Dental Anxiety in Adults
Psychosocial Aspects, Oral Health and Psychological Treatment

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UNIVERSITY OF GOTHENBURG
Gothenburg 2015
To Heidi, Åke and Sofia
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ABSTRACT

The aim of this thesis was to investigate relationships between dental anxiety (DA), psychosocial factors, oral health and oral health-related quality of life (OHRQoL) in adults, and to assess the efficacy of cognitive behavior therapy (CBT) as treatment for DA. Study I is a national Swedish survey, studies II and III are clinical cross-sectional studies on patients with severe DA, and study IV consists of a systematic review and a meta-analysis. In the first study, a weak relationship was found in the general population between Antonovsky’s salutogenic concept of sense of coherence (SOC) and DA. Strong relationships were found between high DA and low OHRQoL, irregular dental care and smoking. In study II, a high prevalence of attention deficit hyperactivity disorder (ADHD), as indicated by a screening questionnaire, was found in patients with severe DA. ADHD was further associated with a higher level of DA and lower self-rated/subjective oral health. There were also indications of poorer objective oral health associated with ADHD, although these relationships were not statistically significant. In study III, the results showed a substantial dissatisfaction with orofacial esthetics in patients with severe DA. Ratings of orofacial esthetics were related to both psychological and oral health. Furthermore, the results confirmed the validity of the Orofacial Esthetic Scale (OES), when used on patients with severe DA. The systematic review in study IV included randomized controlled trials evaluating CBT for severe DA, and identified ten scientific publications originating from seven different RCTs. Some support was found for CBT resulting in a clinically significant reduction in DA immediately after treatment and at follow-up after one and two years. Also, some support was found for CBT enhancing the acceptance of conventional dental treatment. These results were all based on a low quality of evidence according to the GRADE (Grading of Recommendations Assessment, Development, and Evaluation) system. In conclusion, DA is an important condition associated with avoidance of dental care and health-related outcomes. Dissatisfaction with orofacial esthetics and ADHD both seem to be associated with DA and in the general population a weak relationship between the SOC and DA was found. There are effective psychological methods that can reduce or eradicate DA in adult individuals, although there are substantial knowledge gaps to be investigated.

Keywords: adults, attention deficit hyperactivity disorder, cognitive behavior therapy, dental anxiety, esthetics, oral health, oral health-related quality of life, psychological health, psychological treatment.

ISBN: 978-91-628-9392-7 (printed)
SAMMANFATTNING PÅ SVENSKA

Tandvårdsrädsla är ett vanligt förekommande problem som kan leda till att individen skjuter upp eller helt undviker tandvård, vilket i sin tur kan få allvarliga konsekvenser för munhälsan. Grav tandvårdsrädsla har även visat sig ha samband med psykosociala faktorer som social isolering och psykisk ohälsa.


This thesis is based on the following studies, referred to in the text by their Roman numerals.


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<tr>
<td>ADHD</td>
<td>Attention Deficit Hyperactivity Disorder</td>
</tr>
<tr>
<td>ASRS</td>
<td>The World Health Organization Adult ADHD Self-Report Screener</td>
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<td>BT</td>
<td>Behavior Therapy</td>
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<td>CBT</td>
<td>Cognitive Behavior Therapy</td>
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<td>DA</td>
<td>Dental Anxiety</td>
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<td>Dental Fear Survey</td>
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<td>DMFT</td>
<td>Decayed, Missing and Filled Teeth</td>
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<td>GRADE</td>
<td>Grading of Recommendations Assessment, Development, and Evaluation</td>
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<td>HADS</td>
<td>Hospital Anxiety and Depression Scale</td>
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<td>HADS-A</td>
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<td>HADS-D</td>
<td>Hospital Anxiety and Depression Scale—Depression Subscale</td>
</tr>
<tr>
<td>OES</td>
<td>Orofacial Esthetic Scale</td>
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<td>OHIP</td>
<td>Oral Health Impact Profile</td>
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<td>OHIP-5</td>
<td>Oral Health Impact Profile, five-item version</td>
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<tr>
<td>OHRQoL</td>
<td>Oral Health-Related Quality of Life</td>
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<td>SCB</td>
<td>Statistics Sweden</td>
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</table>
SOC       Sense of Coherence
SOC-3     Concept of Sense of Coherence, three-item scale
INTRODUCTION

In popular culture, dental treatment is often associated with physical discomfort and psychological distress. This relationship is frequently exploited in the media and may be used as scary examples of dental treatment or in comic interpretations. In science, psychological distress associated with dental treatment, or the anticipation of dental treatment, has been a subject of inquiry for over 90 years [1]. The first article indexed in Medline that used the term dental anxiety (DA) was published in 1946. In the article, Coriat described DA as “an excessive dread of anything being done to the teeth,” and states that “one of the most widely spread forms of fear in everyday life is the fear of going to the dentist” (p 365) [2]. The first epidemiological surveys of DA were performed in the 50s and early 60s, [3, 4] and these, together with more contemporary research, have proven Coriat right in that DA is a commonly occurring phenomenon. Today, it is also known that high DA is associated with poor oral health and a range of psychosocial consequences, marking it as an important health-related condition worthy of further investigation.

Several terms have been used in scientific language to describe psychological distress related to dentistry. Although the terms focus on somewhat different aspects, they are often used interchangeably. The most common terms that will be explained here are: DA, dental fear and dental phobia. After an initial description of these terms, DA will be used consistently in this thesis as an overarching term for both DA and dental fear.

Anxiety, fear and phobia

Fear is often described as the evolutionarily inherited emotional response to a dangerous or life-threatening stimulus [5]. An emotional response is a set of characteristics including a subjective emotional experience (e.g., to feel afraid), action tendencies directed towards survival of the individual or species (e.g., escape), and a means of communication with other individuals (e.g., that something dangerous is present). Fear is mostly characterized by our fight-or-flight response, which is our basic tendency to escape dangerous situations, or, if under attack and escape is not possible, to defend ourselves. Even though flight is the primary tendency for most individuals, fight may become the predominant response under certain learning conditions. Apart from the fight-or-flight response, fear can also be associated with a freezing
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response, also called tonic immobility [5]. Dental fear is the description of fear elicited by dental treatment.

The term anxiety is more difficult to define than fear and is more oriented towards cognitive processes regarding an anticipated future threat or aversive situation [5], which, in DA, is related to dentistry. These processes often involve strong physiological activation. In anxiety, low perceived control over future, potentially aversive events has been described as a crucial factor [6], and Barlow states that “anxiety could be characterized roughly as a state of helplessness, because of a perceived inability to predict, control, or obtain desired results or outcomes in certain upcoming personally salient situations or contexts” (p. 1249) [5]. In line with this, anxiety associated with dentistry has been linked to perceptions of dental care as uncontrollable and unpredictable [7].

Lautch defined dental phobia in the early 70s as “a special kind of fear out of proportion to the demands of the situation, which will not respond to reason, is apparently beyond voluntary control and leads to avoidance of dental treatment where this is really necessary” [8] (p. 151). Today, dental phobia is a psychiatric condition diagnosed as a type of specific phobia according to diagnostic manuals [9, 10]. In the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) [9], a specific phobia is described as a marked anxiety or fear, that is out of proportion and “actively avoided or endured with intense fear or anxiety” [9] (p. 197). The anxiety, fear or avoidance should also be persistent and lead to substantial distress or impairment. Dental phobia is a part of the blood-injection-injury subgroup of specific phobias, which also include injection phobia and phobias related to other medical procedures.

Prevalence

The prevalence of DA has been investigated in the general public and specific populations, such as university students and patients in dental care [11]. Estimates of the prevalence of DA have varied substantially between studies. In a comparison of studies that used the same instrument [the Dental Anxiety Scale (DAS)], Raadal and Skaret found estimates of prevalence ranging from about four to 30 per cent [12]. Differences in the populations studied and cutoff levels may explain these observed differences. Despite substantial variation, several reports of the prevalence of DA in the general population give estimates of about 20 % [13-15] and about five per cent for severe DA [16-18]. A common finding in prevalence studies is that DA is more common in women than in men [19]. In children and adolescents,
Klingberg and Broberg estimated the prevalence of DA to nine per cent in their review of the literature [20].

Dental anxiety in adults is generally measured using self-report questionnaires. The questionnaires range from one-item scales to measures that consist of 20 items or more. Typical questions ask the participant to rate the anticipated level of anxiety, fear or particular behavior that may be elicited by situations or stimuli associated with dental care. Existing scales differ in their conceptual models and capture the phenomena of DA in different ways [21, 22]. The Dental Anxiety Scale (DAS) [23] is the most commonly used measure to assess DA in adults [24]. This four-item self-report scale asks the participant to rate the anticipated level of tension or anxiety the day before a visit to the dentist, while sitting in the dentist’s waiting room, when the dentist is about to start drilling, and when the dentist is about to remove calculus. In the Modified Dental Anxiety Scale (MDAS), a fifth question concerning dental injections has been added to the original scale, as well as a change to the response options to make them more uniform [25]. Another often used scale that covers several different types of reactions and situations is the Dental Fear Survey (DFS) [26], which consists of 20 items. Apart from self-report data, aspects of DA in adults have also been measured by observing the behavior of the patient. In the treatment situation, the dentist may rate the treatability of the patient [27], and data on the regularity of dental care visits may indicate the level of avoidance [28]. Psychophysiological measures, mainly heart rate and muscle tension, are also sometimes used to measure the physiological aspects of DA [29].

