Pain in Pediatric Dentistry

Experiences, attitudes and knowledge from the perspective of the child, the adolescent and the dentist

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ABSTRACT

Aim. The overall aim of this thesis was to study the frequency and intensity of general and oral pain, and oral discomfort in Swedish children and adolescents (with or without a disability). A further objective was to analyse dentists' knowledge about and attitudes to pain and pain management in the young patient.

The specific aims were:

To study the frequency and intensity of pain in children and adolescents' (without a disability) caused by dental treatment and everyday pain events; to analyse their pain experiences, using the Children's Pain Inventory (CPI), in relation to their gender, age, and dental anxiety.

To reduce the number of questions in the extended CPI and propose a short-version of the CPI that also includes dental treatment questions for use in clinical pain-scanning studies.

To study the prevalence of oral pain and discomfort in children and adolescents with an intellectual or physical disability, using the Dental Discomfort Questionnaire (DDQ) (and compare the DDQ with dental health, and oral hygiene as well as dietary habits), in relation to matched controls.

To study the knowledge about and attitudes to pain and pain management in children among Swedish general dentists by adapting an existing instrument for use among medical professionals to dentists: Dentists' Knowledge and Attitudes on Children's Pain perception (DKA-CPP).

Material and methods. The reports of 368 children and adolescents (8-19 year olds) on the 38 items CPI were analysed. Dental anxiety was evaluated by the Dental Anxiety Scale (DAS). The most frequently experienced CPI pain events were processed by Exploratory Factor Analysis in order to reduce the length of the questionnaire. A total of 188 (12-18-year-olds) with a disability (and their matched controls) were studied regarding DDQ. Dental records were analysed. Three hundred and eighty-seven general dentists were evaluated regarding their knowledge about and attitudes to pain and pain management.

Results. Half of the children and adolescents undergoing invasive dental procedures, 'Dental injection', 'Tooth drilling' or 'Tooth extraction', had experienced them as painful. The pain intensity experience was enhanced by higher dental anxiety, having a disability, being younger than 14 years old, or being female. The children and adolescents with a disability had statistically significantly higher DDQ scores compared with controls (despite similar dental health in both groups). The reduced CPI resulted in four factors (twelve items, of which one factor included 'Dental injection' and 'Dental X-ray'), explaining 79 % of the variance among the items. Dentists with more professional experience and/or female dentists applied more pain management strategies.

Conclusion. Young children, children with a disability and those with higher dental anxiety should be recognised as more susceptible to pain and should be offered additional care and pain relief during invasive dental treatments. The short CPI is proposed to be applied in clinical studies. Dentists should ensure all children customised and pain-free dental treatments.

Keywords child, adolescent, disability, oral, dental, discomfort, pain, invasive procedure, everyday, dentist, knowledge, attitude, questionnaire

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SAMMANFATTNING PÅ SVENSKA

Det övergripande syftet med avhandlingen var att studera svenska barn och ungdomars vardags-, orala- och tandvårdsrelaterade upplevelser av obehag och smärta. Målet var också att studera allmäntandläkares kunskaper och attityder gällande smärta och smärtbehandling hos den unga patienten.

De specifika målen i de fyra studierna var

- att studera förekomsten och intensiteten av vardags och tandvårdsrelaterad smärta hos barn och ungdomar utan funktionsnedsättning, (genom Children's Pain Inventory (CPI)) i relation till ålder, kön och tandvårdsrädsla.
- att reducera CPI formulärets frågor och föreslå en förkortat CPI version som också inkluderar tandvårdsrelaterade frågor för att kunna användas i kliniska studier.
- att studera barn och ungdomars (med funktionsnedsättning) upplevelser av orala obehag och smärta (Dental Discomfort Questionnaire (DDQ)) i relation till tandhälsa, oralhygien- och kostvanor, samt jämföra med matchade kontroller);
- att studera svenska allmäntandläkares kunskaper och attityder beträffande barns smärta och behandlingen av den, att adaptera ett befintligt formulär (som använts inom sjukvården) för att användas bland tandläkare (Dentists' Knowledge and Attitudes on Children's Pain perception (DKA-CPP)).

Material och metod. 368 barn och ungdomar (8-19 åringar) svar på CPI (38 frågor) analyserades. Tandvårdsrädsla evaluerades med Dental Anxiety Scale (DAS). De mest frekventa CPI incidenterna bearbetades med Exploratory Factor Analysis (EFA). DDQ för 188 barn och ungdomar (12-18 år) med funktionsnedsättning, samt deras köns och ålders matchade kontroller utvärderades. Tandläkarjournaler analyserades med avseende på tandhälsan. 387 allmäntandläkare evaluerades avseende kunskap och attityder genom DDK-CPP.

Resultat. Hälften av barnen och ungdomarna vilka hade upplevt invasiv tandvård såsom "*Oral injektion*", "*Tandborrning*" eller "*Tanduttagning*" hade upplevt den som smärtsam. De barn som antingen var tandvårdsrädda, hade funktionsnedsättning, var yngre än 14 år eller flickor rapporterade högre smärtintensitet. Barnen med funktionsnedsättning hade statistisk signifikant högre DDQ i jämförelse med de matchade kontrollerna, trots liknande tandhälsa i båda grupperna. Det reducerade CPI formuläret innehöll 4 faktorer (12 variabler, varav en faktor inkluderade "*Oral injektion"* och "*Oral röntgen"*), som förklarade 79 % av variansen för de studerade variablerna. Tandläkare med större yrkeserfarenhet, och/eller kvinnliga tandläkare använde sig av flera strategier för att behandla smärta hos barn.

Konklusion. Yngre barn, barn med funktionsnedsättning samt tandvårdsrädda kan anses vara mer smärtkänsliga och erbjudas extra omvårdnad och utökad smärtbehandling vid invasiv tandvård. Det förkortade CPI formuläret rekommenderas för kliniska studier. Tandläkare ska erbjuda alla barn individuellt anpassad och smärtfri tandvård.

LIST OF PAPERS

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

I. Krekmanova L, Bergius M, Robertson A, Sabel N, Hafström C, Klingberg G, Berggren U. Everyday- and dental pain experiences in healthy Swedish 8-19 year olds: an epidemiological study. Int J Paediatr Dent 2009;19:438-447.

