What is the worst thing you think might happen? What could you do to control this?
- What words or images do you have in your mind when you go to the dentist? Could you draw or tell us about any images you might have?
- What are you scared other people might think of you when you are at the dentist?

Feelings

- How do you feel about going to the dentist?
    - *At home beforehand, in the waiting room before your visit, when you are in the dentist's chair?*
- Do you have any other feelings other than being anxious/worried/afraid when you are at the dentist? Do you feel out of control?
- How do you feel after you have been to see the dentist?

Physical symptoms 'body'

- What happens to your body when you are thinking about going to the dentist, or when you are at the dentist?
- What happens to your senses when you feel this way (*prompt for hearing, vision, smell, taste*)?

- How does it affect you when you feel this way (*prompt for heart beating fast, dry mouth, feeling sick, stomach hurts, sweating and clammy, feeling faint*)?

Behaviours

- When you know you are going to see the dentist when do you start to have anxious/worried or afraid feelings (*prompt for timeframe*)?
- Do you try to avoid going to dentist (*prompt if answer is 'yes' to explain*)?
- Do you sometimes end up missing or cancelling your appointments?
- So when you are actually at the dentist, what do you do then when you have these feelings?
- Do you think these feelings make you behave differently (*prompt for some people say they feel grumpy or angry/ some people say they talk more or become very quiet*)?
- When you feel like that do you do anything to help you cope with those feelings?

The resource

- How would you feel about having a resource which is designed to make you feel better about going to the dentist?
- Ideally, what would this resource look like? How long? Done in an hour or 2 or over a few weeks? What format? E.g. workbook, booklet, website, app. What should it have in it? Activities? Videos? Young people's stories?
- Would you prefer a resource you went through on your own? Or one you did with your parent? Or a dentist or nurse? Should this support be provided face-to-face or via email or telephone or text?
- What would stop you using a resource?

Closing

- Covered everything, or is there anything else you want to raise? What could dentists do to make going to the dentist easier for young people? What would you tell a friend about how to cope with going to the dentist?
- How would they themselves rate their level of dental anxiety: mild, moderate or severe?

Next steps

- Thank the participant
- Reassurance again about confidentiality
- Discuss that the findings will be used to development a guided self-help CBT resource reduce dental anxiety, and the results will be published in a scientific journal. We will send all the participants a report so they know what we found out
- Give the participant a gift voucher

Appendix 4. Topic Guide for cognitive and usability interviews in Study 3

Developing a dental anxiety questionnaire app for use on a tablet

Topic Guide

Introduction

- Purpose is to talk to young people (and parents/carers) about the design of the web-based app for an iPad and how we could make it relevant and young person friendly – it is to find a way of easily measuring how anxious a young person is at the dentist without using paper questionnaires, which other young people don't tend to like!
- Young person will try out the questionnaire on an iPad. They will then be asked to 'think aloud' whilst completing it.
- Interview will last as long as they wish, but on average 30 minutes
- Use of a digital recorder by researcher
- Not a test, and no right or wrong answers (young person is the expert)
- Doesn't have to talk about anything they don't want to
- Participation is voluntary (can change their mind and stop the interview at any point)
- Answers will be private
- Can choose a name that they would like to be referred to on the tape
- Complete consent forms for participant and parents/carers
- Complete MCDAS

Think Aloud task prompts

- What are you thinking now?
- What are your thoughts about this page?
- Can you tell me more about that?
- How are you feeling?
- How do you feel about that?
- Why did you click on that?

Specific design questions

- What features did you like/dislike about the dental anxiety questionnaire app?
- Appearance (colours, colour thermometer for scale, graphics)
- Font and font size
- Welcome/home page (content, features to establish trustworthiness/credibility)
- Introduction and completion text (length, should there be an audio option?)
- Navigation (e.g. preferences for scrolling or one item per page, progress bar)
- Error messages (clear?)
- Need for animations, photos
- Do you want to see your results immediately or have your dental professional talk to you about them?
- Need for a help button (text, audio?)
- Any other design features

Interview

- Describe previous experience of tablet computers and smartphones?
- Do you think the dental anxiety questionnaire app would be useful for you?
- When/where would you want to complete the app
- Could you tell me about whether you would be interested in using it?
- What would you think about using it a second time, third time?
- Can you see problems or concerns with using it?
- Is there anything else you would like it to do?
- Is there anything you can suggest to make the app more fun and interesting

System Usability Scale

- What did you think about the questionnaire?
- Were there any parts of the questionnaire that were hard for you to understand?

Next steps

- Thank the participant
- Reassurance again about confidentiality

- Discuss that the findings will be used to development the dental anxiety questionnaire app and the results will be published in a scientific journal. We will send all the participants a report, so they know what we found out (post/email?)
- Give the participant a gift voucher and ask them to sign receipt