Following a decline in dental caries and improved pain reduction methods during the 20th century [1], the question has been raised whether this is related to a decrease in DA. Smith and Heaton [11] concluded in 2003, in their review of studies made in the United States on college students and general population samples, that there was no clear evidence of changes in the prevalence of DA during the past 50 years. A difficulty when comparing studies over time is the variation between studies of measures and populations. However, there are some indications that the prevalence of DA is decreasing. National surveys in the United Kingdom, with two questions assessing DA, found a substantial decrease in DA between 1988 and 1998, but also a constant prevalence of severe DA at about ten per cent in the adult population during this time period [30]. A recent study from Norway found a small and non-significant decrease (from 17.5 to 16.7 %) in the prevalence of DA in 25-year-olds between 1997 and 2007 [31]. When adjusting for gender, education and regularity of dental care, the odds ratio for having DA in 1997 was 1.4 as compared to 2007.
In prospective studies, the prevalence of DA has been found to increase in adolescence and early adulthood [32], and to decrease in the middle-aged and elderly [33]. Cross-sectional studies have also found that older adults have a lower degree of DA [16, 34, 35]. The prevalence of dental phobia has been investigated in a few studies originating in Sweden [36], USA [37] and the Netherlands [14], with estimates ranging from 2.1 to 3.7 %, showing that dental phobia is common compared with other phobias in the general population [14].

**Etiology and psychosocial aspects**

Why is it that some people are immensely afraid of going to the dentist whereas others seem not to care at all? This question has been pondered by clinicians, researchers and, of course, by patients. As with several other forms of anxiety and phobia, most adults with DA report that their DA started in childhood, although a substantial number reports its onset in adolescence or adulthood [38-40]. In a study of adults in the general population, Locker et al. reported that among dentally anxious individuals, 50.9 % stated that their DA started in childhood, while 22.0 % stated adolescence and 27.1 % adulthood [39]. In comparison, Berggren and Meynert found, in a clinical sample of adult patients referred or self-referred to a specialized DA clinic, that 84.3 % reported that their DA started before the age of 16, indicating an earlier onset in clinical samples [40]. A prospective general population study showed an increasing prevalence of DA during adolescence and early adulthood, but also that a high degree of remission occurred during this period, indicating that it is a period of instability regarding DA [32].

Barlow and colleagues have postulated the theory of “triple vulnerabilities” in the development of anxiety [5]. In this theory, they point to the interplay between generalized and specific vulnerabilities in the development of anxiety. A generalized biological vulnerability consists of genetic components related to the development of anxiety. A generalized psychological vulnerability seems to come from specific childhood characteristics that lead to a diminished sense of control. Generalized biological and psychological vulnerabilities have been related to an increased risk of developing anxiety in general. Specific vulnerabilities are psychological in their nature, in that they consist of early experiences where a person learns from caregivers that something is dangerous. This may be expressed as modeling of the behavior of caregivers or by transmission of information. Generalized and specific vulnerabilities interplay and may be sufficient in themselves to develop strong anxiety or may contribute to the
effect of a traumatic event. If, for example, someone is stuck in an elevator, the existence of generalized vulnerabilities might increase the risk of developing claustrophobia.

Dental anxiety has also been found to be multifactorial in its etiology [39, 41-43]. Several authors have divided etiological factors of DA into the categories of endogenous and exogenous [39, 42, 44]. Endogenous refers to internal personal factors or vulnerabilities, such as personality and general fearfulness, while exogenous refers to external events or circumstances, such as trauma or vicarious learning which includes modeling the behavior of others.

Regarding exogenous factors, a substantial amount of research in adults has reported an association between DA and previous aversive experiences during dental treatment. These experiences may involve pain, being frightened, experiencing nausea, and extreme helplessness or embarrassment, as well as experiences of the dentist being heavy-handed, cold, providing too little information, rude or uncaring. [39, 40, 45-47]. A literature review regarding children also found an association between restraining and punishing behavior of the dental staff and fearful behavior of the child [48], and there appears to be a general relationship between invasive dental treatment and DA [43, 49]. Another risk factor for DA in children is having dentally anxious parents [39, 50, 51], suggesting that vicarious learning may lead to a specific vulnerability for DA in line with Barlow’s theory of triple vulnerabilities. Some results also point to sexual assault and abuse as possible risk factors for DA [46, 52], indicating a role for trauma outside of the dental care environment. However, many other potentially traumatic events have failed to show an association with DA [46].

Personality factors and psychological disorders have also been linked to DA, mainly in cross-sectional studies, although some prospective data indicate that predispositions at an earlier age may influence the trajectory of DA later in life [53, 54]. In a prospective study of middle-aged and elderly women, with measures taken in 1968-69 and again in 1992-93, Hägglin et al. [53] found that a high level of the personality trait of neuroticism and a low level of the personality trait of extraversion in 1968-69 were associated with a chronic trajectory of DA. This finding is also supported by cross-sectional studies linking DA especially to high neuroticism but also to low extraversion [8, 55]. Also, while a possible link between DA and genetic factors have been suggested [56, 57], a part of this link may be due to a genetic factor shared by DA and neuroticism [57]. Neuroticism is associated with a general tendency towards anxiety and anxiety disorders [5], and points
to a generalized vulnerability in the development of anxiety and fears in line with Barlow’s theory [5]. In further support of generalized vulnerability also affecting DA, both cross-sectional and prospective studies on samples drawn from the general population have found an association between DA and other fears as well as psychological disorders [43, 53, 54, 58], and point to a gradient where a higher level of DA is associated with an increasing number of psychological disorders [54]. Clinical studies in patients with severe DA also show that additional fears seem to be the rule [59], and the general symptom level of anxiety is reported to be high [60]. Additional psychological disorders, especially anxiety disorders, also seem to be common in patients with dental phobia or severe DA [61, 62].

An association between DA and certain characteristics of attention deficit hyperactivity disorder (ADHD) has been found in children [63]. ADHD is a psychiatric condition [9, 10], characterized by the two symptom clusters of inattention and impulsivity/hyperactivity. At first, ADHD was identified as a disorder in children but is now also known to be a common, although underdiagnosed, disorder in adults [64, 65], with a prevalence of 2-3 % in the general adult population [66]. In clinical populations, however, the occurrence of ADHD is estimated to be considerably higher, as seen by a prevalence rate of 27.9 % in a psychiatric sample of adult patients with anxiety disorders [67]. A strong association between ADHD and anxiety has also been established [64, 68]. Blomqvist et al. [63] found an association between ADHD with many symptoms of impulsivity/hyperactivity and DA in children. In adults, there is a general lack of research into dental implications of ADHD [69], and a possible relationship between ADHD, DA and oral health in adults is unknown.

Another psychosocial concept that has been associated with DA is the sense of coherence (SOC) [70-72]. In Antonovsky’s salutogenic theory, which focused on factors associated with good health instead of risk factors for disease, the SOC was central [73]. The SOC concept evolved from studies on individuals who had maintained good health despite histories of severe trauma [73]. Antonovsky described three internalized psychosocial dimensions; comprehensibility, manageability and meaningfulness, and together, these three components make up the SOC. The three dimensions are shaped by our upbringing and environmental factors, from childhood to the mid-twenties, and seem to be related to how we cope with stress both in everyday life and in extreme situations. According to the theory, individuals with a strong SOC are less prone to develop stress and better suited to deal with stress once it has occurred. The SOC has been strongly associated with perceived health and with mental health in particular [74]. This has called for
the argument that SOC may be equal to mental health; however, in their literature review, Eriksson and Lindström arrive at the conclusion that although the SOC is close to mental health and well-being, it is not the same thing [74]. A few studies have investigated the relationship between SOC and DA in adults [70-72]. In these studies, the presence of DA has been associated with a weak SOC. It may be that a strong SOC is a protective factor against developing DA, explaining why a weak SOC has been associated with DA. A clearer understanding of the relationship between SOC and DA may lead to important insights into the nature of DA. Therefore, the relationship between SOC and DA needs to be investigated further. A good place to start may be to investigate this association in a larger general population sample.

Apart from the above-mentioned psychosocial aspects, DA seems to be associated with an additional range of psychosocial problems [75-77]. Patients with severe DA have reported effects on relationships, social and working life because of DA [60, 77, 78]. Longer avoidance of dental care have also been associated with an increase in negative social effects in this patient group [77]. Patients with severe DA have also reported feelings of anger, shame and depression because of their DA [60]. After treatment at a specialized DA clinic, patients have reported significant changes in the frequency of tension headache and stomach problems, and reduced abuse of alcohol [28]. Successful treatment at a specialized DA clinic has also been associated with a decreased number of sick days [28, 79]. Also, in a sample from the general population, Locker reported that 93.1% of individuals with DA reported at least one psychosocial consequence [76]. Eighty-four per cent reported psychological reactions, such as worry about one’s teeth or feeling foolish, 60.4% reported problems regarding social relationships, such as a need to hide their DA from others, and 21.2% reported that they hide their teeth when they laugh or smile. The burden of psychosocial problems seems to be relative to the number of other fears in patients with severe DA and in the general population [75, 76]. There are some mixed results, but on the whole, there appears to be a relationship between DA and low socioeconomic status [80-84]. In general, a relationship between low socioeconomic status and poor psychological health, as well as other health-related outcomes, is a common finding [85, 86]. The reasons for this kind of relationship are probably complex, although low socioeconomic status is associated with less access to resources and generally fewer possibilities to influence one’s life situation, which may result in increased levels of stress.
Oral health–related aspects

Dental anxiety has been found to be related to deterioration in oral health and to avoidance of dental care [15, 40]. There is support of general relationship between avoidance of dental care and an increased self-perceived oral treatment need and DA seems to amplify this process [87]. Indications of poorer oral hygiene behaviors [88, 89] and a substantially higher prevalence of smoking [90, 91] have also been found in adults with DA, indicating that other factors than avoidance may also contribute to poor oral health in this group.