II. Krekmanova L, Hakeberg M, Robertson A, Klingberg G. Common experiences of pain in children and adolescents-an Exploratory Factor Analysis of a questionnaire. Swed Dent J 2013;37:31-38.

III. Krekmanova L, Hakeberg M, Robertson A, Braathen G, Klingberg G. Perceived oral discomfort and pain in children and adolescents with intellectual or physical disabilities as reported by their legal guardians. Eur Arch Paediatr Dent 2016;17:223-230.

IV. Krekmanova L, Hakeberg M, Robertson A, Klingberg G. Dentists' Knowledge and Attitudes toward Child Pain Perception (DKA-CPP) – A novel measurement to understand pain management in dental care for children and adolescents. In manuscript.

CONTENT

ABBREVIATIONS

ADHD	Attention Deficit Hyperactivity Disorder
CFSS-DS	Children's Fear Survey Schedule-Dental Subscale
CPI	Children's Pain Inventory
DA	Dental Anxiety
DAS	Dental Anxiety Scale
DDQ	Dental Discomfort Questionnaire
DF	Dental Fear
DFA	Dental Fear and Anxiety
DKA-CPP	Dentists' Knowledge and Attitudes on Children's Pain Perception
DMFT	Decayed Missing Filled Teeth
EFA	Exploratory Factor Analysis
IASP	International Association for the Study of Pain
PDS	Public Dental Service
RVG	Region Västra Götaland
SES	Socio-economic status
VAS	Visual Analogue Scale
WHO	World Health Organization

1 INTRODUCTION

Dental care may be associated with pain and discomfort. Furthermore, these phenomena may be intertwined with fear and anxiety**,** especially among children with treatment needs. The young patient's pain expectations may also add to the situational complexity. Consequently, dental health care may be perceived as a challenging—sometimes even insurmountable—problem. For the patient with a disability, such negative experiences may add to an already difficult situation. The general view today is that the young dental patient is particularly vulnerable during the 55yhperiod of childhood and adolescence, because of different cognitional prerequisites and developmental processes, intellectual as well as physical, compared with adults.

1.1 Pain

1.1.1 Definition

The International Association for the Study of Pain (IASP) has defined pain as: 'An unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage'. In the definition, *sensory* refers to the senses, such as touch, hearing, taste, smell or sight; creating a sensation, which is conveyed *per se* from the sensory organs, through a nerve impulse to the nerve centres. The term *emotional* is associated with the state of mind or with feelings, as well as the prevailing motivation at the time of the experience (1). Furthermore, *actual or potential tissue damage* suggests that a tissue injury is not required for the experience of pain. However, the individual's own cognition, maturity, knowledge and understanding of pain is a prerequisite for how danger or tissue damage is interpreted in a given situation, resulting in the experience or absence of pain. This means that pain is not tied to a stimulus in the IASP definition. By similar reasoning, a nerve impulse that is transferred to the central nervous system (CNS) is not to be equated with a painful experience (1, 2). The expression *tissue damage* refers to the inflammatory processes following tissue injury or infection that trigger the transmission of a physiological impulse, manifested, for example, as toothache (3).

The definition of pain has been remodelled over time, according to the prevailing scientific view at a specific point in time. For example, in the 1960s, the view on pain developed towards a multifactorial and subjective experience, with sensory, cognitive, and emotional dimensions. Accordingly, in 1968, McCaffery stressed that pain is 'whatever the experiencing person says that it is, existing whenever and wherever the

person says it does' (4). The view to promote the patient's own pain definition, expressed by self-reporting, is well intentioned, but it still excludes children with developmental intellectual disabilities (who may not, fully or at all, be capable of expressing their experiences).

Table 1**. Definitions in alphabetical order, as stated by IASP*, United Nations Convention on the Rights of the Child (UNCRC)** and the World Health Organization (WHO), Treede et al.****, Carr et Goudas***** (1, 5-8)*

Term	**Definition**
Acute pain*****	The normal, predicted physiological response to an adverse chemical, thermal or mechanical stimulus associated with surgery, trauma and acute illness. Acute pain is defined as pain lasting less than three to six months.
Adolescence***	A period of human growth and development that occurs after childhood and before adulthood, from ages ten to19.
Analgesia*	Absence of pain in response to stimulation that would normally be painful.
Central sensitisation*	Increased responsiveness of nociceptive neurons in the central nervous system to their normal or subthreshold afferent input.
Child**	An individual below the age of 18 years.
Childhood**	A separate space from adulthood, recognising that what is appropriate for an adult may not be suitable for a child.
Chronic/ long-lasting pain****	Pain in one or more anatomical regions that persists or recurs over a period longer than three months and is associated with significant emotional distress or significant functional disability (interference with activities of daily life and participation in social roles) and that cannot be better explained by another chronic pain condition.
Everyday pain	Acute pain experienced during everyday events, such as 'got a splinter in the finger' or 'hit my toe'.
Nociceptor*	A high-threshold sensory receptor of the peripheral somatosensory nervous system that is capable of transducing and encoding noxious stimuli.
Nociceptive neuron*	A central or peripheral neuron of the somatosensory nervous system that is capable of encoding noxious stimuli.
Nociceptive pain*	Pain that arises from actual or threatened damage to non-neural tissue and is due to the activation of nociceptors.
Nociceptive stimulus*	An actually or potentially tissue-damaging event transduced and encoded by nociceptors.

Table 1, continued

Noxious* stimulus	A stimulus that is damaging or threatens damage to normal tissues.
Pain Intensity*	The external measurable part of pain.
Pain threshold*	The minimum intensity of a stimulus that is perceived as painful. Properly defined, the threshold is really the experience of the patient, whereas the intensity measured is an external event.
Pain tolerance level*	The maximum intensity of a pain-producing stimulus that a subject is willing to accept in a given situation.
Peripheral sensitisation*	Increased responsiveness and reduced threshold of nociceptive neurons in the periphery to the stimulation of their receptive fields.
Procedural pain	Pain initiated by medical and dental procedures/treatments, such as vaccination or tooth-drilling.
Recurrent pain	Acute pain that returns, such as headache, shoulder and back pain, among others.
Sensitisation*	Increased responsiveness of nociceptive neurons to their normal input, and/or recruitment of a response to normally sub-threshold inputs.