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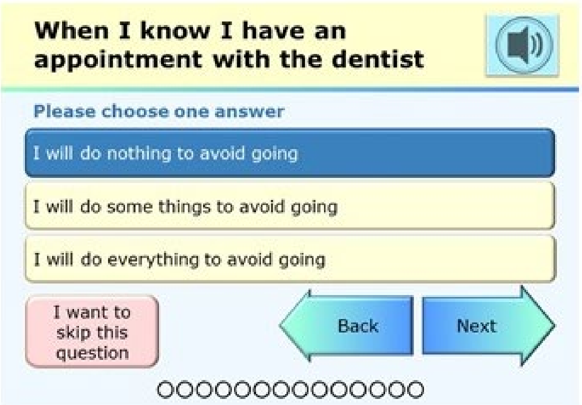
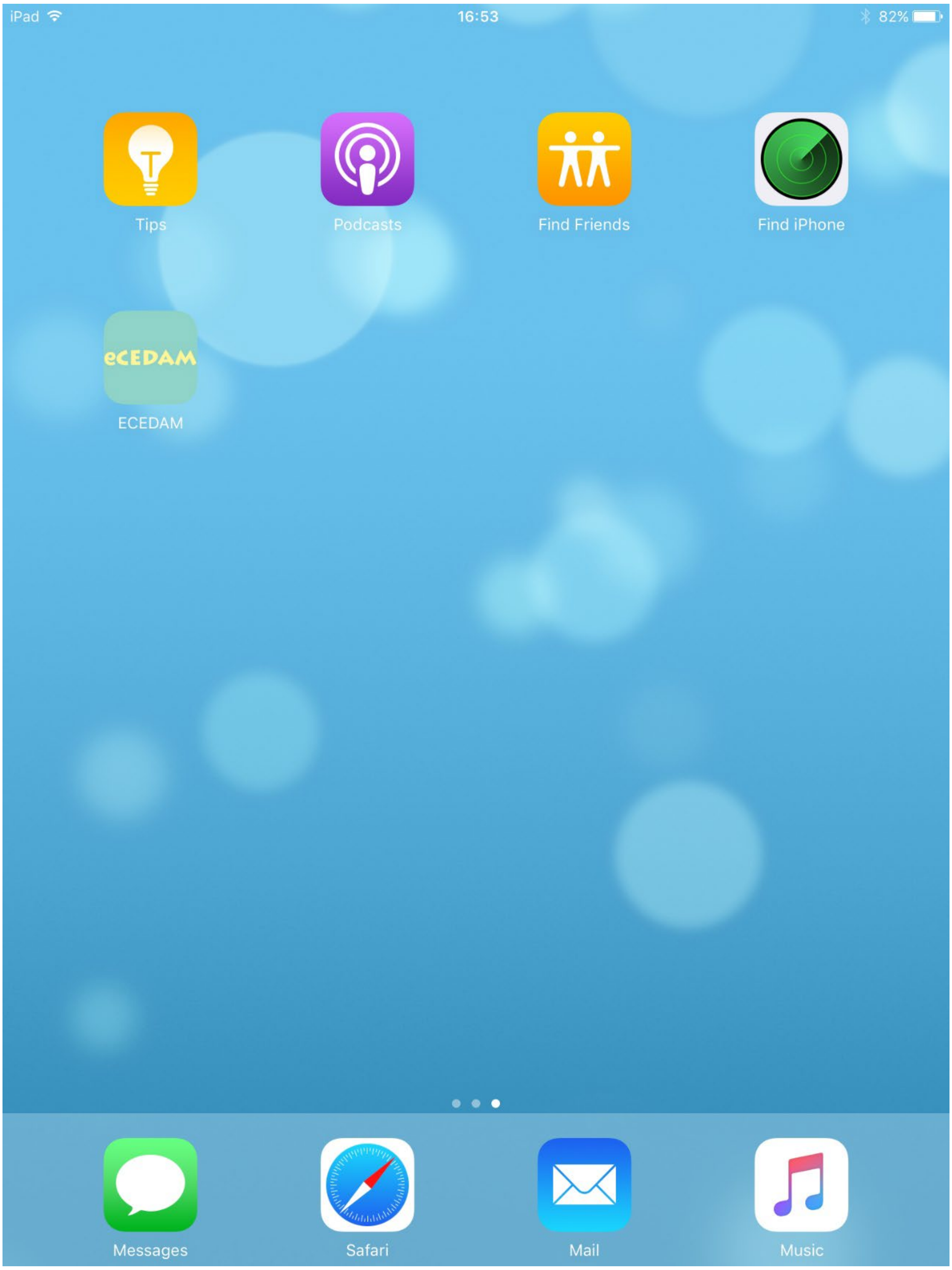
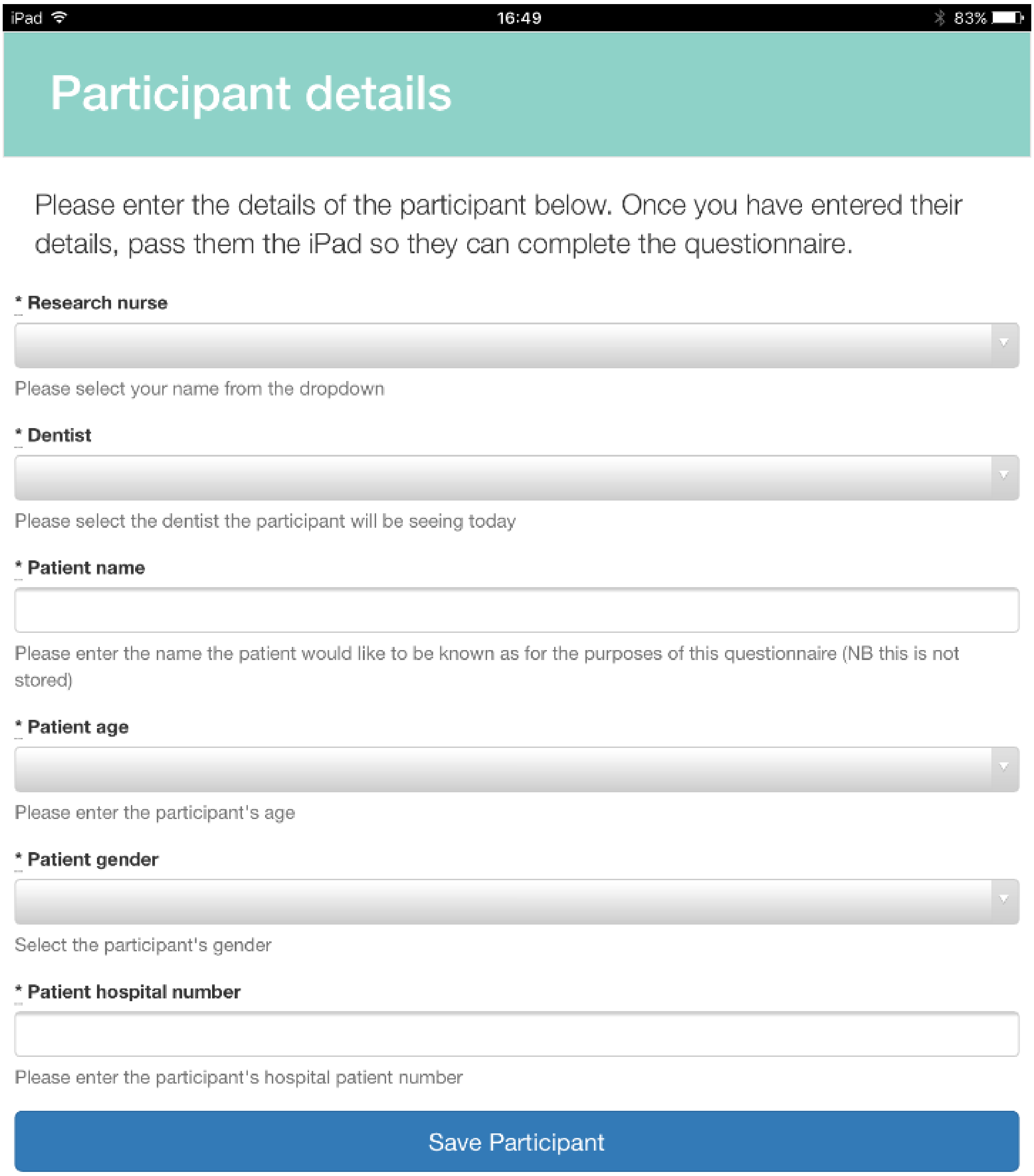
Appendix 5. Iterative cycle development for a single page of eCEDAM



Appendix 6. Screenshots of the eCEDAM prototype





iPad 16:49 83%

Participant details

Please enter the details of the participant below. Once you have entered their details, pass them the iPad so they can complete the questionnaire.

*** Research nurse**

Please select your name from the dropdown

*** Dentist**

Please select the dentist the participant will be seeing today

*** Patient name**

Please enter the name the patient would like to be known as for the purposes of this questionnaire (NB this is not stored)