Regarding avoidance of dental care, it is noteworthy that anxiety and fear in general are linked to different kinds of avoidant behavior [92]. The main survival value of anxiety and fear is often regarded to be the way in which they motivate us to avoid direct or expected danger. Also, in the diagnose of specific phobia, avoidance related to fear and anxiety is emphasized [9]. In research focused on DA, avoidance of dental care has been thoroughly investigated both in clinical and observational studies and revealed as one of DA’s most central features [29]. Avoidance of dental care has mainly been measured through self-report data with questions on time since last dental treatment and regularity of dental care. Clinical studies on patients referred or self-referred to specialized DA clinics have shown estimated avoidance times of 5 to 20 years, with higher levels of DA being associated with increased avoidance [29]. A gradient between the level of DA and avoidance of dental care has also been found in observational data for the general population [35].

However, it is important to note, when discussing the established association between DA and avoidance, that even among the most dentally anxious individuals, a substantial proportion do not avoid dental care. Some research has been carried out to try to identify factors associated with avoidance or non-avoidance in individuals with high DA [93, 94]. In a study by Armfield [93], factors such as smoking, tooth-brushing less than twice a day and anxiety about costs all predicted avoidance of dental care in dentally anxious individuals. The interpretation of these results is somewhat difficult, but it may be seen as unlikely that, for instance, smoking by itself causes individuals to avoid dental care. Rather, smoking is probably an indicator of other factors related to avoidance and may be a surrogate measure of health behavior in general.

Oral health and quality of life

As mentioned above, in addition to avoidance of dental care, DA has also shown a strong relationship with poor oral health. Oral health was defined by
the World Health Organization (WHO) in 2012 as “... a state of being free from mouth and facial pain, oral and throat cancer, oral infection and sores, periodontal (gum) disease, tooth decay, tooth loss, and other diseases and disorders that limit an individual’s capacity in biting, chewing, smiling, speaking, and psychosocial wellbeing” [95]. This definition reflects a high focus on disease in the definition of oral health; functional and psychosocial aspects are mentioned, but only as consequences of disease. Already in 1948, however, the WHO defined health in general as “a state of complete physical, mental and social well-being and not merely the absence of illness or infirmity” [96]. This definition points to psychosocial aspects of health and extends its definition beyond the absence of disease. Contemporary definitions of oral health that exhibit similar features also exist. The fact that “oral health” is an established term, instead of just using “health”, reflects the fact that odontology and medicine have evolved as separate disciplines [97].

In his influential 1988 paper, Locker adapts the WHO International Classification of Impairments, Disabilities and Handicaps model to the oral health context [98]. This conceptual framework builds on the effects and consequences of disease and gives a flow of events from disease to handicap or death (see Figure 1).

![Figure 1. Locker’s conceptual model of oral health [98].](image)

*Figure 1. Locker’s conceptual model of oral health [98].*

*Disease* specifies pathological processes, which, in oral health, may be caries or periodontitis. *Impairment* refers to physical (or psychological in some disorders) changes or abnormalities that may be present at birth or associated with disease and injury. In the case of oral health, this may, for instance, be seen as missing teeth. *Functional limitation* is the consequence of impairment on bodily function. In oral health, this may be the function of chewing. These three aspects taken together, disease, impairment and functional limitation,
capture the concept of disease and the biophysical consequences that arise from it. From there follow the more psychological and social, or sociodental—as some would label them—aspects of oral health. Discomfort implies subjective experiences of discomfort. Pain is a very common symptom of oral disorder and falls into this category. Disability originally included mainly restrictions and limitations in activity and social roles; however, psychological and social well-being may be influenced by disease to the same extent and are now also included in this concept. As an example, oral disorders may lead to an inability to communicate effectively, with effects on social roles and psychological well-being. Handicap refers to disadvantages experienced when one cannot live up to societal standards. Some oral disorders may also contribute to a handicap due to their esthetic implications, which may be related to social acceptance. It should be noted that in today’s Swedish language, the term handicap is less well regarded and the Swedish National Board of Health and Welfare recommends that the term should no longer be used [99], although in this thesis it will still be used in the context described above.

Starting with the disease and impairment parts of Locker’s model, clinical studies on patients with severe DA and avoidant behavior have found a high level of decayed surfaces/decayed teeth, missing teeth and periodontal disease in this group of patients [40, 60, 100, 101]. One study that included a control group found that patients with severe DA had more decayed surfaces, missing teeth and periodontitis, but fewer filled teeth than controls [101]. The occurrence of root remnants was investigated in two studies reporting that almost 40 [29, 100] and 57 % [60], respectively, of patients with severe DA had at least one root remnant. The existence of root remnants indicates longstanding avoidance of dental care, despite an obvious treatment need. Epidemiological studies have also found an association between DA and oral disease and impairment [83, 102-104], indicating that DA is associated with these aspects of oral health also in the general population.

As for functional limitations, discomfort, disability and handicap, there are some self-report data relating to DA, although nowadays, measures of these aspects are mainly included under the title of oral health-related quality of life (OHRQoL). The term OHRQoL emerged in dentistry following the introduction of “quality of life” or “health-related quality of life” in medicine. The concept of quality of life is not clearly defined, either in medicine or in odontology, and there is no clear distinction between the concepts of quality of life and health [105]. However, quality of life has mainly been associated with the psychosocial and functional aspects of illness and is usually intended to incorporate the perspective and values of the patient. However, what is
actually measured with quality of life measures varies, both in dentistry and
in medicine [105, 106]. One of the most widely used measures of OHRQoL,
the Oral Health Impact Profile (OHIP) [107], is based on Locker’s 1988
model of oral health and its items are designed to measure the effects of oral
disorder in the areas of functional limitation, physical pain, psychological
discomfort, physical, psychological and social disability, and handicap [107].
The 14-item version or the OHIP has been used in several studies to evaluate
the association between OHRQoL and DA [103, 108-112]. The results of
these studies show that DA is associated with a substantial decrease in
OHRQoL when measured in this way. These results, together with the
previously described results concerning disease and impairment, indicate that
DA is associated with a full range of health-related problems.

What is lacking when applying Locker’s 1988 model of oral health to quality
of life is the actual “quality” of the experience. However, the underlying
assumption is that if we know enough about the impact of illness on
important life domains, this should tell us something about how the quality of
our lives is affected by illness [106], although this assumption fails to
acknowledge important psychological and social factors. When individuals in
a large general population sample was asked if the condition of their teeth,
gingiva, mouth or false teeth reduced or added to their quality of life, 75 %
reported a link between quality of life and these aspects of oral health [113].
This result points to the existence of a strong link between oral health and
quality of life with a focus on the actual “quality”. The UK Oral Health
Quality of Life (OHQoL-UK) measure asks participants in a similar fashion
if aspects of their oral health influence a selection of important domains, and
if this, in turn, affect their quality of life in a positive or negative way [114].
The OHQoL-UK has been used to assess OHRQoL in relation to DA and
McGrath and Bedi reported a strong association between DA and poor
OHRQoL, also when measured in this way [115]. Despite the differences
between the quality of life measures, these different types of measures
provide us with valuable information; however, we need to know on what
premises they are based so that we know what kind of conclusions may be
drawn from them.

One area that may be affected by oral disorders and is associated with
OHRQoL is dental and orofacial esthetics [113, 116]. Several measures of
OHRQoL include questions related to appearance, and in the well-used OHIP
measure, appearance has been suggested as a separate dimension [117].
However, data show that the OHIP is insufficient as a comprehensive
measure of dental appearance [118]. This calls for research including more
direct measures of appearance to complement measures of disease and
OHRQoL, as appearance may be an important link between the two. The Orofacial Esthetic Scale (OES) [119] consists of questions about the subject’s feelings about the appearance of different parts of the orofacial region, which includes the teeth, gum, mouth and face. Since orofacial esthetics is operationalized as asking about appearance in the OES, the two terms esthetics and appearance will hereafter be used interchangeably. In patients with severe DA [40], as well as in the general population [102], research has provided some indications of an association between DA and dissatisfaction with dental appearance. Since orofacial appearance is related both to oral disorders and OHRQoL, it may have a central position in the negative consequences associated with DA [120]. There has also been some research into how the rating of dental and general appearance is associated with psychological factors; especially that it is negatively associated with depression [121-123]. A more comprehensive and direct investigation of orofacial esthetics and DA is warranted and since DA has been associated with increased level of psychological disorders [54] and other indications of poor psychological health [59, 60, 124], an investigation of orofacial esthetics and DA ought to include measures of psychological health.