1.1.2 Physiological aspects of pain

Descartes described, in 1664, the path of ‘acute’ pain transmission that could imply ‘a specific pain pathway, a single channel from the skin to the brain, that carries messages from a peripheral pain receptor to a pain centre in the brain’(9).

Adding to the specificity theory was the neuromatrix concept. This comprised sensory, affective and cognitive neuromodules (10). It stipulated that ‘Pain is a multidimensional experience produced by characteristic “neurosignature” patterns of nerve impulses generated by a widely distributed neural system—the “body-self neuromatrix”—in the brain’ (10). The idea was that the neurosignature patterns could be triggered by sensory inputs, but also generated independently of them.

The prerequisite for a physiological pain reaction is the anatomical entity of the Central Nervous System (CNS): the brain and the spinal cord as well as the peripheral nervous system. These structures are built by neurons; the nerve cells consisting of cell bodies, dendrites and axons (11). The acute pain evoked by a noxious input is well understood today. Starting on a peripheral level, the nociceptor, a free sensory nerve ending, found, for example, in the tooth pulp, the skin and in muscles, detects a stimulus. A triggering input could be heat/chemical irritation or internal chemical

mediators, such as bradykinin, serotonin, prostaglandin, substance P or histamines. As the stimulus reaches the necessary intensity, an action potential is generated and propagated to the Central Nervous System (CNS).

Two kinds of axons are responsible for the propagation of action potentials from nociceptors. The afferent A delta fibres, which are myelinated and serve as rapid conductors (3-30 meters/sec.), give rise to a sharp and well localised sensation. In contrast, the afferent unmyelinated C fibres are slow conductors (0.5-2.0 meters/sec.), and usually produce a more widespread and diffuse perception. The action potential may be further conveyed to the brain cortex and experienced there as pain (12).

1.1.3 Pain perception, infant to adolescent

In the last decades, factors related to the disciplines of genetics, psychology, socio-economy and culture, have also been recognised as intervening with the physiological pain path (13-15). Even though much is known about the genesis of pain, the whole pain phenomenon is still not fully understood.

In the 1920s, Piaget developed the theory about children's consecutive mental developmental stages, with each stage being dependent on the existence of the previous stage. This theory presumed the child's interaction with the environment, providing the possibility continuously to evolve the child's own sensory-motor and abstract skills. During the interaction, individual intrinsic and extrinsic differences, such as temperamental traits, having a disability, or the capability of attachment and coping, may be decisive for the outcome (16). Seen in this way, the child is being challenged throughout a long period of growth and maturation into adolescence (17).

Today, it is acknowledged that the diverse developmental physical and intellectual abilities cause children and adolescents to understand and express pain differently from adults. Still, until modern time, there have been scientific misconceptions about the infant's and the child's perception of pain, such as:

Infants do not feel as much pain as adults;
Young children cannot determine the location of pain;
Active/playing children or sleeping children cannot be in pain;
Children with an intellectual disability may experience less pain.

These assumptions were ascribed to the immature central nervous system of the infant and the young child, but they have all been proven wrong in

the past decades. For example, studies have shown that 90 % of the brain regions involved in the pain reactions of adults are also activated in newborn children (18-22). Furthermore, individual variances in the genetics of pain have been recognised in the management of acute postoperative pain (23). It is also acknowledged that the infant's untreated pain during interventions or a postoperative period may result in long-term consequences, in the form of a physiologically and/or psychologically altered response to pain (24-27).
Due to the immature processing of the nervous system, the currently prevailing knowledge instead holds that the infant, the young child, the child with an intellectual disability (such as Down syndrome), as well as the adolescent, are more vulnerable to pain than adults (21, 28-30). These observations influence dental health care, by enabling the dentist to identify and respond to individual pain susceptibility based on the patient's pain history.

1.1.4 Desensitisation or sensitisation to pain

It has been discussed whether repeated exposure to pain stimuli lead to a heightened or lowered pain threshold, to desensitisation or sensitisation. Both of these responses may take place on a physiological, i.e. peripheral, and/or central CNS level, as well as on a psychological level. Data suggest that sensitisation (peripheral or central) is expected to occur more often in neonates and younger children (31, 32). One example is a study by Fearon & McGrath, 1996, which observed children three to seven years old in a day care centre and recorded the frequency of painful events in connection with the children's behaviour. The frequent exposure to pain resulted in obvious sensitisation. The more frequently the painful incidents appeared, the more severe the children's emotional reactions (33). On the other hand, adolescents with diabetes mellitus have reported lowered pain intensity over time to pain provoked by the insulin syringe. As the injection in these cases is a recurrent life-saving procedure, at least a psychological influence modulating the pain perception may be present (31).

1.2 Recurrent, everyday and procedural pain

The expressions *acute recurrent*, *everyday* and *procedural* pain, debated in this thesis mirror the terminology used in studies describing the occurrence of various forms of acute pain in children and adolescents.
Acute recurrent pain occurs in prevalence surveys, for instance, as sporadic headache, stomach ache and back or shoulder ache.

Everyday pain, less frequently studied as a term in the literature, partly overlaps 'recurrent' pain but indicates other acute pain-causing events, such as accidental events at home or during sports exercise or playing.
Acute *procedural* pain, on the other hand, is understood to be initiated by medical and dental procedures and/or treatment, such as vaccination or tooth-drilling.

1.2.1 Occurrence of recurrent, every-day and procedural pain

Recurrent pain occurrence

The acute *recurrent* pain prevalence among the young population varies considerably, which may be ascribed to the prevailing psychosocial conditions in society or different pain definitions and studied age ranges. There may also be unrecorded data that add to the uncertainty of the figures. Table 2 depicts experiences of pain in children and adolescents in relation to gender and age.