*** Patient age**

Please enter the participant's age

*** Patient gender**

Select the participant's gender

*** Patient hospital number**

Please enter the participant's hospital patient number

Save Participant

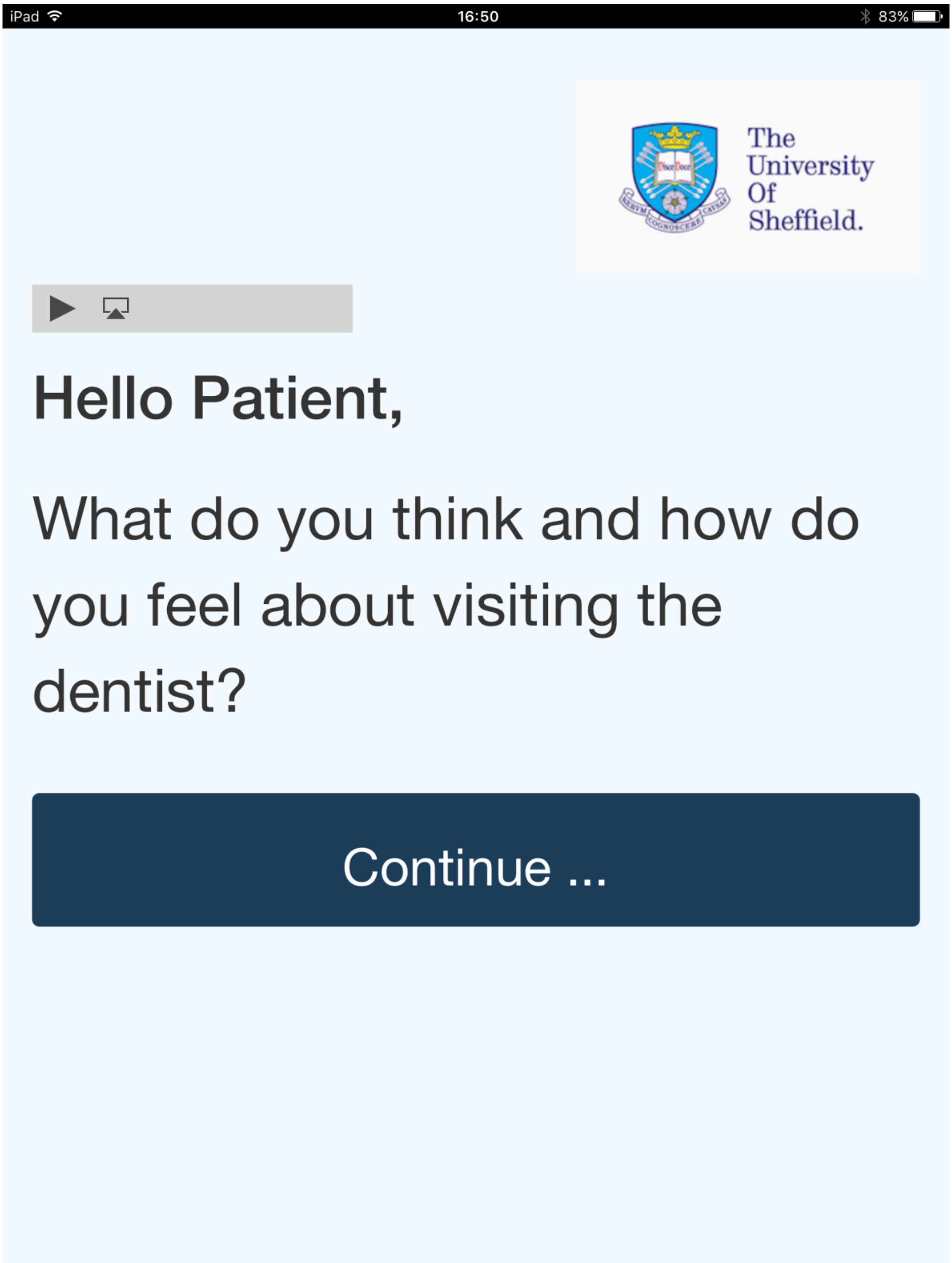
iPad 16:50 83%

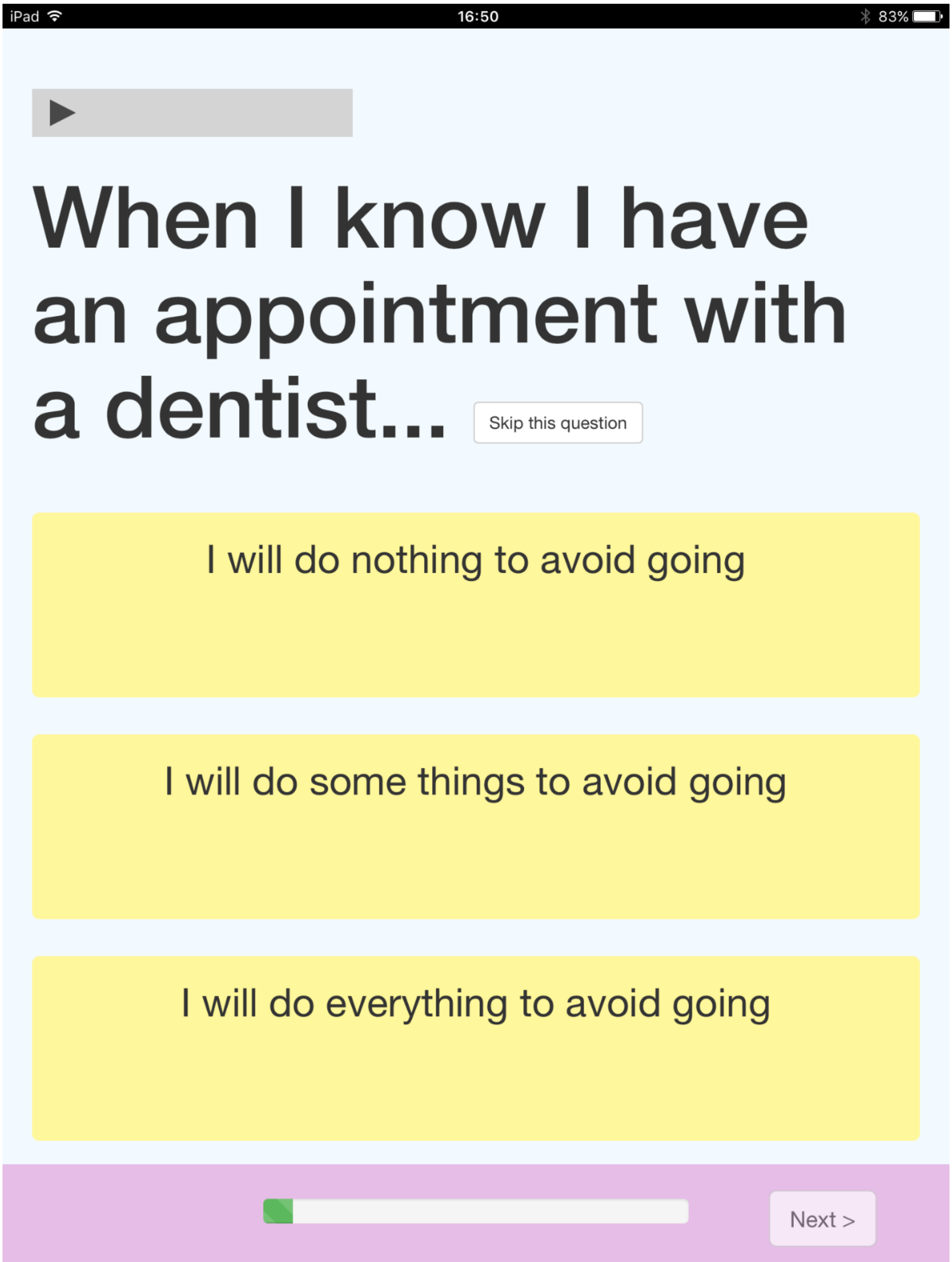
Instructions

For each question please read all the choices and decide which one is most like you today.

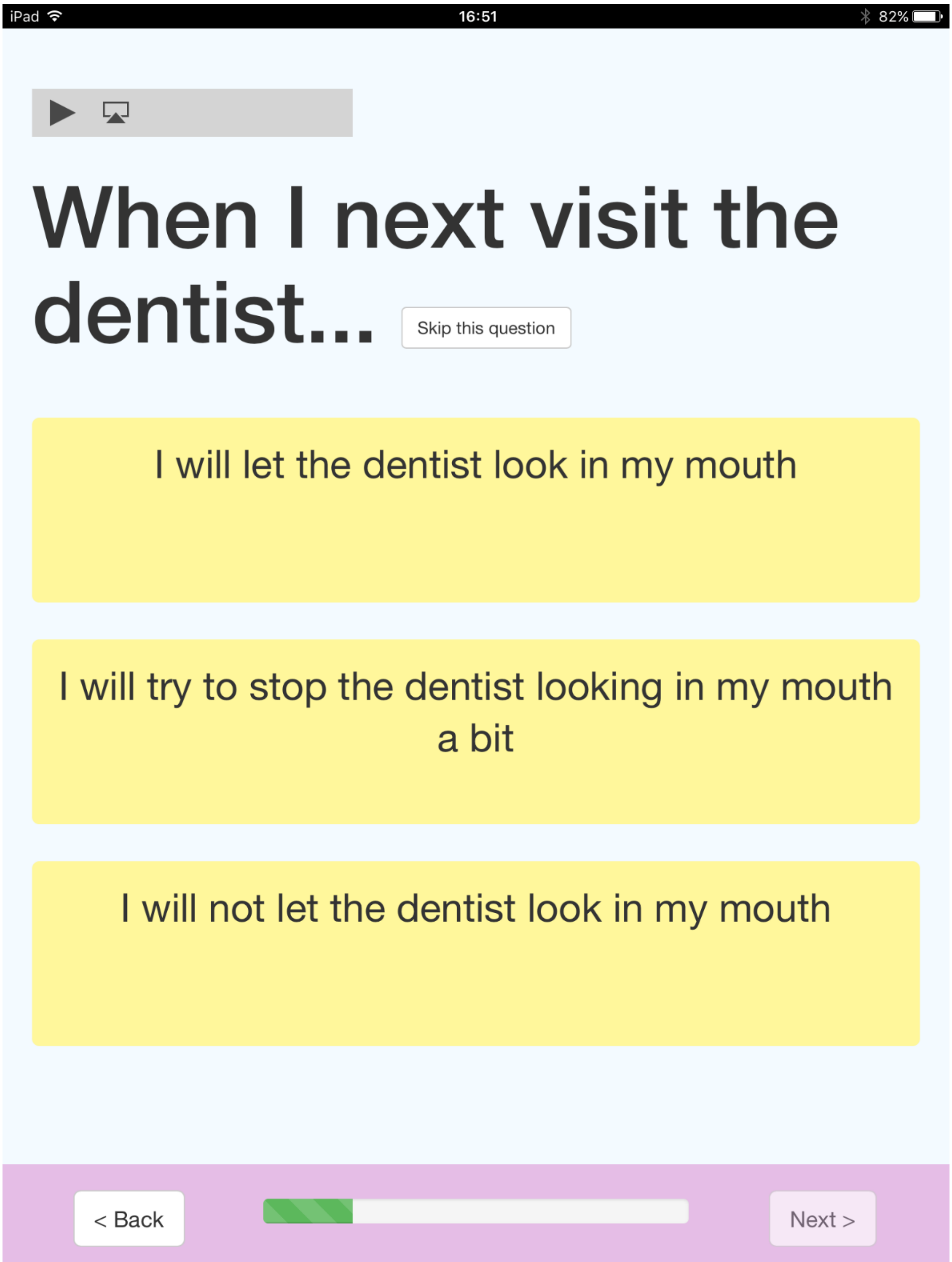
The questions have no right or wrong answers.