The vicious circle of dental anxiety

In order to synthesize different findings regarding DA, oral health and psychosocial aspects in a model, Berggren, as well as other researchers, have proposed a vicious circle of DA [40, 87, 125]. The vicious circle shows how oral health and psychosocial aspects act together over time in the maintenance and elevation of DA and some of its consequences. The vicious circle has had a substantial impact on the research field and has been used as a conceptual model (see, for example [60, 87]). The first step in the vicious circle is avoidance of dental care. Avoidance is central to the many health-related and psychosocial consequences of DA, and individuals with high DA who do not avoid dental care seem to be in a better position concerning oral health and psychosocial aspects [87, 94]. The second step of the vicious circle is the deterioration of oral status that comes with avoidance of dental care. Armfield then argues that the deterioration of oral status may lead to invasive and acute treatments and makes dental visits for an existing problem the third step in the vicious circle [87, 126]. Berggren, on the other hand, focuses on psychosocial consequences and describes feelings of shame and inferiority caused by deterioration of oral status as the third step [127]. Both problem-triggered visits to the dentist and feelings of shame and inferiority are then assumed to increase the existing DA. Although the relationship between DA, avoidance of dental care and oral health has not been
thoroughly investigated until recent years, it has long been anticipated and is included in Coriat’s previously mentioned article from 1946 [2].

![Diagram of the vicious circle of DA]

As a means to validate the vicious circle model, Armfield investigated the patterns of avoidance of dental care, treatment need and seeking dental care for an existing problem and found that 38.5% of individuals with DA in a general population sample seemed to follow the course implied by the vicious circle [87]. Results from de Jongh et al. also indicate that poorer oral health mediate the relationship between avoidance of dental care and fear of being negatively evaluated by others, which further supports the model [125].

**Treatment of dental anxiety**

In reviewing the associations between DA, oral health and psychosocial aspects, DA is recognized as a health-related condition that may have serious implications for health and well-being. Effective ways to manage DA are therefore of significant importance. Beginning in the early 70s, a collaboration between the Public Dental Service (PDS) in Region Västra Götaland, and the University of Gothenburg was established with dentists and psychologists working together at the Clinic of Oral Medicine, PDS, in Gothenburg, Sweden [128, 129]. This was an attempt to help patients with severe DA, not only to obtain good oral health, but also to alleviate their DA.
The psychological treatment method was based on behavior therapy (BT) using the systematic desensitization technique developed by Wolpe [130]. A pioneering case study from the US [131] was also used as inspiration for the Gothenburg group, and over the years, more links have been developed with additional centers performing research into DA [132]. Together with a few other locations worldwide, the collaboration project in Gothenburg is one of the first major integrative efforts of psychology and odontology in order to treat DA, and to evaluate different treatment technologies. Today, a range of treatment methods, both pharmacological [133] and behavioral [134], are used to make dental care possible in patients with DA. In some cases, these methods are used only as a means to get through dental treatment, while, in other cases, there is also the explicit goal to reduce DA in the long term and to normalize the patient’s dental care attendance patterns and ability to cope with conventional dental care routines. Pharmacological methods consist of dental treatment under different kinds of sedation and general anesthesia. Behavioral interventions usually consist of anxiety reduction and coping techniques delivered by the dental team, or more comprehensive psychological treatments delivered by the dental team or a psychologist. Of the comprehensive psychological treatments, interventions based on BT or cognitive behavior therapy (CBT) are the most common, but other types of intervention also exist, with hypnotherapy probably being the best known [134].

Behavior therapy was first developed in the mid-20th century and was based on learning theory, as developed by the behaviorist approach to psychology. Later, cognitive therapy was developed with its focus on cognition, and the two traditions were combined into what is now called CBT. The term CBT thus includes a variety of interventions from behavioral to cognitive. Hereafter, the term CBT will be used for all treatment approaches that build on these types of intervention, although the content may vary substantially between treatments. In the treatment of specific phobias, and anxiety disorders in general, CBT has been established as an effective form of treatment [135]. In a meta-analysis of behavioral interventions for DA and dental phobia in adults, Kvale and colleagues reported medium to large effect sizes, despite extensive heterogeneity between studies [134]. However, this review did not evaluate the quality of the included studies in a systematic fashion, and included studies with different types of study design. In evaluating the effects of interventions aiming to reduce DA, several types of outcome measures are important. The subjective experience of reduced DA is often used as the main outcome and is measured on self-rating scales, of which the previously described DAS and DFS are commonly used. Although the experience of reduced DA is an important outcome it does not by itself
measure the effect of treatment on dental care utilization. Therefore, this type of data needs to be complemented by measures of whether regular dental care can be maintained after treatment for DA. Dentists’ ratings of the treatability of patients may be used to indicate whether dental treatment is possible [27], but follow-up data regarding the actual frequency and content of dental care visits are also considered important [28]. Other outcomes, such as quality of life and OHRQoL, as well as adverse effects, are important in order to establish the effect and drawbacks of interventions aiming to reduce DA.

Because of the substantial health-related and psychosocial effects of severe DA and dental phobia, it is important to establish effective treatments. Since CBT is the major treatment approach in DA and dental phobia, a new systematic review and meta-analysis of its treatment effect in adults, according to established standards of quality assessment, are needed.
AIM

The aim of this thesis was to investigate relationships between DA, psychosocial factors, oral health and OHRQoL in adults, and to assess the efficacy of CBT as a treatment for DA.

Study I
Study I aimed to investigate relationships between DA and the SOC, OHRQoL and health-related behavior in the adult general Swedish population.

Study II
Study II aimed to investigate the presence of ADHD in adult patients with severe DA, and to analyze the relationship between ADHD, oral health and DA in this group.

Study III
Study III aimed to investigate self-rated orofacial esthetics in adult patients with severe DA, and to compare the results with normative data from the general population. Furthermore, this study aimed to investigate the relationship between orofacial esthetics and psychological and oral health, as well as the internal reliability of the OES, in patients with severe DA.

Study IV
This study aimed to investigate the efficacy of CBT as a treatment for severe DA in adults by conducting a systematic review. The specific research question was: Is CBT a more effective treatment for dental phobia or severe DA, with regard to reduction of DA and acceptance of conventional dental treatment, than information, pharmacological sedation, general anesthesia or placebo/no treatment?
MATERIALS AND METHODS

Study I

Design and participants
Study I is a cross-sectional national survey. The sample was randomly selected from inhabitants in Sweden aged 19 years or above using the SPAR registry. The SPAR registry includes everyone who is registered as a resident in Sweden. The data collection was carried out as a telephone survey performed by TNS-SIFO, a telemarketing company focusing on public opinion and marketing surveys. Fixed or mobile phone numbers were used in the data collection. Individuals who could not speak Swedish or who had secret phone numbers could not be included. The response rate was 49.7 %, leaving a final study group of 3500 individuals. There were missing items on most of the measures, excluding those individuals from analyses regarding that particular measure. On the SOC measure, 245 individuals were excluded because of missing items, which was the highest number in all the measures. The study was approved by the Regional Ethical Review Board in Gothenburg (reg. no. 801-12).

Measures
Dental anxiety was measured using the Dental Anxiety Question (DAQ) [136]. The DAQ is a single-item measure consisting of the question “Are you afraid of going to the dentist?”, with the response options “no”; “a little”; “yes, quite”; or, “yes, very”. Single-item measures of DA are common in epidemiological surveys and Neverlien reports good validity of the DAQ [136]. In the analysis, the result of the DAQ was dichotomized into a low DA group, including those with no or little DA, and a high DA group, including those with moderate (response option “yes, quite”), or extreme (response option “yes, very”) DA. In some analyses, the extreme outcomes on the DAQ were also analyzed: no DA (response option “no”) and extreme DA (response option “yes, very”).

The SOC measure consisted of three items (SOC-3) [137]. The scale was developed to capture the three dimensions of SOC: manageability, meaningfulness and comprehensibility. Each item consisted of a question that was operationalized to measure one of the dimensions [137]. The questions for each dimension were: manageability; “Do you usually see a solution to problems and difficulties that other people find hopeless?”, meaningfulness;
“Do you usually feel that your daily life is a source of personal satisfaction?”, and comprehensibility; “Do you usually feel that the things that happen to you in your daily life are hard to understand?”. Each item was scored from zero to two with reversed scoring for comprehensibility, giving a total score of zero to six with a high score indicating a low SOC. Acceptable reliability for the scale has been reported [137]. The validity of the scale was based on the face validity of the items and how these capture the essence of the three components of the SOC construct. Although correlations between the SOC-3 and Antonovsky’s original scale for measuring the SOC construct have also been reported, with a correlation of -0.72 for the 29-item version of the original scale and -0.67 for the 13-item version [138]. Since the SOC-3 and Antonovsky’s original scale go in opposite directions, the correlation coefficients become negative. The scale has been used in a substantial amount of research, although it has also been criticized, in part because of its inability to distinguish between individuals with a very strong SOC [138]; however, in study I, the focus is on sorting out individuals with the lowest SOC and, consequently, this does not present as a problem. In the analyses of the results in study I, the total score of the scale was dichotomized, defining participants as having either a strong SOC or a weak SOC. Several different types of dichotomization have been used in previous research. The division made by the originators of the scale was used in this case, which is in line with previous studies on the general Swedish population and sorts out individuals with the weakest SOC [139, 140].