TABELL 2 pådenna sida

Epidemiological surveys indicate that around 400,000 of the approximately two millions Swedish youths below the age of 18 years, experience recurrent pain in the form of headache, abdominal pain and back pain (34-41).
Furthermore, in a survey of 2597 Swedish schoolchildren, 30 % of the 10-18-year-olds reported weekly headache due to stress. The boys in this survey responded more often to stress, with either headache or abdominal pain, than the girls. Over 40 % of the children experienced either headache or abdominal pain as often as several times a month. In contrast, the co-occurrence of diverse pain was reported more often by the girls (42).
Holm et al., 2012, described 154 Swedish 8-16-year-olds, seeking paediatric primary care due to pain mostly located to the head, shoulders and back, as well as the limbs and stomach. Fifty per cent of the children and adolescents were reported to have pain with pain-free intervals (43).
Among 28.899 Swedish schoolchildren aged 12-19 years, Nilsson et al. found the prevalence of self-reported temporomandibular disorder (TMD) pain to be 4.2 %. The pain prevalence increased with age. Moreover, among the 5-17-year-olds, TMD pain was strongly associated with headache. Girls typically report pain more frequently than boys in studies (39, 40, 44-48).

Everyday pain occurrence

Under ordinary and healthy circumstances, the everyday encounters with pain constitute a child's major source of experiencing pain; for example, through more or less severe daily events: '*Stubbed the toe'*, '*Got hit by a ball'* or '*Got a splinter'*. Events of this kind have been reported by 67-94 % of children and adolescents (49, 50).
In the literature, *everyday* pain includes pain events such as headache, abdominal pain and earache, and is partly interchangeable with the definition of recurrent pain (33, 49, 51, 52).

In contrast, children with a disability may experience daily pain that is often connected with their condition. One example is painful epileptic seizures or a myotonic muscle state in children with cerebral palsy. In a proxy report, *assisted stretching* was indicated to be the everyday activity most frequently associated with pain (53).
The combined occurrence of *recurrent* and *everyday* pain may possibly provide the most representative picture of the child's regular pain exposure.

Procedural pain occurrence

For medical or dental procedural pain to be experienced, an individual need for a diagnostic or therapeutic procedure must be present. This need may be the result of conditions such as disabilities, general health problems, sickness and oral health. An example of a potentially painful process is the '*needle injection',* which has been reported to be the most frequent pain-causing medical procedure among children with cerebral palsy (53).

In dental care, procedural pain may be provoked by the use of tissue-invasive instruments such as oral anaesthesia syringes, tooth drills or extraction forceps, among other instruments. In the literature, these procedures have been often reported and discussed as potentially causing dental fear (DF) or behaviour management problems (BMP). The primary aim of studies has less often been to analyse the occurrence of painful dental procedures among the young population (54-56). This makes it difficult to estimate the occurrence of procedural pain.

Procedures that are not tissue-invasive, such as orthodontic treatment, have traditionally been seen as not inducing pain. This has been contradicted by researchers, who report that a majority of the young patients studied perceive dental separators and orthodontic wires as painful (57, 58).

It may be hypothesised that a young individual with frequent experiences of acute *recurrent* and *everyday* pain events might become sensitised to pain, which could aggravate the dental care situation. The dentist's awareness of the young patient's pain history and pain status is important in order to prevent suffering.

1.3 Factors that may influence pain perception

1.3.1 Intellectual and physical functions

The World Health Organization (WHO) has stated: 'Disabilities is an umbrella term, covering impairments, activity limitations, and participation restrictions. An impairment is a problem in body function or structure; an activity limitation is a difficulty encountered by an individual in executing a task or action; while a participation restriction is a problem experienced by an individual in involvement in life situations' (59).

As intellectual and physical functions may vary within a group diagnosed with the same condition (for example, Down syndrome or cerebral palsy), the actual medical diagnosis may be of secondary importance in relation to a child's actual functioning.

In order to give different operators, such as clinicians, researchers, policymakers and family members, the possibility to document an individual's characteristics of health and functioning, the WHO published the International Classification of Functioning (ICF) in 2001. This version was further modified, specifically to evaluate the young population, children and youths (ICF-CY). The ICF-CY offers a conceptual outline in a common language and terminology for the recording of difficulties manifested in infancy, childhood and adolescence. It identifies physical and intellectual functions, activity limitations and participation restrictions, as well as environmental factors important for children and youth (60).

It should be especially considered that children and adolescents with disabilities face individual barriers that are often greater than those of others, potentially affecting their self-caring ability and, possibly, their own oral and dental health. These circumstances might lead to additional dental treatment needs and a further risk of experiencing pain.

1.3.2 Anxiety and fear

Definitions

Anxiety and fear are consistent with normal reactions to unknown situations throughout the developmental period of childhood and adolescence. The terms *anxiety* and *fear* have a similar and, to some extent, shared meaning; however, they are defined as separate entities, according to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), (American Psychiatric Association 2013) (61).

Anxiety is defined as: 'The apprehensive anticipation of future danger or misfortune accompanied by a feeling of worry, distress, and/or somatic symptoms of tension'. The focus of the anticipated danger may be internal or external.

Fear is defined as: 'An emotional response to perceived imminent threat or danger associated with urges to flee or fight'.

Dental fear

Dental Fear (DF) and Dental Anxiety (DA) have often been interchangeably used in the literature and also combined in the term *Dental Fear and Anxiety* (DFA), which is not a clearly defined entity. DFA has been used, for instance, to refer to strong feelings also included in dental phobia (characterised by persistent and unreasonable fear, immediate response to phobic stimuli and by the individual recognising the irrational

fear). In this thesis, the term DFA will further be used to denote both dental fear and dental anxiety.
The dental setting is a typical example of a potent fear-provoking situation, often encountered early in life when the child is particularly vulnerable. The risk of evolving DFA exists during all stages of childhood and adolescence, as dental interventions may give rise to insecurity, discomfort and pain. The origin of DFA is often complex and cannot be explained merely by one factor, in terms of cause and effect. Researchers have pointed out traumatic and painful dental treatment in early childhood as an associated factor for evolving DFA, influencing the patient's view on dental care into adulthood (62-65). The child's age and sex are considered to influence the occurrence of DFA. There is a trend for young children and girls to report pain more frequently, although the results from different studies are inconsistent. Other intrinsic and extrinsic factors, such as the child's temperament, culture and family background may further contribute to the development of DFA. The unpredictable relationship of the DFA-initiating factors demonstrates the complexity of fear and anxiety. In order to estimate the occurrence of DFA among the young population, diverse approaches have been employed, such as the child evaluating itself through a self-report form, the parent's proxy report or the dentist's observation of the child's behaviour. Various instruments and cut-offs, determining the degree of DFA, have also been used. The different methods have together resulted in a range of prevalence figures. DFA has been estimated to occur in 9 % of Swedish children (66-68).