Start questionnaire


iPad
16:50
83%
The University Of Sheffield.
Hello Patient,
What do you think and how do you feel about visiting the dentist?
Continue ...


iPad
16:50
83%
When I know I have an appointment with a dentist...
Skip this question
I will do nothing to avoid going
I will do some things to avoid going
I will do everything to avoid going
Next >


iPad
16:50
82%
When I know I have an appointment with a dentist...
Skip this question
I will tell my parents I don't mind going
I will tell my parents I would rather not go
I will tell my parents I really don’t want to go
< Back
Next >


iPad
16:51
82%
When I next visit the dentist...
Skip this question
I will let the dentist look in my mouth
I will try to stop the dentist looking in my mouth a bit
I will not let the dentist look in my mouth
< Back
Next >

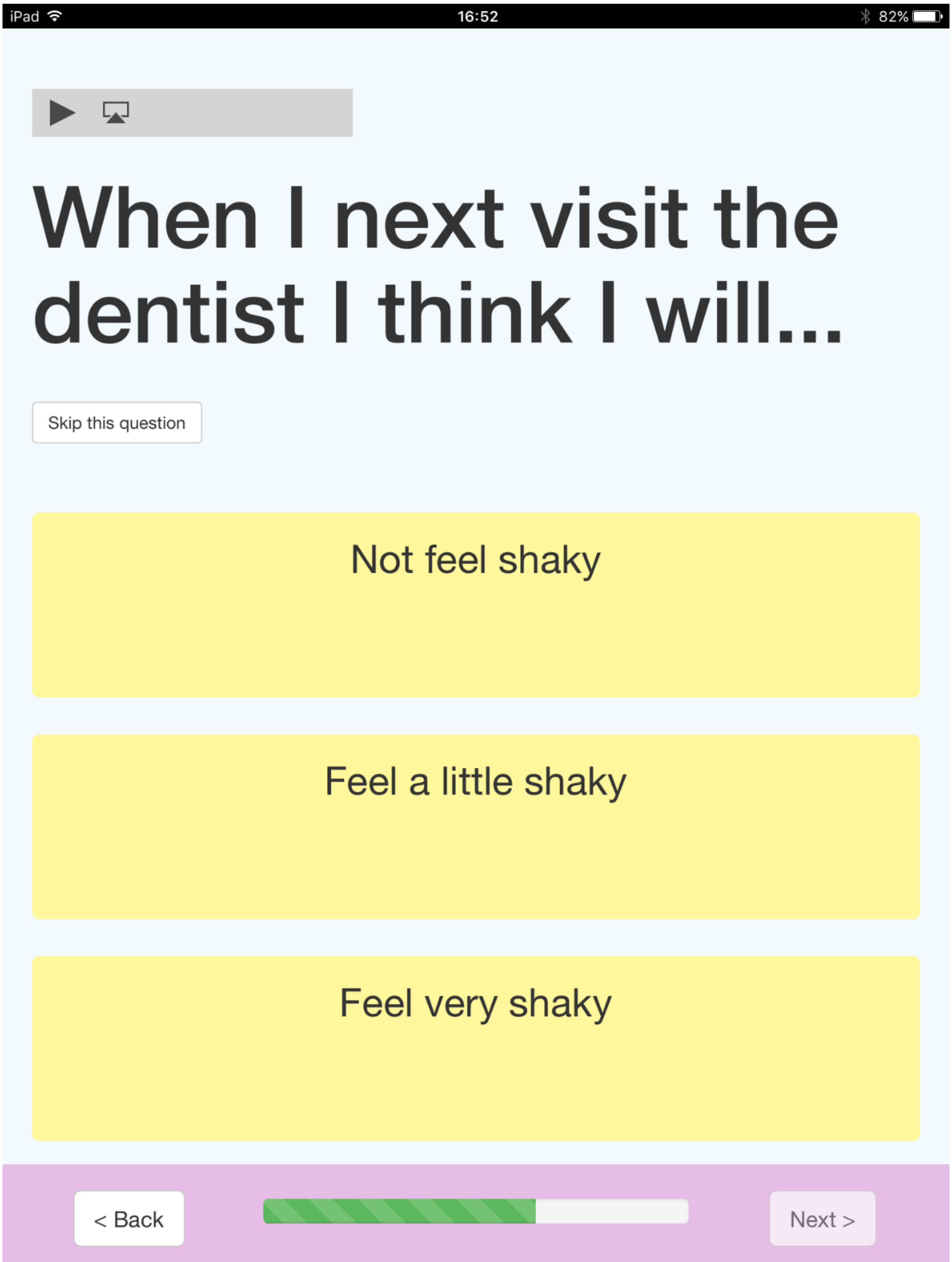

iPad
16:51
82%
When I next visit the dentist I think...
Skip this question
I will not get worried if the dentist tells me I need to have something done
I will get a little worried if the dentist tells me I need to have something done
I will get really worried if the dentist tells me I need to get something done
< Back
Next >

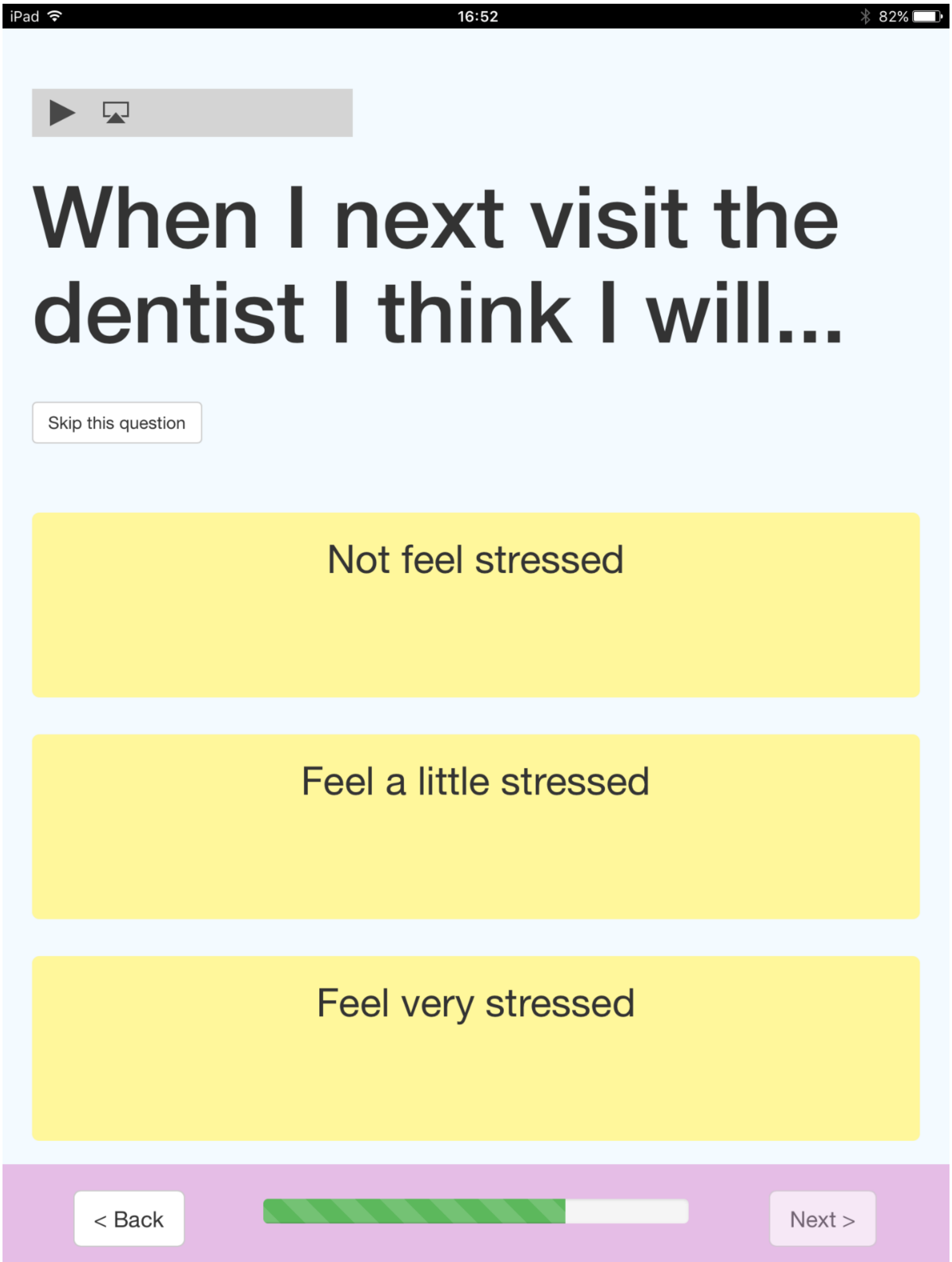

iPad
16:51
82%
When I next visit the dentist I think...
Skip this question
If I asked the dentist to stop what they were doing they would definitely stop
If I asked the dentist to stop what they were doing they might stop
If I asked the dentist to stop what they were doing they would not stop
< Back
Next >