Oral health-related quality of life was measured using an abbreviated version of the OHIP consisting of five items (OHIP-5) [141, 142]. The Swedish version of the OHIP-5 has shown a strong correlation with the original 49-item scale and acceptable reliability [142]. The items ask about the presence of problems in the domains of functional limitation, physical pain, psychological discomfort, physical disability and handicap. The respondent rates each question from “never” to “very often”, indicating the frequency of the particular problem described by each item. As previously described, the OHIP was based on Locker’s 1988 model of oral health, and was not initially presented as a measure of OHRQoL, but instead, as its name implies, as a measure of specific impacts related to oral disorders [107]. This conceptual way of measuring OHRQoL is very common [105], and the different versions of the OHIP are widely used. Different OHIP scoring methods exist, and it is sometimes scored in a way that dichotomizes each item, showing whether the particular item is indicative of an existing problem or not. Which response option that indicates an existing problem is not defined, and different levels are used in existing research. This study follows previous research concerning DA and SOC in this dichotomization [108, 143]. In the analyses
of study I, it was decided that zero to one indications of an existing problem would indicate a “high OHRQoL”, and two to five indications would be indicative of a “low OHRQoL”

Oral and general health-related behavior was measured with a series of items including tooth-brushing and flossing frequency, attendance of dental care and smoking. The answers to these questions were all dichotomized for analysis. Tooth-brushing was divided into once a day or less, or twice a day or more. Flossing frequency was divided into once a week or more often, or less frequently than once a week. Attendance of dental care was divided into “regular” (once every other year or more often), or “irregular” (less frequently than every other year, only acutely or never), and smoking was divided into daily smokers and not daily smokers.

Level of education and country of birth was measured with two questions. Education was divided into “low” (≤ 9 years), “medium” (10 - 12 years), and “high” (> 12 years). For country of birth, the following response options were available: Sweden; another Nordic country; non-Nordic country.

In order to examine the representativeness of the sample, comparisons were made between the study sample and total population data from Statistics Sweden (SCB) [144] regarding age, gender, level of education and country of birth.

Study II and III

Design and participants

Study II and III are cross-sectional clinical studies performed at the Clinic of Oral Medicine, the Public Dental Service, Region Västra Götaland, Gothenburg, Sweden, which specializes, i.a., in the treatment of patients with severe DA and dental phobia. The two studies were approved by the Regional Ethical Review Board in Gothenburg (nr 395-10), and written informed consent was given by all the participants. The two studies were based on the same data collection, although in study III, the data collection period was longer, resulting in larger number of subjects.

A consecutive sample of patients, referred or self-referred to the Clinic of Oral Medicine because of DA, was included in the studies. The data collection was performed as a part of the standard intake procedure at the clinic and included questionnaires, an adapted visual examination by a dentist, and a full radiographic examination. The following exclusion criteria
were applied to the two studies: 1) not understanding Swedish, or 2) having a severe mental or physical health problem that made it impossible for the patient to fill out questionnaires. In study III, some individuals sought treatment for other reasons than DA and they were likewise excluded.

In study II, of 141 patients who sought treatment for DA at the clinic, four were excluded because of language difficulties, 12 were excluded because of a severe mental or physical health problem, and 13 declined participation in the study. Of the remaining 112 patients, two were excluded because of missing items on the World Health Organization Adult ADHD Self-Report Scale (ASRS) Screener, which was the main measure in the study, leaving a final study group of 110 patients.

In study III of 207 patients seeking treatment, two sought for other reasons than DA and were therefore excluded. Another four were excluded because of a lack of understanding of Swedish, and 13 were excluded because of a mental or physical health problem. Thirty-three patients declined participation and another three patients were excluded because of more than two missing items on the OES, which was the main measure in the study. A final number of 152 patients were included in the study.

**Measures**

Measures included in Study II and III are listed in Table 1.

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**Clinical measures**

The numbers of decayed, missing and filled teeth were calculated for each patient, with the third molars excluded. Decayed, missing and filled teeth were used as individual measures or combined into the DMFT index. The DMFT index was originally developed to measure the effects of caries but
the reason why a tooth is filled or missing may also be due to other factors [98], which means that DMFT is not exclusively a measure of caries. Furthermore, the DMFT index does not clearly indicate treatment need, which is essential to patients with severe DA since previous research has shown a higher prevalence of decayed and missing teeth and a lower prevalence of filled teeth in this group. In study III, instead of using the DMFT index in the regression analysis, a summary score of the number of decayed and missing teeth was used. Previous research has also shown that root remnants are an important indicator of oral health in this group of patients and was included in these studies and defined as total destruction of the tooth crown. The presence of root remnants is an indicator of oral disease but also of avoidance of dental care, despite an obvious treatment need. The clinical measures were all recorded by a dentist who is also a co-author of Study II and III (K.B.).

**Questionnaires**
The World Health Organization Adult ADHD Self-Report Scale (ASRS) Screener [145, 146] was used in study II to identify possible cases of ADHD. The original ASRS instrument consists of 18 items, six of which with the highest concordance with a clinical diagnosis of ADHD are included in the ASRS screener [145]. A diagnosis of ADHD is often established by a comprehensive assessment by a psychiatrist or psychologist and any screening instrument is therefore bound to involve a substantial amount of error. The ASRS screener has the advantage of being a very quick way to screen for ADHD while maintaining reasonable psychometric properties. Two different scoring methods have been proposed for the ASRS screener and this study uses the one recommended by Kessler et al. [146]. Scored in this way, the ASRS screener had a sensitivity of 64.9 %, a specificity of 94.0 % and a total classification accuracy of 91.5 % in a sample of health plan subscribers in the US [146], and a sensitivity of 81 % and a specificity of 70 % in an international sample of patients seeking treatment for substance use disorders [147]. Since high DA is associated with a higher prevalence of psychological disorders and lower socioeconomic status, the sample in study II is probably not equal to the general population, but is probably not identical to a sample of patients seeking treatment for substance use disorders either. Despite the differences, these figures hint at the range in which the sensitivity and specificity probably lie. Semi-structured clinical interviews were used as the golden standard for assessing ADHD in the abovementioned validity studies [146, 147], which may also differ from the way ADHD is diagnosed in routine care. The total score of the ASRS screener can also be divided into four strata based on the likelihood of having an ADHD
diagnosis. This makes the symptomatology of ADHD more nuanced as a continuum, rather than merely provide a dichotomous diagnostic indication.

The Dental Fear Survey (DFS) [26] was used in both studies to assess DA. The DFS is one of the most widely used measures of DA and consists of 20 items covering anticipation anxiety, avoidance, physiological arousal and fear of specific stimuli. The DFS was originally not intended as a summary measure of DA, but more as an inventory of various aspects of DA [22]. Nevertheless, it is very frequently used as a summary measure and Schuurs and Hoogstraten [22] conclude in their review of the scale that the published data point to “a reasonably high reliability of DFS” (p. 332), and “in general, to at least some validity“ (p. 333). Reviews of available DA measures have recommended the use of the DFS in research [22, 24]. The many areas covered by the DFS also make it suitable for division into subscales. Two factor analyses, the first including four samples from different locations with demographic differences [148], and the second based on a sample of students [149], led to almost identical results, giving the subscales of anticipation anxiety and avoidance, fear of specific stimuli, and physiological arousal. However, a factor analysis of a clinical sample of patients with severe DA revealed a somewhat different factor structure, yielding the subscales of avoidance of dental care, physiological arousal, anticipatory anxiety, fear of drilling and fear of needles [150]. The subscales of this latter factor analysis are used in study II.

The presence of general anxiety and depression was assessed using the Hospital Anxiety and Depression Scale (HADS) [151]. The HADS is a screening questionnaire developed for use in medical settings [151], with the idea that only items that are not so easily confounded with symptoms of somatic disorders should be included. The HADS has been widely used and a systematic review has shown good psychometric properties [152]. The HADS consists of 14 questions divided into two subscales consisting of seven items each. The first subscale consists of symptoms of anxiety (HADS-A) and the second of symptoms of depression (HADS-D). For both HADS-A and HADS-D, a specific cutoff has been reported as the best level for identifying potential clinical cases of anxiety and depression [152]. The use of the HADS as a screening questionnaire for identifying specific psychiatric disorders was questioned recently, when studies of sensitivity and specificity of the HADS-D were reviewed [153]. The results showed a considerable variation between studies and the authors concluded that the sensitivity and specificity of the scale must be highly dependent on the context where it is applied [153]. Despite these results, the HADS gives an indication of the severity of the symptoms of anxiety and depression and, apart from being a
screening questionnaire, can also be used as a continuous measure of these dispositions.

The patients’ perception of their orofacial esthetics was measured in study III using the Orofacial Esthetic Scale (OES) [119]. The OES consists of seven items measuring how a person feels about the appearance of different aspects of his/her face, mouth, gingiva and teeth, and an eighth item consisting of a global assessment of the person’s orofacial esthetics. Each item is given a score that indicates the self-rated level of satisfaction or dissatisfaction with the particular aspect and a summary score may also be calculated. The OES was developed for use in prosthodontic patients and has shown good discriminatory validity in this group [154]. Good reliability of the scale has also been reported, both in prosthodontic patients and in the general population [154, 155]. Normative data for the general Swedish population have also been provided [156]. Since the scoring of the items range from very dissatisfied to very satisfied and no specified level that separates dissatisfied from satisfied is supplied, dissatisfaction and satisfaction are regarded as a continuum and not as separate concepts. This conceptualization will remain when reporting and discussing the results, where more dissatisfaction and less satisfaction will be considered as interchangeable terms.

Self-rated oral health was measured using a visual analog scale with the question “How do you rate your overall oral health?”, from 0 = “worst possible”, to 100 = “best possible”.

Background variables were collected using the clinic’s standard intake questionnaire, and included questions about regularity of dental treatment and years since last full dental treatment. Dental treatment had the response options of regular (once every, or every other, year), more seldom, only when having acute symptoms, and never. Years since the last full dental treatment had the response options of: < 5 years, 5-9 years, 10-14 years, 15-19 years, 20-25 years, > 25 years, and “I have never had full dental treatment”. Data on age and gender were also collected.