1.3.3 Oral health

The WHO definition of oral health is: 'A state of being free from chronic mouth and facial pain, oral and throat cancer, oral sores, birth defects such as cleft lip and palate, periodontal disease, tooth decay and tooth loss, and other diseases and disorders that affect the oral cavity (and may limit an individual's capacity in biting, chewing, smiling, speaking, and psychosocial wellbeing). Risk factors for oral diseases include unhealthy diet, tobacco use, harmful alcohol use, and poor oral hygiene' (69).

Dental Caries

Dental caries is recognised as the 10th most prevalent condition worldwide, affecting 621 million children and constituting a major public health challenge (70). The global age-standardised prevalence and incidence of untreated caries have remained unchanged between 1990 and 2010 (71). A decrease in the prevalence of dental caries has been demonstrated in industrialised countries. There may be multiple factors responsible for this development, such as advanced prevention, use of fluoride, changes to the diagnostic criteria, and treatment decisions.

Dental caries statistics are traditionally presented as the mean DMFT (Decayed, Missed, and Filled Teeth of the permanent dentition) figure. The distribution of dental caries is generally skewed among children and adolescents, and, as a result, the mean DMFT figure gives distorted information about the most affected individuals. Bratthall, 2000, introduced The Significant Caries Index (SiC) to help visualise the group with the greatest needs, calculated from the third of the population with the highest DMFT figures (72).

In Sweden, the National Board of Health and Welfare reported in 2015 that the number of caries-free children and adolescents aged 3, 6, 12 and 19 years old were 96 %, 76 %, 68 % and 36 %, respectively. The data were based on manifest dental caries diagnosed at the above dental examination ages. A drawback was that approximately 1/6-1/5 of the age groups, in relation to the total child and adolescent population, was not included (73, 74). There is sparse knowledge and great uncertainty about the caries situation in children and adolescents with a disability, as no specific interconnected data are available. The data suggest that the dental caries prevalence is not significantly different at group level, compared to children without disabilities.

1.3.4 Attitudes, social and cultural aspects

Attitude is a complex cognitive process, the way the individual thinks and feels about someone or something. In the simplest case, attitudes, beliefs, and behaviour should be related. Children are susceptible to traditional and cultural approaches and prone to adopt the prevailing attitudes in their environment. The environment's expectations of the child's pain reaction can therefore be powerful and influence what the child believes and how it acts. The formation of attitudes, however, is a life-long modulating process. Altered living circumstances and requirements may modify the individual's views and behaviour (33, 75, 76).

1.3.5 Dentists' knowledge and attitudes

A dentist's personal attitude to pain may instinctively be brought into his/her professional practice, thus intervening with current knowledge about pain. As a result, dentists may not, for instance, ask about the patient's pain experience or offer available pain management.

So far, few studies have reported on dentists' knowledge and attitudes to pain and pain management in children and adolescents (77-80).

In a Finnish survey, Murtomaa et al., 1996, found that half of the dentists did not routinely ask the young patient about pain-related issues (77).

In a Swedish study, Wondimu et al., 2005, reported that local anaesthesia was used consistently by only 36 % of the dentists when performing restorative treatment in young patients. Also, 35 % of the dentists stated unresponsiveness to the patient's experiences of pain and psychological management (79). Furthermore, 42 % of the dentists expressed that 'Children occasionally report pain when they have no obvious reason for it'. In a Danish study, Rasmussen et al., 2005, reported that male dentists used topical anaesthesia in children less frequently than their female colleagues (78).

1.4 The questionnaire as sampling method

The use of a questionnaire is a common method to gather information on the various experiences of children and adolescents. However, there are several matters to be considered before using a questionnaire in a young population.

From the young patient's point of view, filling in the form should be a quick process and the form should be easy to understand and respond to. This is also desirable from society's point of view, as research is time-consuming and expensive.

From the researcher's perspective, the form should be as informative as possible, which may be a paradox, as a long questionnaire may be demanding for the responder. However, the form's variables are often related to each other, exploring the same issue from different angles, and sometimes repeating information. For these reasons, it may sometimes be desirable to revise and reduce questions in a form, thereby making it more usable in clinical and research settings.

When used with children and adolescents, the form's design, length and layout should be adapted to the target group's comprehension level (81). Thus, factors such as the responders' degree of maturity and their age,

gender, and cultural background should be taken into account. Furthermore, the terminology should be carefully selected. In order to improve the understandability of the questionnaire, the question and response process should be exemplified.

Self-reporting, also termed the gold standard and made verbally or in writing, is considered the best approach to gather information about a subjective experience, such as pain. First-hand information assumes the respondent 'speaking the truth'. But it should be considered that various situational and environmental factors may influence the authentic statement given by the child or adolescent (82).

Self-reports, as used in questionnaires, presume a specific developmental degree of maturity, linguistic skills and literacy, as well as honesty and frankness on the part of the responder. Children and adolescents with difficulties of cognitive and communicative function may not be able to express themselves sufficiently through this method. In these cases, a supplementary proxy report; i.e., an observer's report, may be helpful. However, it has been recognised that the reports by legal guardians and nurses on children's pain should be considered estimates rather than factual statements (82-87).

1.4.1 Methods for measuring pain and discomfort

Children's Pain Inventory

The Children's Pain Inventory (CPI), a self-report concept, was introduced by McGrath at the Children's Hospital in Western Ontario, Canada in 1990. The aim was to provide hypothetical pain situations for children in order to substitute the graded levels of experimental pain used in validity studies on adults. The CPI contained a list of 25 events, generally regarded as pain-provoking situations, and five events typically considered as not being pain-provoking experiences. These events described familiar recreational and medical situations and conditions with varying extent of tissue damage. In this way, the occurrence of the events could be estimated. A Visual Analogue Scale (VAS), a 160 mm long line, was further used to mark the pain intensity of each event. In the 1980s, VAS lines of 100 mm, 150 mm and 165 mm were occasionally used for this purpose (52, 88, 89). McGrath *et al*. subsequently presented combined CPI lists based on a 100 mm VAS. Examples of pain intensity of experienced items among 5-16-year-olds were '*Earache'* 43.0, '*Stubbed the toe*' 35.4, '*Burn'* 56.9 (52, 89). Based on the reports of 175 children, higher pain intensity was

reported for items such as '*Bruise'*, '*Cold'*, '*Finger prick'*, and *'Broken arm'* (49).