iPad
16:51
82%
When I next visit the dentist I think...
Skip this question
I will not be worried that it will be painful
I will be a little worried that it will be painful
I will be very worried that it will be painful
< Back
Next >


iPad
16:51
82%
When I next visit the dentist I think...
Skip this question
Nothing will go wrong
Something will go a bit wrong
Something will go very wrong
< Back
Next >


iPad
16:51
82%
When I next visit the dentist I think...
Skip this question
I will have a lot of control over what happens in the appointment
I will have a bit of control over what happens in the appointment
I will not have any control over what happens in the appointment
< Back
Next >


iPad
16:52
82%
When I next visit the dentist I think I will...
Skip this question
Not feel shaky
Feel a little shaky
Feel very shaky
< Back
Next >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When I next visit the dentist I think I will...
Skip this question
Not feel stressed
Feel a little stressed
Feel very stressed
< Back
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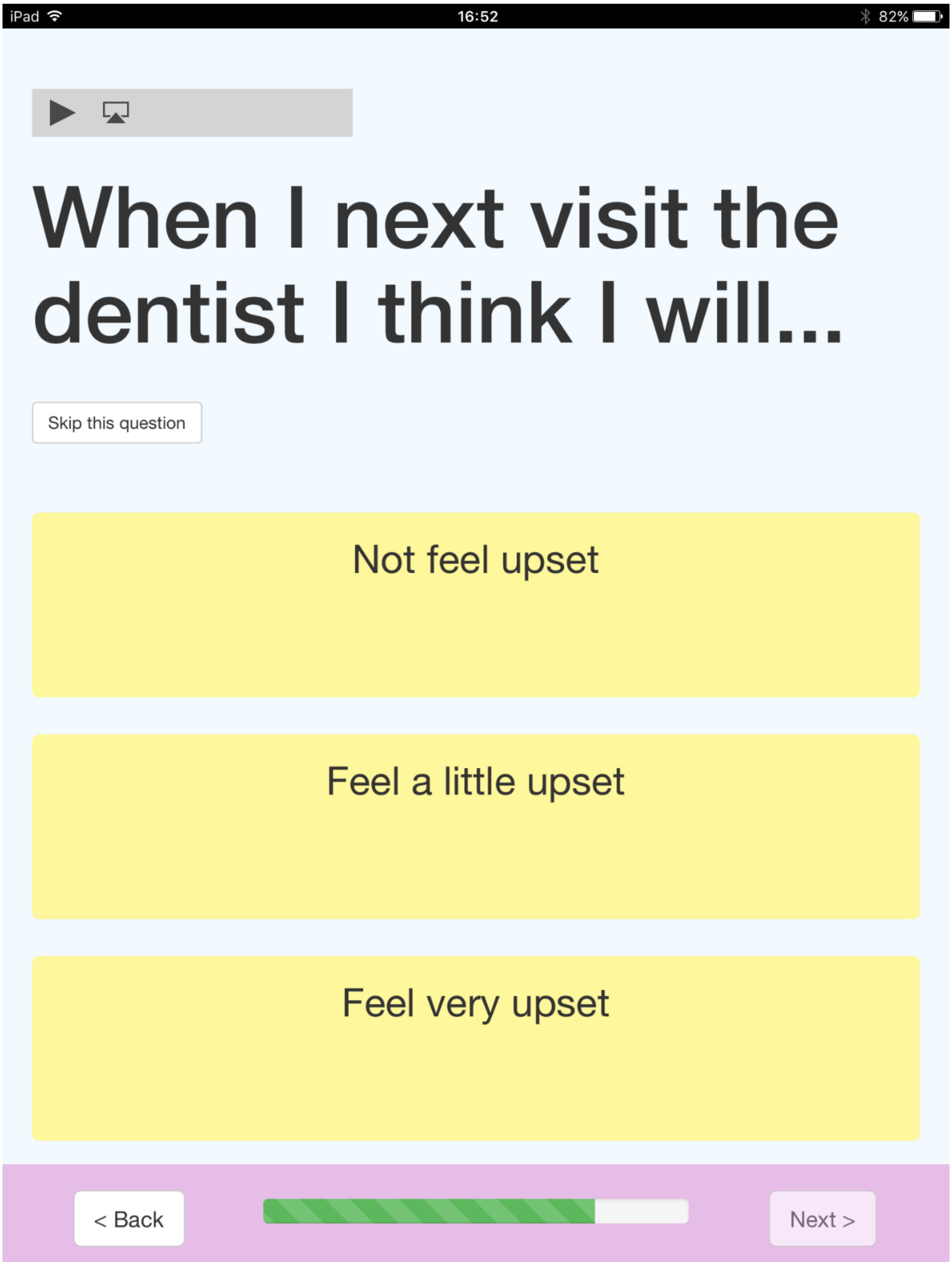
iPad
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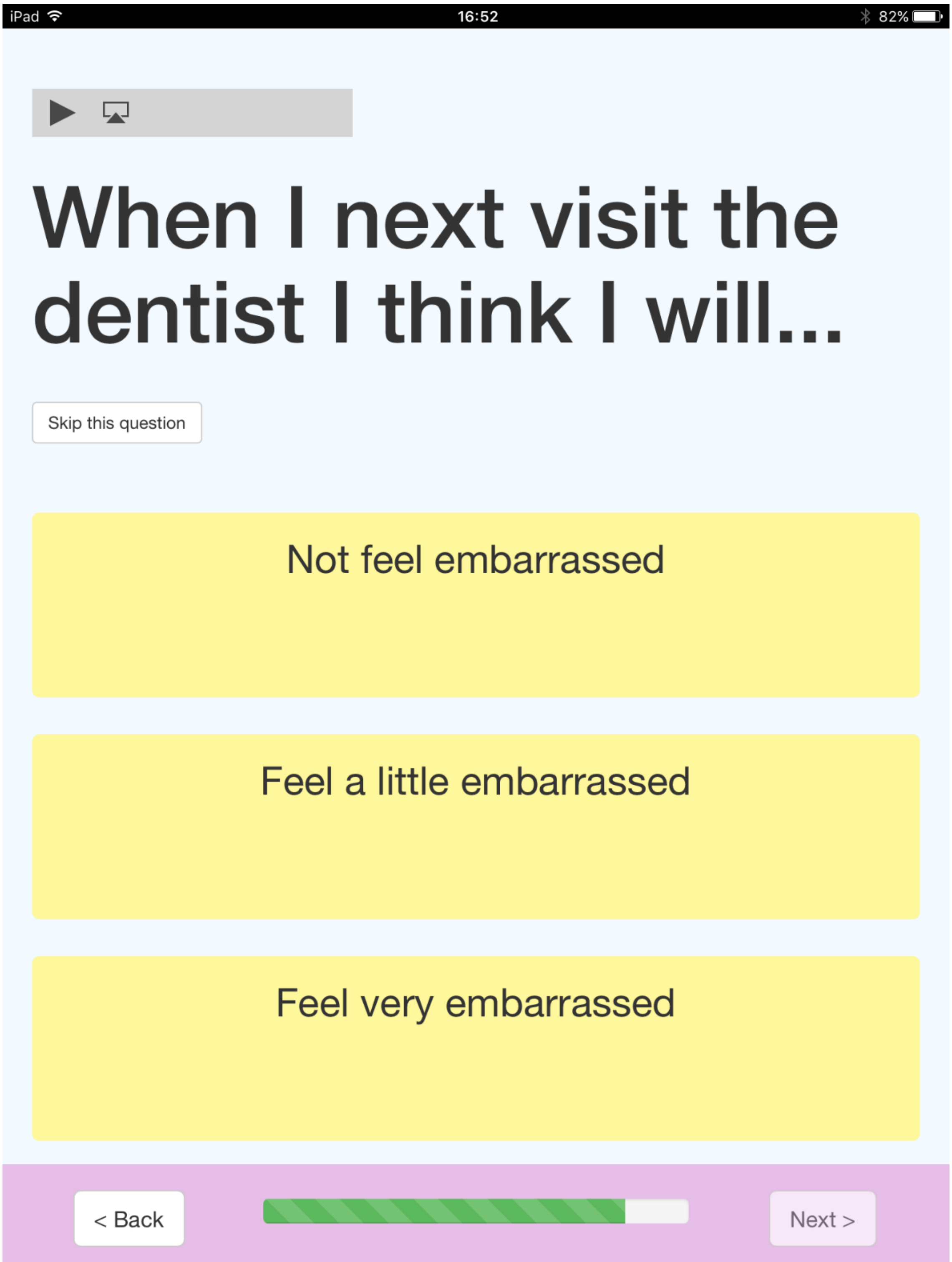
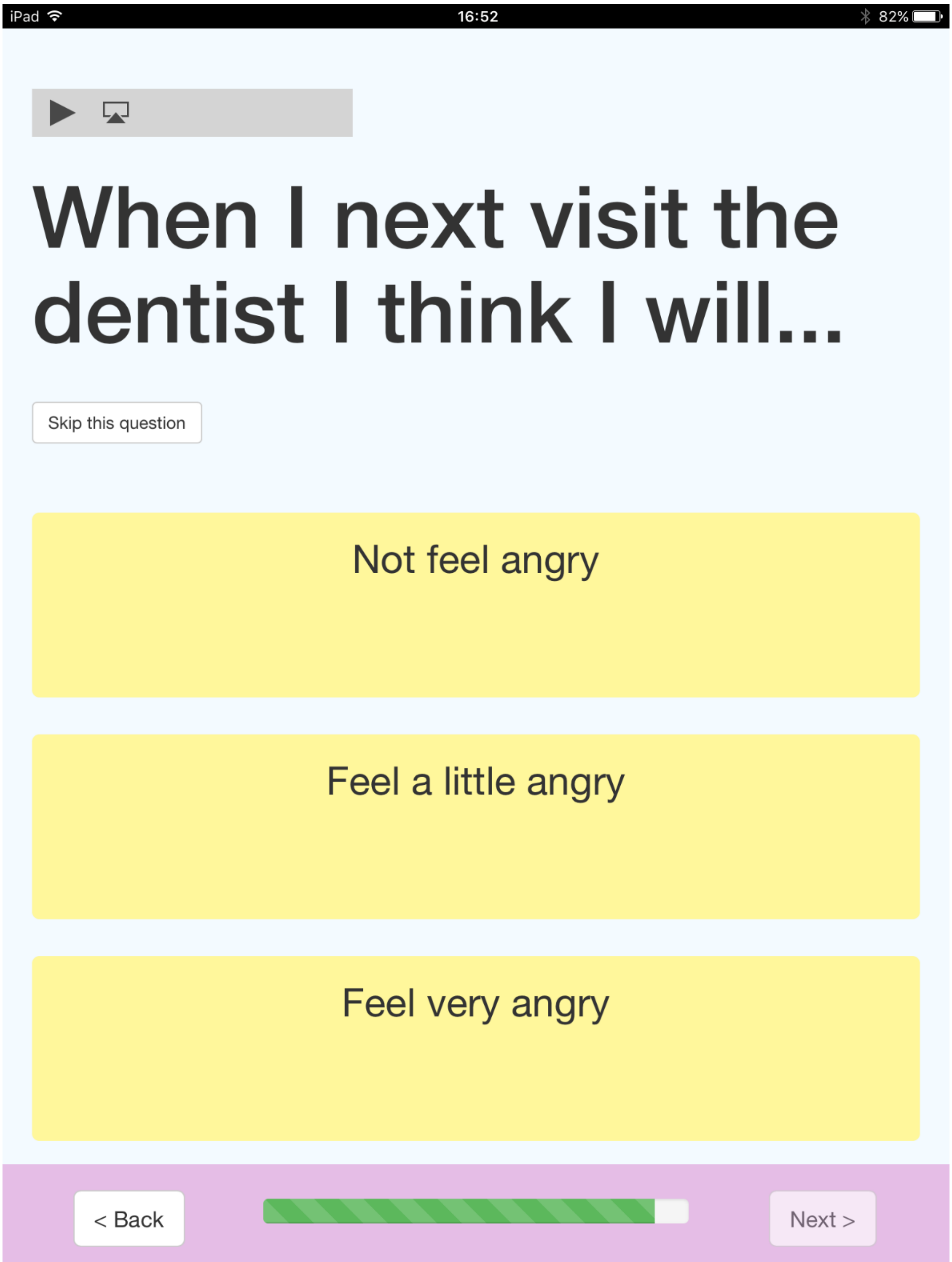
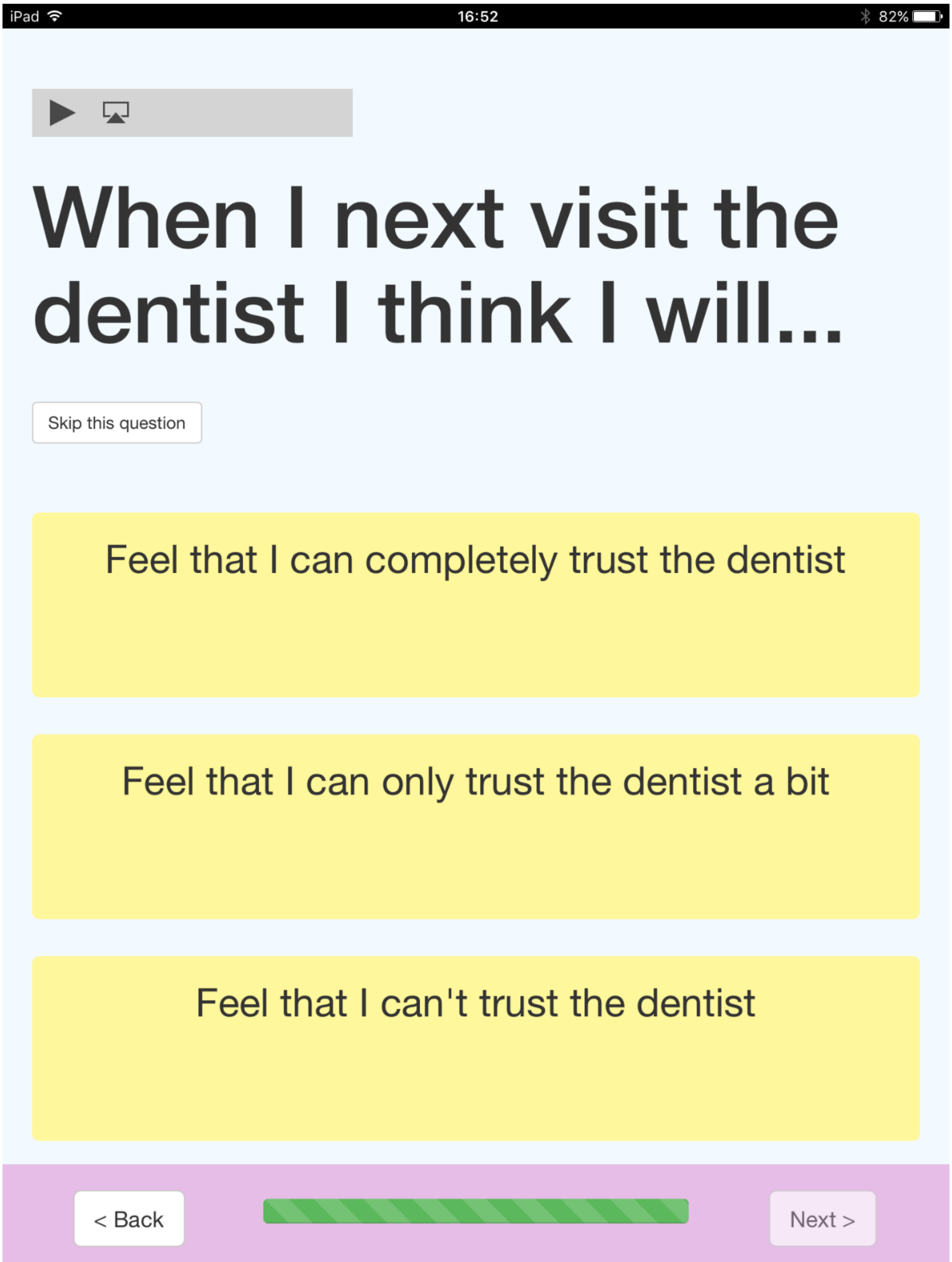
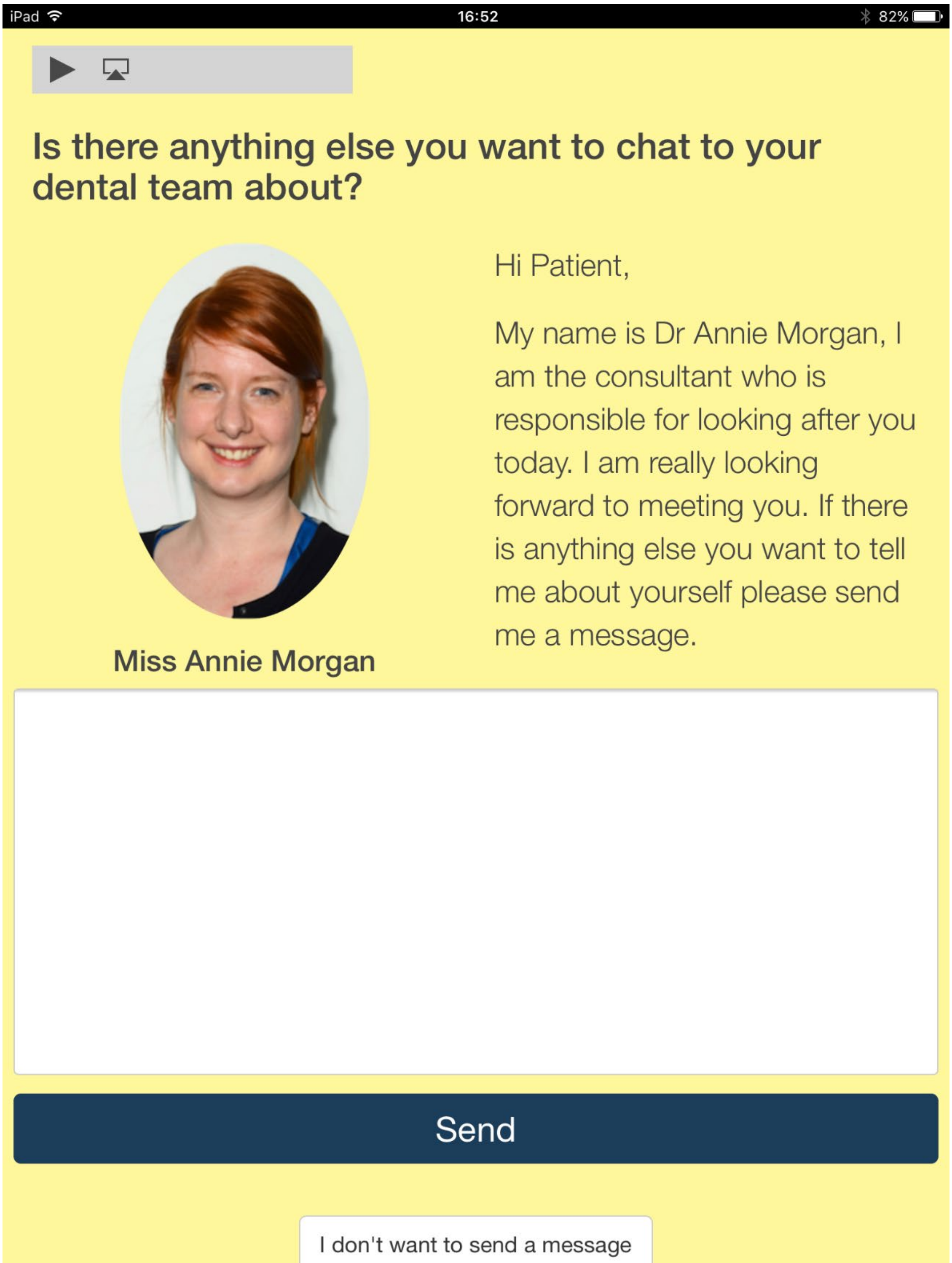
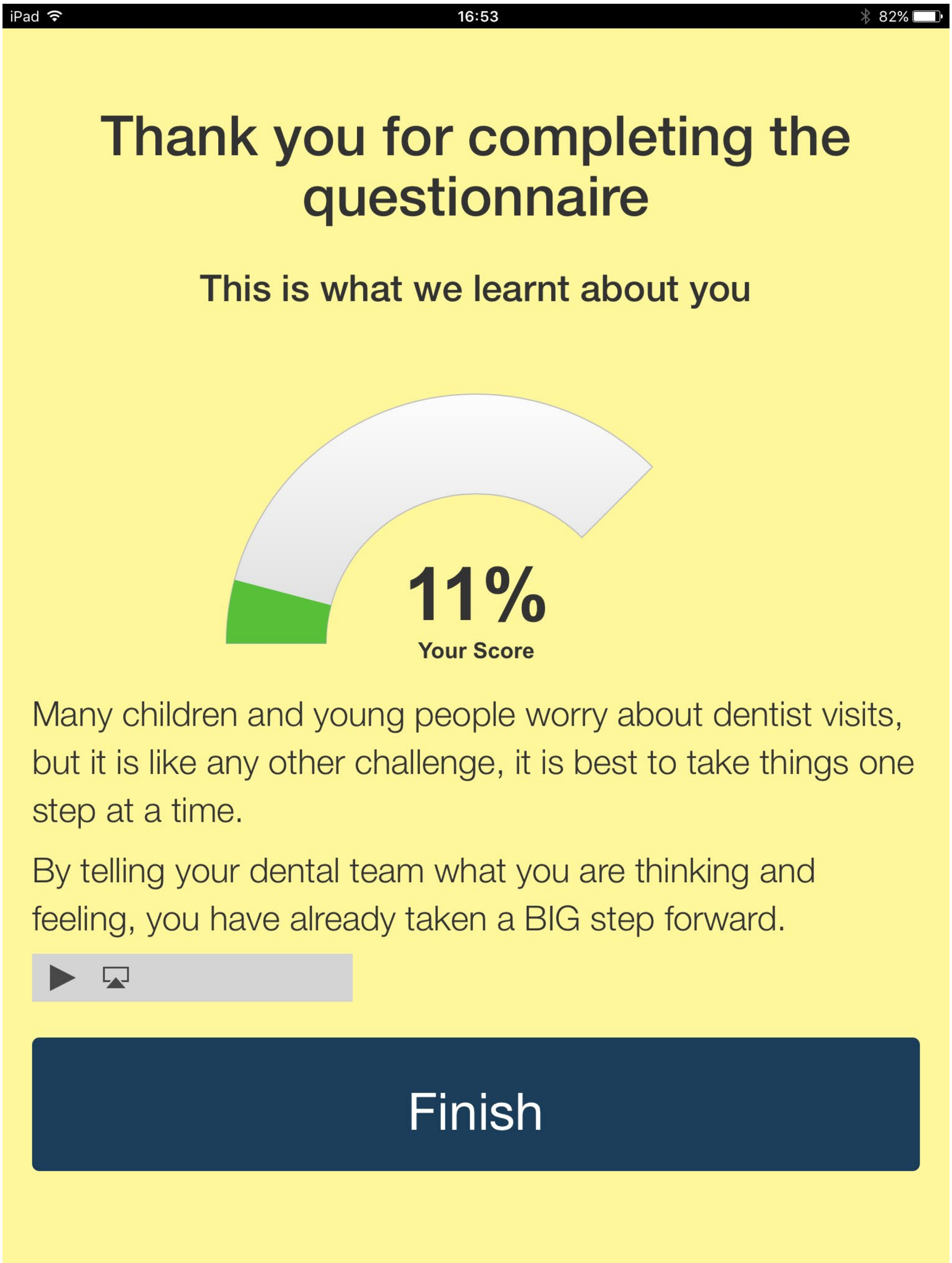
82%
When I next visit the dentist I think I will...
Skip this question
Not feel upset
Feel a little upset
Feel very upset
< Back
Next >