**Study IV**

**Design**

This study consists of a systematic literature review and a meta-analysis. The study was carried out as a Heath Technology Assessment (HTA) project in collaboration with the HTA Center at Sahlgrenska University Hospital, Region Västra Götaland.
Criteria for studies to be included were defined according to the Patients, Interventions, Controls, Outcome (PICO) methodology [157]. The PICO criteria were the following:

**Patients** = Adults (≥ 18 years) with dental phobia (according to DSM-IV or ICD-10) or severe DA (according to validated scales/instruments).

**Interventions** = Interventions based on CBT, including exposure therapy, systematic desensitization, or relaxation therapy.

**Comparisons** = Information, pharmacological sedation, general anesthesia or placebo/no treatment.

**Outcome** = Level of DA as measured using validated scales, acceptance of conventional dental treatment (without pharmacological sedation), dental treatability ratings, quality of life and OHRQoL, and complications.

A Randomized Controlled Trial (RCT) is generally considered to be the best study design for evaluating treatment. For this reason, it was decided only to include reports with RCT or systematic reviews as their study design.

A literature search was then performed by two librarians at the HTA Center during September, 2011, covering the databases of Pubmed, The Cochrane Library, Embase, CINAHL, PsycInfo, and additional HTA databases. The search covered papers published from January 1970 to August 2011. Reference lists of relevant articles were also scrutinized for additional references. Publications in English, Danish, Norwegian and Swedish were accepted. The main mesh terms used for the literature search were “Dental Anxiety”, “Behavior Therapy”, “Cognitive Therapy”, “Desensitization, Psychological”, and “Relaxation Therapy”. For a complete list of the literature search, see supporting information to study IV, Table S1. The literature search found a total of 1434 publications, 990 of which remained after duplicates had been removed. The abstracts of these articles were read by the librarians, who excluded 848 publications that did not fulfill the PICO criteria. Full text versions of the remaining 142 articles were assessed by the librarians, who excluded another 86 of these articles for not fulfilling the PICO criteria. The reasons for excluding these articles were: wrong patients/population (two articles), wrong intervention (one article), wrong comparison (14 articles), wrong outcome (one article), wrong study design (63 articles), other (five articles). The remaining 56 full text articles were assessed for eligibility by the research group, which excluded another 46 articles that can be found in the supporting information to study IV, Excluded
references. A final number of ten articles were included in the synthesis. A flow chart of the selection process can also be found in supporting information to study IV, Fig. S1.

Assessment of quality and grading of evidence
In general, the quality of RCTs differs, and RCTs of insufficient quality may be associated with bias, often exaggerating the treatment effect [158]. The reports of RCTs have been inadequate in many instances, rendering assessment of their methodological quality impossible. For example, many reports fail to state the method of randomization used, as well as the primary endpoints. In an attempt to improve the reporting standards of RCTs, the CONSORT (Consolidated Standards of Reporting Trials) statement was published in 1996, and was subsequently updated in 2001 and 2010 [158]. The CONSORT statement provides a checklist of what to include when reporting an RCT in order for the reader to be able to assess the quality of the trial. In this literature review and meta-analysis, the methodological quality of the included studies were assessed using a checklist for RCTs from the Swedish Council on Technology Assessment in Health Care (SBU) [159], which is in concordance with the CONSORT checklist, resulting in an overall rating of the quality of each study as high, moderate or low.

Once an estimate of the treatment effect thorough a meta-analysis or individual studies has been made, as well as an assessment of the methodological quality of the included studies, the overall quality of evidence of this treatment effect may be graded. In study IV, the GRADE (Grading of Recommendations Assessment, Development, and Evaluation) system [160] was followed in order to summarize all aspects into a single statement of the quality of evidence of the estimate. The GRADE system uses the following definition: “the quality of evidence indicates the extent to which we can be confident that an estimate of effect is correct” (p. 1490) [160], and builds on an approach where each important outcome is given its own quality of evidence graded at one of the following levels [161] (p. 926):

- High quality— “Further research is very unlikely to change our confidence in the estimate of effect”;
- Moderate quality— “Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate”;
- Low quality— “Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate”, and
• Very low quality— “Any estimate of effect is very uncertain”.

The following domains were assessed in a collective judgment, in order to obtain an overall evidence level for each outcome: Study limitations, which may indicate a risk of bias; consistency, which refers to the degree of similarity between estimates from different studies; directness, which refers to whether there is a direct link between the intervention and the ultimate outcome and if the interventions and populations are similar to those being considered; precision, which refers to the degree of certainty of an estimate; publication bias; magnitude of effect; and other important factors such as dose-response gradient.

Each included article, as well as the evidence for each outcome, was assessed separately by at least three reviewers and the overall assessment was then agreed by all authors of study IV. Disagreements were solved by consensus.

**Statistics**

In study I-III, descriptive statistics and inference testing were used. Bivariate comparisons were made with the t-test or the Mann-Whitney U-test depending on the distributions and sample sizes. Multivariate analyses were made using the ANOVA and the Kruskal-Wallis test. Logistic regression was also used in study I and linear regression in study III. Comparisons of proportions were made using the chi-square test or Fisher’s exact test. Correlations were calculated using the Pearson or Spearman coefficients. All calculations in study I-III were made using SPSS v. 19.0 (IBM Corp, Armonk, NY, USA), except for comparisons between the study sample and Swedish total population data in study I, and between the study sample and previously published general population data in study III. These calculations were made using Excel 2010 (Microsoft Corp.). The chosen level of significance in study I-III was $p < 0.05$, except for some analyses with multiple comparisons in study III, where a Bonferroni correction was made.

In study IV, weighted mean differences were used for continuous outcomes and heterogeneity between studies was determined using the chi-square test and the $I^2$ statistic ($p > 0.1, I^2 < 25$) [162]. In order to examine publication bias, a funnel plot was used. The statistical analyses were made using the Review Manager (RevMan), version 5.1 (Copenhagen: The Nordic Cochrane Center, The Cochrane Collaboration, 2011).
RESULTS

Study I

In the initial bivariate analyses regarding DA, the high DA group had a larger proportion of individuals with low OHRQoL, irregular dental care, daily smoking and female gender. Sense of coherence, socioeconomic status, tooth-brushing and dental flossing were not related to DA. To further investigate the relationship between SOC and DA, the extreme groups of DA, i.e., individuals who stated no vs. extreme DA, were compared with regard to the presence of a weak SOC. This revealed a statistically significant difference between the groups, with a larger proportion of a weak SOC in individuals with extreme DA. Two multivariate logistic regression analyses were then performed. The first had high and low DA as the dependent variable, while the second analysis used the extreme groups of DA as the dependent variable. The independent variables were the same for both logistic regressions and were based on the results of the bivariate analyses and theoretical considerations. Low OHRQoL, irregular dental care, daily smoking and female gender all strongly predicted high or extreme DA in the two analyses. The sense of coherence was not a statistically significant predictor in any of the two logistic regressions, but had an odds ratio of 1.54 (95% CI: 0.95-2.51) in the extreme groups analysis.

A non-participation analysis was carried out with comparisons between the study sample and data for the total Swedish population aged 19 or above, as supplied by Statistics Sweden (SCB). That analysis revealed that the study sample was more highly educated and consisted of fewer individuals born outside of Sweden. Also, the study sample had a somewhat higher mean age and consisted of slightly more women. The differences were mainly small and the study sample consisted of a wide range of individuals, as indicated by age, gender, education and country of birth.

Study II

In this clinical study, a high mean level of DA was found and a majority of patients reported dental care only in acute situations or never at all. Sixteen per cent of the included patients were identified as probable cases of ADHD using the ASRS screener (the ADHD group). The ADHD group had a higher level of DA and a poorer self-rated oral health compared with the remainder of the sample. There was also a tendency towards poorer clinical oral health
in the ADHD group, although these results were not statistically significant [ADHD group vs. non-ADHD group, mean (SD): decayed teeth 8.1 (7.4) vs. 6.4 (6.9); missing teeth 4.7 (5.2) vs. 3.3 (4.3); filled teeth 4.8 (4.3) vs. 6.8 (4.9); root remnants 2.4 (4.4) vs. 1.8 (3.2)]. Further analysis based on a four strata division of the total score from the ASRS screener revealed an extremely low self-rated oral health in the group most likely to have ADHD.

**Study III**

As in study II, a high level of DA and substantial avoidance of dental care were found in the sample. An initial analysis of the internal reliability of the OES revealed a strong Cronbach’s alpha for the first seven items, as well as when all eight items were included. High inter-item correlation was also seen between the two items measuring face and facial profile, and between the items measuring the mouth, gingiva and teeth. However, the items measuring the face and facial profile were less correlated to the items measuring the mouth, gingiva and teeth. The mean level of the individual items and the summary score of the OES were compared with general population data reported by Larsson [156], revealing a strong dissatisfaction with the orofacial appearance in the study group. Compared with the general population, patients with DA were generally less satisfied with all aspects of their orofacial esthetics, including the face, mouth, gingiva and teeth. Group comparisons revealed a relationship between less satisfaction with self-rated orofacial esthetics and older age, irregular dental care, and depressive symptoms, as well as with the occurrence of root remnants and increasing numbers of decayed teeth. Correlation analyses revealed an inverse relationship between satisfaction with orofacial esthetics and symptoms of depression and general anxiety, the number of decayed and missing teeth, root remnants, and self-rated oral health. Age, the sum of decayed and missing teeth, self-rated oral health, and symptoms of depression or general anxiety were all significant predictors of the OES summary score in linear regression analyses.