Visual Analogue Scale

Various instruments, including the Visual Analogue Scale (VAS), have been used since the 1920s in the social and behavioural sciences to measure a variety of subjective phenomena, such as quality of life, mood, stress and health. The VAS has also been widely used as a pain-assessing instrument and has been evaluated for validity and reliability among individuals ≥ 8 years old (52, 90, 91). The VAS is a horizontal line of 100 mm, defined at the left end as *No pain* (0) and at the right end as *Worst possible pain* (100). To indicate the pain intensity, a marking is placed on the line. The VAS score is then determined by measuring in millimetres from the left hand end to the marking. The cognitional challenge for the individual is to understand that the only measured characteristic is the pain intensity and that this figure increases going from left to right on the line.

Researchers have elaborated on the horizontal versus the vertical position of the VAS, as well as on different lengths, end marks and end phrases for the VAS. The horizontal VAS has been shown to produce a more uniform distribution (92). The end phrase *Worst possible pain* has been shown to yield fewer extremes compared with other phrases, such as *Intense* or *Unbearable pain* (93). For the indication of *Mild* or *Moderate pain* the upper limits on the VAS have been proposed to be 35 and 60, respectively (94).

Other pain assessment instruments

Various pain assessment instruments (self-report and observational scales) have been used in paediatric hospital wards worldwide.
Common scales in use, showing self-reporting of acute procedural, post-operative or disease-related pain, are the Faces Pain Scale (FPS) by *Bieri et al*., 1990, and the Numerical Rating Scale (NRS) by von Baeyer al., 2009 (95, 96).

Observational scales used for the estimation of pain in preverbal children and children unable to understand a self-report scale are, for example, FLACC (Face, Legs, Activity, Cry, and Consolability) and CHEOPS (Children's Hospital Eastern Ontario Scale) for children with postoperative conditions, estimating crying, facial expression, verbal expression, torso position, touch behaviour, and leg position (97, 98).

Dental Discomfort Questionnaire

The Dental Discomfort Questionnaire (DDQ) was developed to detect behaviour in young children, observed by their parents, indicating toothache. Decayed teeth may cause negative and changed manners of eating, chewing and sleeping, or may also be manifested as individually disturbing habits. Interviews were made with the parents of referred toddlers with toothache and caries. The information gathered resulted in the Dental Discomfort Questionnaire with eight questions. The DDQ has since been developed stepwise, with regard to the quality and number of the included questions. The children groups studied have ranged from preverbal children; i.e., toddlers, to children with learning disabilities (6-13-year-olds) (99-103). The DDQ can be described as a proxy report for children unable to communicate their dental discomfort and pain adequately.

1.4.2 Methods for measuring dental fear

Dental Anxiety Scale

The Dental Anxiety Scale (DAS) is a self-report instrument used both in clinical and research settings to measure DFA. The scale deals with overall dental anxiety and has mainly been used on adolescents and adults. The instrument contains four imagined dental situations, each with five response alternatives scoring 1-5, with a total score range of 4-20. A DAS cut-off value for dental fear of ≥ 15.0 has been suggested (104-109). There are established normative mean DAS values for the adult population, ranging between 7.87 (SD = 3.51) and 9.4 (SD = 2.9), but no established values for the younger population (110). Neverlien & Johnsen, 1991, used DAS in a group of 10-12-year-olds, resulting in a DAS mean of 8.44 (SD = 3.61). Blomqvist et. al., 2007, studied DAS in 13-year-olds with ADHD and their matched controls. The DAS mean for the controls was 6.5 (SD = 1.7) and 7.4 (SD = 3.5) for the children with ADHD (108, 110, 111).

Children's Fear Survey Schedule - Dental Subscale

The psychometric scale Children's Fear Survey Schedule-Dental Subscale (CFSS-DS) was developed by Cuthbert & Melamed, 1982, to measure DFA in children (112). This scale, including 15 questions, covers more aspects of the dental setting than the DAS. Each question in the CFSS-DS scores 1-5, from 'not at all afraid' to 'very afraid', with a total range score of 15-75. The scale has been widely used and validated for internal consistency, validity and reliability in different populations, both as a self-report scale in 8-17-year-olds, and a proxy report (66, 67, 113, 114). Three general factors have been distinguished in the validation process of the scale; fear of highly invasive procedures, fear of less invasive treatment aspects and fear of medical aspects and strangers (115).

1.5 Ethical considerations

Every child is subjected to the good intention and knowledge of the legal guardian as well as society's capability to act for the good of the individual. It is easy to ignore, directly or indirectly, the child's autonomy and will in different situations, for example, when inviting her/him to participate in research. Children and adolescents with intellectual or physical disabilities may be even more subjected to the will of others, due to additional limitations or challenging living circumstances. This makes ethical considerations necessary whenever children are in focus (116).

To further children's needs and welfare, societies worldwide have joined together to represent children's rights and prevent them from being physically or psychologically harmed (during the sensitive period of growth and maturation).

Several landmark documents promote the child's interest by stating their human rights: *The Declaration of the Rights of the Child* (1924), *The Code of Medical Ethics* (1949), *The Declaration of Helsinki* (1964), *The Belmont Report* (1979), and *The Convention on the Rights of the child* (1989) (116).

In Sweden, there are six regional boards, appointed by the government, which are entrusted with the task of executing the Ethical Review Act, updated in 2008. The ethical vetting board inspects research plans and protocols involving humans within the field of medical science; medicine, pharmacology, odontology, the science of health care and clinical psychology (117, 118).

In addition to the right to be protected in vulnerable situations, children are also entitled to participate actively in matters concerning their own wellbeing. They should be helped to express their own views and thoughts,

be properly informed about possible choices, and enabled to influence decision taken about them.

All children are also entitled to the highest standard of planned and fairly delivered dental health care, performed in collaboration with their families (118, 119).