iPad
16:52
82%
When I next visit the dentist I think I will...
Skip this question
Not feel embarrassed
Feel a little embarrassed
Feel very embarrassed
< Back
Next >


iPad
16:52
82%
When I next visit the dentist I think I will...
Skip this question
Not feel angry
Feel a little angry
Feel very angry
< Back
Next >


iPad
16:52
82%
When I next visit the dentist I think I will...
Skip this question
Feel that I can completely trust the dentist
Feel that I can only trust the dentist a bit
Feel that I can't trust the dentist
< Back
Next >


iPad
16:52
82%
Is there anything else you want to chat to your dental team about?
Miss Annie Morgan
Hi Patient,
My name is Dr Annie Morgan, I am the consultant who is responsible for looking after you today. I am really looking forward to meeting you. If there is anything else you want to tell me about yourself please send me a message.
Send
I don't want to send a message


iPad
16:53
82%
Thank you for completing the questionnaire
This is what we learnt about you
11%
Your Score
Many children and young people worry about dentist visits, but it is like any other challenge, it is best to take things one step at a time.
By telling your dental team what you are thinking and feeling, you have already taken a BIG step forward.
Finish

iPad 16:53 82%

The University Of Sheffield.

Acknowledgements

Thank you to all the children, young people and families who contributed to this study. You were brilliant.

By Sarah Baker, Cathy Creswell, Zoe Marshman, Annie Morgan, Tim Newton, Jenny Porritt, Helen Rodd, Chris Stokes, Ashley Towers, and Chris Williams

Appendix 7. Participant questionnaire booklet for Study 3

Development of a web-based version of the Children's Experience of Dental Anxiety Measure for clinical assessment/phase 3/participant questionnaire booklet/v1
06/01/2016

Recruitment number: _______

Recruitment date: __ __/__ __/ 2016

Children's Experience of Dental Anxiety Measure
Participant Questionnaire Booklet

Thank you for agreeing to take part in our study.

In this booklet you will firstly find some sets of questions for us to learn about you. We are then going to ask you to fill in two different versions of the same questionnaire about how you feel about going to the dentist, and then, finally, what you thought about those questionnaires.

Don't worry; there are no right answers and no wrong answers to any of the questions, and someone will be there to help you if you need it.

We would be very grateful if you could answer all the questions using the instructions.

Section 1

Firstly, we would like to learn about you.

1) Are you a boy or a girl? (please tick)

I am a boy.................................. ☐

I am a girl .... ☐

2) How old are you?

I am ....years

3) What is your home postcode ☐☐☐☐ ☐☐☐

Please ask your parents/carers for help if you need it.

Section 2

Now, we are going to ask you to fill in two different types of the same questionnaire about how you feel about going to the dentist. Your research team member is going to tell you if you are going to do the questionnaire in this booklet first, or on a tablet computer first. After a quick 15 minutes break, we are going to get you to swap and fill it in again on the type you didn't use first time.

Please turn over for the next part of the questionnaire

Please turn over for the next part of the questionnaire

For each question please read all the choices and then decide which one is most like you **today.** There are no right and no wrong answers to any of the questions.

When I know I have an appointment with the dentist	
(please tick one box)	
☐	I will do nothing to avoid going
☐	I will do some things to avoid going
☐	I will do everything to avoid going

When I know I have an appointment with the dentist	
(please tick one box)	
☐	I will tell my parents I don't mind going
☐	I will tell my parents I would rather not go
☐	I will tell my parents I really don't want to go

When I next visit the dentist	
(please tick one box)	
☐	I will let the dentist look in my mouth
☐	I will try to stop the dentist looking in my mouth a bit
☐	I will not let the dentist look in my mouth

When I next visit the dentist	
(please tick one box)	
☐	I will not get worried if the dentist tells me I need to have something done
☐	I will get a little worried if the dentist tells me I need to have something done
☐	I will get really worried if the dentist tells me I need to have something done

Please turn over for the next part of the questionnaire

When I next visit the dentist	
(please tick one box)	
☐	If I asked the dentist to stop what they were doing they would definitely stop
☐	If I asked the dentist to stop what they were doing they might stop
☐	If I asked the dentist to stop what they were doing they would not stop

When I next visit the dentist I think	
(please tick one box)	
☐	I will not be worried that it will be painful
☐	I will be a little worried that it will be painful
☐	I will be very worried that it will be painful

When I next visit the dentist I think	
(please tick one box)	
☐	Nothing will go wrong
☐	Something will go a bit wrong
☐	Something will go very wrong

When I next visit the dentist I think	
(please tick one box)	
☐	I will have a lot control over what happens in the appointment
☐	I will have a bit of control over what happens in the appointment
☐	I will not have any control over what happens in the appointment

When I next visit the dentist I think I will	
(please tick one box)	
☐	Not feel shaky
☐	Feel a little shaky
☐	Feel very shaky

Please turn over for the next part of the questionnaire

When I next visit the dentist I think I will	
(please tick one box)	
☐	Not feel stressed
☐	Feel a little stressed
☐	Feel very stressed

When I next visit the dentist I think I will	
(please tick one box)	
☐	Not feel upset
☐	Feel a little upset
☐	Feel very upset

When I next visit the dentist I think I will	
(please tick one box)	
☐	Not feel embarrassed
☐	Feel a little embarrassed
☐	Feel very embarrassed

When I next visit the dentist I think I will	
(please tick one box)	
☐	Not feel angry
☐	Feel a little angry
☐	Feel very angry

When I next visit the dentist I think I will	
(please tick one box)	
☐	Feel that I can completely trust the dentist
☐	Feel that I can only trust the dentist a bit
☐	Feel that I can't trust the dentist

Please turn over for the next part of the questionnaire

Section 3

We would like you to think about the two types of questionnaires you filled in today about how you feel about a dental visit. We want to learn what you thought about the two questionnaires, and if you thought one type was better than the other one.

Please read the sentences about each and then tick the box that best matches what you think.

"I think the paper questionnaire where I write my answers is good for children and young people"

Please tick the box you agree with the most

I agree a lot	I agree a bit	I disagree a bit	I disagree a lot	I don't know
☐	☐	☐	☐	☐

"I think that the questionnaire on a tablet computer is good for children and young people"

Please tick the box you agree with the most

I agree a lot	I agree a bit	I disagree a bit	I disagree a lot	I don't know
☐	☐	☐	☐	☐

And did you think one type of questionnaire was better for children and young people than the other?

Please tick one box

☐ I would chose the paper questionnaire

☐ I would chose the questionnaire on a tablet computer

☐ I don't have a type of questionnaire I prefer

Please turn over for the next part of the questionnaire

Section 4

This part is about the questionnaire on the tablet computer. Please tick the box that best matches what you think about filling in the questionnaire on the tablet computer. Remember there are no right answers and no wrong answers.

Please tick the box that best matches what you think

	Strongly disagree	Disagree a bit	Neither disagree or agree	Agree a bit	Strongly agree
	1	2	3	4	5
I think that I would be happy to fill in the questionnaire on the tablet again					
I found the questionnaire on the tablet too difficult					
I thought the questionnaire on the tablet was easy to use					
I think I would need someone to help me to use the questionnaire on the tablet					
I found the different parts of the questionnaire on the tablet all worked well together					
I thought there was too many problems with the questionnaire on the tablet					
I think that most people would learn to use this questionnaire on the tablet very quickly					
I found the questionnaire on the tablet tricky to use					
I felt very confident using the questionnaire on the tablet					
I needed to learn a lot of things before I could get going with this questionnaire on the tablet					

Overall, I would rate the user-friendliness of this product as:
(Please circle your answer)

The worst	Very poor	Poor	Okay	Good	Excellent	The best

Please turn over for the next part of the questionnaire

Section 5

Please tick the box that best describes your ethnic group. You only need to tick one box on this page. Ask your parent or carer to help you with this section if you need to.

A: White

- ☐ English/Welsh/Scottish/Northern Irish/British
- ☐ Irish
- ☐ Gypsy or Traveller
- ☐ Any other White background, write in below

B: Mixed/multiple ethnic groups

- ☐ White and Black Caribbean
- ☐ White and Black African
- ☐ White and Asian
- ☐ Any other Mixed/multiple ethnic background, write in below

C: Asian/Asian British

- ☐ Indian
- ☐ Pakistani
- ☐ Bangladeshi
- ☐ Chinese
- ☐ Any other Asian Background, write in below

D: Black/African/Caribbean/Black British

- ☐ African
- ☐ Caribbean
- ☐ Any other Black/African/Caribbean background, write in below

E: Other ethnic group

- ☐ Arab
- ☐ Any other ethnic group, write in below

Thank you for helping us today