**Study IV**

The systematic literature review identified ten scientific publications originating from seven different RCTs. Five of the articles came from two trials with separate reports for follow-up data. There was considerable variation across studies in the type and length of interventions and with regard to by whom they were performed. All studies reported DA scores using the DAS and four studies also used the DFS. Four of the included
studies were of moderate quality and six of low quality. Five studies reported enough data to be included in a meta-analysis of the post-treatment effect on DA, showing a statistically significant effect of CBT with a mean decrease of 2.7 DAS scores. When the studies were divided by control conditions, a statistically significant effect of CBT treatment remained, with a mean decrease of 2.0 DAS scores when general anesthesia and sedation were used as control conditions, and a mean decrease of 3.3 DAS scores when no treatment was used as control condition (e.g., waiting list condition). A meta-analysis of the follow-up data from two trials collected one or two years after treatment was also performed, giving a statistically significant effect of CBT treatment compared with general anesthesia and sedation with a mean decrease of 2.3 DAS scores. No meta-analysis could be performed using the DFS. Only one study, which was of moderate quality, could be used to evaluate the effect of CBT on the acceptance of conventional dental treatment and dentist rating of dental treatability. This study showed that 80% of the patients in the CBT treatment group successfully completed two sessions of dental treatment, compared with 53% in the general anesthesia group. No data on quality of life, OHRQoL or complications were reported in any of the publications.

In essence, some support was found for CBT giving a clinically significant reduction in DA, as measured with the DAS after treatment and at follow-up after one to two years. These results are based on a low quality of evidence, according to GRADE. Furthermore, some support was also found for CBT improving the acceptance of conventional dental treatment, compared with general anesthesia. This result is also based on a low quality of evidence.
DISCUSSION

Psychosocial aspects, oral health and psychological treatment

The results of this thesis pointed to important health-related and psychosocial aspects associated with DA and, importantly, to the effect of psychological treatment in severe DA and dental phobia. In the first study, DA was associated with a substantial impact on OHRQoL, irregular dental care, female gender and smoking in the general population. These results are supported by previous research indicating the general tendency associated with DA to avoid dental care, followed by health-related consequences, as described by the vicious circle of DA [15, 40, 87, 112]. The results are also supported by previous research investigating the relationship between DA and gender [19] and smoking [90, 91]. The OHIP-5 scale that was used to measure OHRQoL in study I is based on Locker’s [98] previously described model of oral health. The OHIP-5 items ask about the presence of five common impacts of oral illness, where an elevated prevalence was associated with high DA in study I. The link between DA and poor OHRQoL in the general population has also been established in previous research using different conceptualizations of OHRQoL [103, 112, 115], pointing to a strong relationship between the two concepts. Apart from being a risk factor for oral disease, smoking has been associated with avoidance of dental care in individuals with DA [93] and in the general population [163, 164]. These relationships are probably not all direct and smoking may be an indicator of other important health-related behaviors and psychosocial factors.

A relationship between SOC and DA was also found in this thesis in the general population. Although this relationship was weak, studies on other samples with more elaborate measures confirm the relationship between SOC and DA. Especially in a study by Jaakkola et al., a strong relationship was found in 18-year-olds [72]. The SOC-3 scale used in study I to measure the SOC may not be as precise as the original SOC scales developed by Antonovsky and may capture the concept of SOC in a slightly different way, which also makes comparisons more difficult. All studies that have investigated the link between the SOC and DA are cross-sectional and say nothing about cause-and-effect relationships; however, according to the theory, a strong SOC is a protective factor against stress and facilitates the use of coping strategies once stress has occurred. Analyses have shown a mediating role of the SOC between trauma and psychological health.
including symptoms of anxiety [165, 166]. If this is true, a high SOC may also protect against developing DA after an aversive event or stress related to dentistry. A weak SOC, on the other hand, may increase the risk of developing DA, thus explaining the relationship found in this thesis and in previous research. A strong SOC may also contribute to facilitate the use of coping strategies in dentally anxious individuals, thereby limiting the negative consequences of DA, which may be a topic for further research.

Moving on to the clinical samples in this thesis, in patients with severe DA an elevated prevalence of ADHD was found, as indicated by a screening questionnaire. The occurrence of ADHD was also related to a higher level of DA. Previous research has found a high comorbidity between ADHD and anxiety in general [64, 68]. Although the etiology behind the combination of these two conditions is unclear [68], it probably includes several alternative pathways [167]. The relationship between ADHD and DA has previously been examined in children, where no general relationship was found [63, 168]. Instead, Blomqvist et al. found that children who had an ADHD diagnosis with many symptoms of hyperactivity/impulsivity had a higher mean level of DA compared with controls [63], indicating a key role for this symptom cluster in the relationship between ADHD and DA. In a previous study, Bergdahl and Bergdahl [169] found that DA in adults was associated with high ratings on the temperament dimension of Novelty Seeking in Cloninger’s Temperament and Character Inventory (TCI). High scores on the temperament dimension, which includes impulsivity, is generally found in children with ADHD [170-172], providing further evidence of a link between DA and the characteristics of ADHD, especially the hyperactivity/impulsivity cluster. It has also been shown that children with ADHD are more impulsive during dental examination [173], and these results taken together may indicate a possible trajectory leading to DA, which involves a higher risk of aversive experiences in the dental treatment situation due to more uncooperative behavior and poor communication. Children with ADHD also have more caries [174], which may call for more invasive treatment, further increasing the risk of aversive experiences. In a version of the EASI (Emotionality, Activity, Sociability, and Impulsivity) temperament survey adapted to adults, the temperament dimension of impulsivity was found to be related to DA in 15-year-olds [175] and adults [176]. The impulsivity dimension in the EASI describes impatience and lack of perseverance, with items more similar to the inattentiveness symptoms of ADHD than to symptoms of impulsivity/hyperactivity. It has been indicated that as children with ADHD grow older, their clinical characteristics often become less dominated by hyperactivity/impulsivity, whereas symptoms of inattentiveness remain to a greater extent [177, 178]. This could mean that the two symptom clusters of
hyperactivity/impulsivity and inattention may have different relations to DA depending on age. As found in study II, a general relationship between symptoms of ADHD and DA seems to exist in adults, and if impulsivity/hyperactivity are more related to the development of DA, these symptoms may largely have subsided in adults.

Another finding in this thesis was that patients with severe DA were generally found to be dissatisfied with their orofacial appearance, compared with the general population. This was true for all aspects, including the appearance of the face and facial profile, as well as the teeth, gingiva and mouth. These results point to what may be conceptualized as an important consequence of DA that has received little attention in past research. Orofacial esthetics probably has a key role in many of the psychosocial consequences experienced by individuals with high DA, with important implications for their quality of life. If dissatisfaction is conceptualized as an aspect of psychological well-being, it could be argued that self-rated orofacial appearance is in itself an aspect of OHRQoL that can be included in the disability category in Locker’s 1988 model of oral health. This kind of conceptualization is supported by results in study III, pointing to an association between dissatisfaction with appearance and symptoms of depression and general anxiety, clearly linking dissatisfaction to psychological well-being. Regarding the social consequences of appearance, previous research has provided data showing that we tend to attribute personality characteristics to individuals based on the esthetic properties of their teeth [179-181]. Embarrassment related to dental appearance and oral health has also previously been associated with DA [110, 120]. The finding that the orofacial esthetics are related to both psychological and oral health is supported by previous research on self-ratings of dental appearance [121], indicating that feelings about appearance may be associated with a variety of factors. These results further add to the increased consequences of DA experienced by individuals with additional symptoms of poor psychological health.

In summarizing the health-related and psychosocial factors related to DA and reported in this thesis and in previous research, the need for effective treatment of this important health-related condition becomes very clear. The results of this thesis also showed that CBT gives a significant reduction in DA, as measured with the DAS, and contributes to the acceptance of conventional dental care. However, these results were based on a low quality of evidence according to the GRADE system. The systematic review included in this thesis was based on strict, internationally established methodological criteria. An important part of the inclusion criteria was to
only include studies on populations with severe DA or dental phobia, in order to make the results generalizable to the treatment of patients who are normally referred or self-referred to specialized DA clinics or who can be considered highly dentally anxious. A previous systematic review have reported medium to large effect sizes of behavioral interventions in DA [134]. That review included different types of study designs and did not assess the methodological quality of the included studies in a systematic fashion, or grade the quality of evidence. Almost simultaneously with study IV, Gordon et al. published another review of treatment approaches to DA in adults [182]. This review included randomized clinical trials but did not include a systematic evaluation of the methodological quality of the included studies or a meta-analysis. An effect of the strict inclusion criteria used in this thesis is that the number of included studies was considerably smaller than in the two other reviews; however, the studies that were included were more comparable. A shared finding in all the published reviews is the heterogeneity between the CBT interventions used to treat DA in the included studies. This makes the generalization of results somewhat more difficult, but also points to the range of possible, effective interventions that may be applied in different settings. Furthermore, the lack of methodological quality and the small sample sizes noted in study IV was also emphasized in the review by Gordon et al. [182]. None of the included trials in study IV reported on quality of life, OHRQoL, or aversive effects as outcomes. Considering the substantial association between low OHRQoL and DA, as reported in this thesis and in previous research, as well as data indicating an increase in OHRQoL [111] and other positive psychosocial effects after treatment for DA, OHRQoL seems to be an important outcome measure. In general, more RCT studies of better design, including outcomes such as the level of DA, acceptance of conventional treatment, OHRQoL and aversive effects, are needed to fully assess the efficacy of CBT in severe DA and dental phobia.