As there is always a risk that children may be harmed when included in research, it could be argued that knowledge from studies performed on adults should be extrapolated to the young population. The priority should be to answer research questions without involving children, if this is at all possible. However, conclusions extrapolated from surveys performed on adults often have limited relevance for children and could even be harmful. This further challenges the researcher to consider the ethical dilemmas from different points of view and not allowing the obstacles to be an excuse for excluding children. As any aspect of the research setting could be ethically questioned, there are checkpoints to be addressed before the enrolment of the child in a survey.

The informed consent wording should be formulated on the basis of the anticipated intellectual level of the child, as it intends to tell the child about the study's aim and outline. The informed consent requirement also aims to engage the child in active decision-making about whether or not to participate. In Sweden, a child ≥ 15 years old can give informed consent, even if the legal guardian will not do so. Furthermore, a child aged ≥ 12 years may her/himself refuse participation, but not agree to participation without a legal guardian's assent.

In cases where the child is unable to give informed consent, for instance, because of an intellectual disability, this may be obtained from the legal guardian acting on behalf of the child, as long as it is expected to increase knowledge that is not otherwise obtainable, is relevant to the studied group and involves a negligible risk of injury. The ethical dilemma in such scenarios is that the child's autonomy and will may be ignored, irrespectively of whether the guardian assents or dissents (118).

Furthermore, confidentiality, ensuring the secrecy of all personal data throughout all research stages, is crucial. The ethical dilemma is that the child may not fully comprehend her/his own privacy or how personal risk-taking may influence her/himself.

Another important point is that the consent may be withdrawn at any time during the survey without stating a reason and without any negative consequences for the participant. Even if this possibility seems just, the

child may not feel in the position to withdraw the participation, because of its dependent situation.

In conclusion; however high the ethical standards may be, and regardless of whether all formal requirements are met, the child's position remains exposed and should therefore be continuously highlighted.

2 AIM

The overall aim was to explore children's and adolescents' experiences of general, oral and dental treatment pain. The aim was also to gain insight into the knowledge and attitudes of dentists to pain and pain management in children.

The specific aims were:
To study the occurrence and intensity of pain in children and adolescents (without disabilities), and to analyse the reported pain experiences in relation to sex, age, and dental anxiety;

To condense the Children's Pain Inventory (CPI) to be suitable for clinical studies among children and adolescents; to expose hitherto undiscovered dimensions of the CPI pain variables and thereby improve the psychometric properties of the CPI;

To study the occurrence of oral pain and discomfort in children and adolescents with intellectual or physical disabilities, using the Dental Discomfort Questionnaire (DDQ), and to compare the results with those of children without disabilities; to analyse the relationship between the DDQ and dental health, oral hygiene and dietary habits.

To explore the attitudes and knowledge of Swedish dentists to pain and pain management in children and adolescents, and to analyse for underlying explanations.

2.1.1 Hypothesis

Children and adolescents without disabilities experience everyday and dental treatment pain according to their age, gender and degree of dental anxiety. Pain reports by Swedish children are similar to previously studied child populations regarding occurrence and intensity.

The extended 38-question CPI form can be shortened using a statistical instrument.

Children and adolescents with intellectual or physical disabilities experience more oral discomfort and pain than age and sex-matched controls. Dental caries is more common in children with disabilities; oral hygiene procedures are carried out less often and the intake of food is more frequent, compared with controls.

The dentist's gender, age, years of professional experience, proportion of working time devoted to treating children or adolescents, and being a parent, influence the dentist's knowledge or attitudes to pain and pain management in children and adolescents.

3 PATIENTS AND METHODS

3.1 Background and study population

The studies were conducted in Region Västra Götaland (RVG) with a population of 1.6 million people. The RVG, with Södra Älvsborg, Fyrbodal, Skaraborg, Södra Bohuslän and the City of Göteborg (> 500,000 inhabitants) is the largest administrative region in Sweden (120, 121). The Public Dental Service (PDS) in RVG comprises clinics for general dental care, specialised dental care and hospital dentistry. In 2012, the PDS employed 50 % of all dentists working in the region, including 138 specialists and 567 general dentists working at 128 clinics. Approximately 95 % of the children and 50 % of the adults in the Västra Götaland region received their dental care within the PDS.

3.1.1 Dental care system for children in Sweden

All children and adolescents in Sweden up to and including the age of 19 are offered comprehensive regular dental health care, including specialist care when needed. Starting in 2017, there is a political intention to extend further the age limit for free dental health.

3.1.2 Study groups

The following groups were studied in this thesis:

Children and adolescents without a disability,
Paper I-II, III (control group).
Children and adolescents with a disability, Paper III.
General dentists at PDS clinics in RVG, Paper IV.

Study group of children and adolescents without disability (Paper I-II)

A total of 383 healthy children and adolescents (aged 8-19 years), hereafter referred to as 'without disability', were invited to participate (Paper I). They were regular patients and consecutive attendants at three general PDS clinics in the City of Göteborg (reflecting different social and economic backgrounds as well as different levels of oral health). The exclusion criteria were difficulties with the Swedish language or young age. The compiled data from these children and adolescents were also used for further exploration in the methodological study (Paper II).

Study group of children and adolescents with disability (Paper III)

A total of 188 children and adolescents, 81 girls, 107 boys (aged 12-18 years), were identified through the Child and Adolescent Habilitation Unit (CAHU), Göteborg and Södra Bohuslän, and invited to participate. The children were divided into four groups based on their intellectual and physical disabilities:

- D1 moderate to severe intellectual disability (IQ ≤ 49), aged 12-18
- D2a mild intellectual disability (IQ 50 – 70), aged 12-14
- D2b mild intellectual disability (IQ 50 – 70), aged 15-18
- D3 physical disability, aged 12-18

The exclusion criteria were not speaking Swedish or a diagnosis of an autism spectrum disorder.

Control group of children and adolescents without disability (Paper III)

A control group of children without a disability was identified for the participants with a disability, and matched for sex and age (± 3 months) and the clinic responsible for the dental care.

Study group of general dentists (Paper IV)

All general dentists employed at the PDS clinics in RVG were eligible and invited (the age categories were given as < 25 years, 25-35 years, 36-45 years, 46-55 years and older than 55 years). The survey was anonymous.

3.2 Methods

Three questionnaires were used: CPI original form, DDQ, and DKA-CCP, as depicted by Appendix I-III and shown in the back of this thesis. The DAS form is presented in Appendix I. Appendix IV illustrates the CPI variables that were statistically processed in order to condense the original form questions.