**In and out of the vicious circle**

Starting with the etiology, ADHD and SOC may both be considered potentially influential in the development of DA. ADHD may contribute to the occurrence and persistence of DA, while a strong SOC may protect against it. Since a strong SOC is seen as a protective factor, a weak SOC may be considered as lacking this protective factor or as a risk factor in itself. Either way, both ADHD and a weak SOC may be described as endogenous in the endogenous/exogenous causality dimensions. Traditionally, the surrounding environment has been considered very important to the
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development of a weak or strong SOC, resulting in an internalized way of responding and relating to the world. ADHD, on the other hand, is mainly seen as a neurodevelopmental disorder, although there is some controversy regarding this. In this way, ADHD and SOC exemplify the importance of distinguishing between different factors that may all be regarded as endogenous in the development of DA. Interestingly, a strong SOC has been found to be protective against the development of ADHD symptoms, pointing to a relationship between these two concepts [183]. Apart from being associated with higher levels of DA, ADHD was also related to lower self-rated oral health in this thesis. This may further be contrasted with a strong SOC, which has been related to improved oral health and more positive health behavior in previous research [184-186]. Regarding ADHD, it is important to note that the results in this thesis were found in a sample with a generally very high level of DA and poor oral health. This may indicate that individuals with ADHD belong in the category with the worst prognosis with regard to DA and oral health in the vicious circle of DA. Apart from the factors discussed above, this may be due to the general difficulties of organization and planning and the high comorbidity with other psychiatric disorder, including substance use disorders, associated with ADHD [64].

As described previously, avoidance of dental care is an important factor in DA that seems to be essential for the deterioration of oral health and the psychosocial consequences associated with DA. Consequently, avoidance of dental care is the first step in the vicious circle of DA, and reducing avoidance is essential in trying to stop the vicious circle and to improve oral health. In the results from study III, female patients with severe DA more often had regular dental care and better oral status than the male patients. Other research has also found less avoidance related to DA in women than in men [94], giving an indication of differences in behavior regarding this aspect. In the treatment of DA, breaking the avoidance of dental care is essential. Behavioral interventions that build on the techniques of exposure or systematic desensitization are an integral part of CBT in DA and are highly focused on breaking avoidance. In these types of interventions, the patients come into contact with dental care in controlled forms, guided by a professional, and learn ways to handle their anxiety and reduce it. Further research is needed to strengthen the results found in this thesis regarding the effect of CBT, and one important area is to determine to what extent CBT reduces avoidance and if regular dental care can be maintained after treatment. The reports included in study IV also described the use of cognitive interventions, which may be used to alter cognitive thinking patterns regarding dental care and specific stimuli.
In study II and III, as well as in previous research, a variety of psychological symptoms have been associated with an increase in negative consequences associated with DA, thus amplifying the course described by the vicious circle and associated factors. However, the causality between DA, psychological symptoms and consequences has not been established. It is possible that psychological symptoms in general constitute a vulnerability regarding DA and reinforce its consequences. This may be an effect associated with the psychological symptoms themselves, or an indication of a generalized vulnerability, such as high levels of neuroticism. However, it may also be the case that poor oral health and its consequences have an impact on mental health that is considerably stronger than what is often recognized. The latter is supported by the reports of positive psychosocial effects from treatment at specialized DA clinics \[28, 79\]. The question has also been raised whether endogenous factors are related to the outcome of DA treatment. In a study by Vassend et al., the role of personality characteristics in the treatment of DA was examined and the conclusion was drawn that they did not predict the treatment result when the initial level of DA was controlled for \[187\]. Psychological symptoms as well as other endogenous factors probably interact with DA and its consequences in many ways. Well-designed prospective studies are needed to investigate this further.

**Strengths and limitations**

This thesis consists of four studies. The first is a general population survey of a large national sample. The second and third studies are clinical studies on patients referred or self-referred to a specialized DA clinic, and the fourth study is a systematic review and meta-analysis. Because of the cross-sectional design in studies I, II and III, no conclusions about causality can be drawn from these studies.

In study I there was a response rate of 49.7 %. The sample was better educated, somewhat older, and consisted of more individuals born in Sweden and slightly more women than the total adult Swedish population. Although, the differences were generally small, it cannot be ruled out that this influenced the results. Socioeconomic indicators were not related to DA in study I, which may be associated with the sample characteristics. Despite some differences between the study sample and the Swedish population, the sample in study I was comparatively large and consisted of individuals with a wide range of social and demographic characteristics. The instruments used to measure DA and SOC in study I were also very short and may therefore be
less precise. Regarding the SOC-3 measure and as previously described, it may not capture the phenomena of SOC in the same way as Antonovsky’s original scale. However, it may be important to investigate the same phenomena using different types of measures to determine whether the conclusion is still valid when the measures are conceptualized in slightly different ways.

Study II and III used a combination of well-established questionnaires and clinical measures to investigate a clinical sample at a specialized DA clinic. Despite some patients declining participation in the studies, the samples must be considered as representative of patients with severe DA. A strength of these two studies is the ability to investigate the interplay between psychosocial characteristics and objective as well as subjective/self-rated oral health. The ASRS screener that was used to measure ADHD in study II is associated with some error when compared with the golden standard for assessing an ADHD diagnosis. However, the proportion of adults with a positive screen was considerably high in study II, indicating a high prevalence of ADHD also when some measurement error is accounted for. The power to detect statistically significant differences between groups regarding clinical dental status was low in study II because of a relatively small number of subjects in the ADHD group, although the results from this study gave an indication of group differences that may be investigated in a larger sample. To our knowledge, the report on ADHD and DA is the first regarding this relationship in adults. Furthermore, a comprehensive report on orofacial esthetics and high DA has been lacking in the literature and the results from study III have substantially filled this gap.

As far as we know, the systematic review in study IV was the first to use strict criteria according to the GRADE system to evaluate the effect of psychological treatment in severe DA and dental phobia. Consequently, this review is most valuable, in that it assesses the quality of the existing evidence as well as identifies areas of knowledge that need to be strengthened.

In essence, the main strengths of this thesis were the comparatively large representative samples, the use of established methods and validated measures, as well as the novelty of its findings.
CONCLUSION

In this thesis, relationships between DA, psychosocial and oral health-related factors, as well as psychological treatment for DA, were investigated in adults. Dental anxiety was strongly correlated to poor OHRQoL, irregular dental care and smoking in the general population. Also, DA was weakly related to Antonovsky’s salutogenic concept of SOC. In patients with severe DA, ADHD was a common condition related to the level of DA and oral health. Furthermore, patients with severe DA were dissatisfied with all aspects of their orofacial appearance, and their ratings on this matter were related to both oral and psychological health. There are effective psychological methods that can reduce or eradicate DA in adult individuals, although there are substantial knowledge gaps to be investigated.
ACKNOWLEDGEMENTS

First, I would like to express my warmest gratitude to all the participants who generously shared their experiences and made this research possible.

I would also like to express my sincere gratitude to everyone who has helped and supported me in this work.

Special thanks go to:

Associate Professor Ulla Wide Boman, my main supervisor and co-author, who always believed in my capability and shared her expertise in the field, both as a clinician and as a researcher.

Professor Magnus Hakeberg, my supervisor and co-author, who has given me invaluable advice and encouragement.

Klas Blomkvist, LDS, my co-author in study II and III, who have taught me a great deal about odontology during our inspiring conversations, and who has been an incredible resource in the data collection.

Maria Westin, LDS, my co-author in study IV, with whom I shared many interesting thoughts during our intense work with study IV.

Per-Olof Rödström, LDS, head of the Clinic of Oral Medicine, who provided support and made this research possible.

Research coordinator Birgitta Ahlström, for her great work with posters and for general support.

All the colleagues at the Clinic of Oral Medicine, who participated in the recruitment of patients and the data collection, and who supported me in so many ways.

Maud Eriksson, Ann Liljegren, Petteri Sjögren and Annika Strandell at the Health Technology Assessment Center at Sahlgrenska University Hospital, for their great contribution to study IV.

Everyone at the Department of Behavioral and Community Dentistry, for their joy and support.
My colleagues at the Department of Research and Development, Region Kronoberg, for support and inspiring conversations.

Katarina Hedin, PhD, head of the Department of Research and Development, Region Kronoberg, who generously welcomed me to the department when I needed a place to work on my thesis.

My colleagues at Lasarettsrehab and the cancer rehabilitation team, Region Kronoberg, for much joy and encouragement.

My amazing wife Sofia, who has been fantastically supportive and encouraged me over the years, and my lovely children Heidi and Åke.

My parents, Karin and Jan, and my sister Klara, for all their help and support from the day I was born.

My parents-in-law, Kerstin and Leif, who have provided great support to me and my family.

All my friends, who have generously given me of their time and shared moments of joy with me.

**This research received funding from:***

The Local Research and Development Board for Gothenburg and Södra Bohuslän, Region Västra Götaland, Sweden.

The Health Technology Assessment Center, Region Västra Götaland, Sahlgrenska University Hospital, Gothenburg, Sweden.

The Agreement concerning research and education of doctors (TUA).

Institute of Odontology, The Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden.
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