Table 3. *Studied topics, measuring instruments and number of items. Invited children/adolescents, dentists and number of respondents (N). The number of analysed respondents is shown with an asterisk*. The data collection method is shown (Paper I-IV).*

Studied topic/item topic/item	**Measuring instrument**	**No. of items**	**Invited N**	**Respondents N** Analysed*	**Collection method** (Paper)
Experience of Pain	**CPI** Ref (88)	38	383	368*	Questionnaire (I, II)
Dental discomfort and pain	**DDQ** Ref (100)	12	220	D group 142*/188 C group 135*/188, aged	Questionnaire (III)
Dentist's Knowledge and attitudes to Children's Pain Perception	**DKA-CPP** (Ref 121)	56	567	387*	Questionnaire (IV)
Dental Fear	**DAS** Ref (104)	4	383	368*	Questionnaire (I)
Dietary habits	Single items	3	220	D group 134*/188 C group 134*/188	Questionnaire (III)
Oral hygiene habits	Single items	7	220	D group 131-135*/188 C group 131-135*/188	Questionnaire (III)
Caries	**DMFT**		220	D group 142*/188 C group 135*/188	Dental records (III)

3.3 Data collection

3.3.1 Pain experience using the CPI, VAS (Papers I-II)

The Children's Pain Inventory (CPI), represented by two different lists previously used by McGrath, was modified. The 23-item 'acute trauma/disease pain' CPI list and four items from the 'acute treatment-related pains' list, were used (49, 89). The item '*vaccination*' was considered a significant general source of distress for children and adolescents and also included in the modified CPI. An additional ten dental treatment-related items were also added (57, 89, 107). Finally, the modified CPI had 38 items. The responder was instructed to indicate whether the CPI item had been encountered, using the alternatives *yes*, *no*, or *don't know*. If the answer was *yes*, they were asked to specify whether the experience had been painful, answering *yes always*, *yes sometimes*, *no never*, or *don't remember*. If the answer to this follow-up question was *yes always* or *yes sometimes*, the pain intensity was marked on a 100 mm Visual Analogue Scale (VAS), where 0 represented *no pain* and 100 the *worst possible pain*.

3.3.2 Dental Fear and Anxiety using the DAS (Paper I)

The Swedish version of the Dental Anxiety Scale (DAS) was used to assess Dental Fear and Anxiety (DFA) in the group of children without disabilities through four imagined dental treatment-related situations. The sum score range was 4-20 (104).

3.3.3 Discomfort and pain experience using the DDQ (Paper III)

The Dental Discomfort Questionnaire (DDQ) was used to measure discomfort and pain in children and adolescents with disabilities and their matched controls. This measure has twelve items, with the response options *never* (0), *sometimes* (1) or *often* (2), resulting in a total score range 0 to 24 (101).

3.3.4 Oral hygiene and dietary habits (Paper III)

All oral hygiene and dietary items had previously been utilised by Mobley, 2003, in the US. They were also used in Swedish studies performed on the young population (122-126).

3.3.5 Dental Records (Paper III)

Data on dental caries in the permanent dentition (DMFT) were obtained from dental records. The latest documented dental status was obtained. Radiographs were not assessed.

3.3.6 Dentists' Knowledge and Attitudes to Children's Pain Perception (Paper IV)

In order to assess the knowledge and attitudes among dentists to children's pain and pain management, the form Dentists' Knowledge and Attitudes to Children's Pain Perception (DKA-CPP) was used. The questionnaire was a modification based on the studies of Salanterä, 1999, 2000, and Enskär, 2007, applied to medical nurses (127-130)**.** The DKA-CPP included the following pain categories: A) *Views on the care of children in pain,* B) *Physiology of pain*, C) *Pain alleviation*, D) *Pain medication*, E) *Sociology and Psychology of pain*, F) *Pain assessment instruments and methods,* G) *Non-medication methods of pain alleviation,* and H) *Documentation of pain management*; in all 56 items. Background data on the dentists' age and sex, year of graduation, years of professional experience, and proportion of working time devoted to treating children and adolescents, and parenthood, were collected.

3.4 Statistical methods (Paper I-IV)

The Statistical package for the Social Sciences (SPSS) was used for the analyses in *Papers I-IV*. In all papers, descriptive statistics were compiled and an evaluation of the data type and distributional properties of the variables was performed. On the basis of this, it was decided whether to apply parametric or non-parametric statistical methods in the analyses.

Paper I. Differences in the Children's Pain Inventory (CPI) based on sex, age and level of dental anxiety (DAS) were tested with the Student's t-test and the Chi-square test. Cronbach's alpha was used to estimate the internal consistency reliability, and values exceeding 0.70 were deemed acceptable for group comparisons.

Paper II. The CPI data compiled in *Paper I* were analysed using Exploratory Factor Analysis (EFA) (131). This multi-step method could detect abundant and repeated information among the original CPI items and was used to reduce the item set. The EFA discovered dimensions among the items that were not directly observable, the so-called

latent factors. The EFA generated various *factor models*, including different numbers of *latent items*, holding different *loadings* (*affinity)* to each factor. The EFA method is not absolute but *heuristic*, which means that several of the generated EFA *factor models* could be considered a possible outcome. The factor model that was finally chosen was based on an interpretation by the researchers.

Before the EFA was applied to the data set, the items with gender linkage and response frequencies below 80 % were considered to be insufficiently representative to allow for further analysis and were omitted (Figure 1).

The remaining 18 CPI items, marked with an asterisk* (Appendix IV), were examined in a correlation matrix and inserted in the EFA. During the data processing there was no presumption on the number or nature of the latent factors and their items. Various *factor models* were subsequently tested and discussed. A stepwise assessment of each factor model was performed.

All interpretations and decisions on the final number of the extracted factors were consistently based on the following accepted principles:

The Kaiser's eigenvalue > 1 criterion, inspection of the scree plot and the interpretability of the item loadings. Factors with an eigenvalue larger than 1; i.e., explaining more than one variable, are retained.

The orthogonal rotation, using the Varimax method to maximise the variance in the factor model.

The explained variance - the interpretability (in %) of the factor model. Factors that account, in total, for 70-80 % of the variance are retained.

The applicability of the factor model in a clinical and/or research setting.

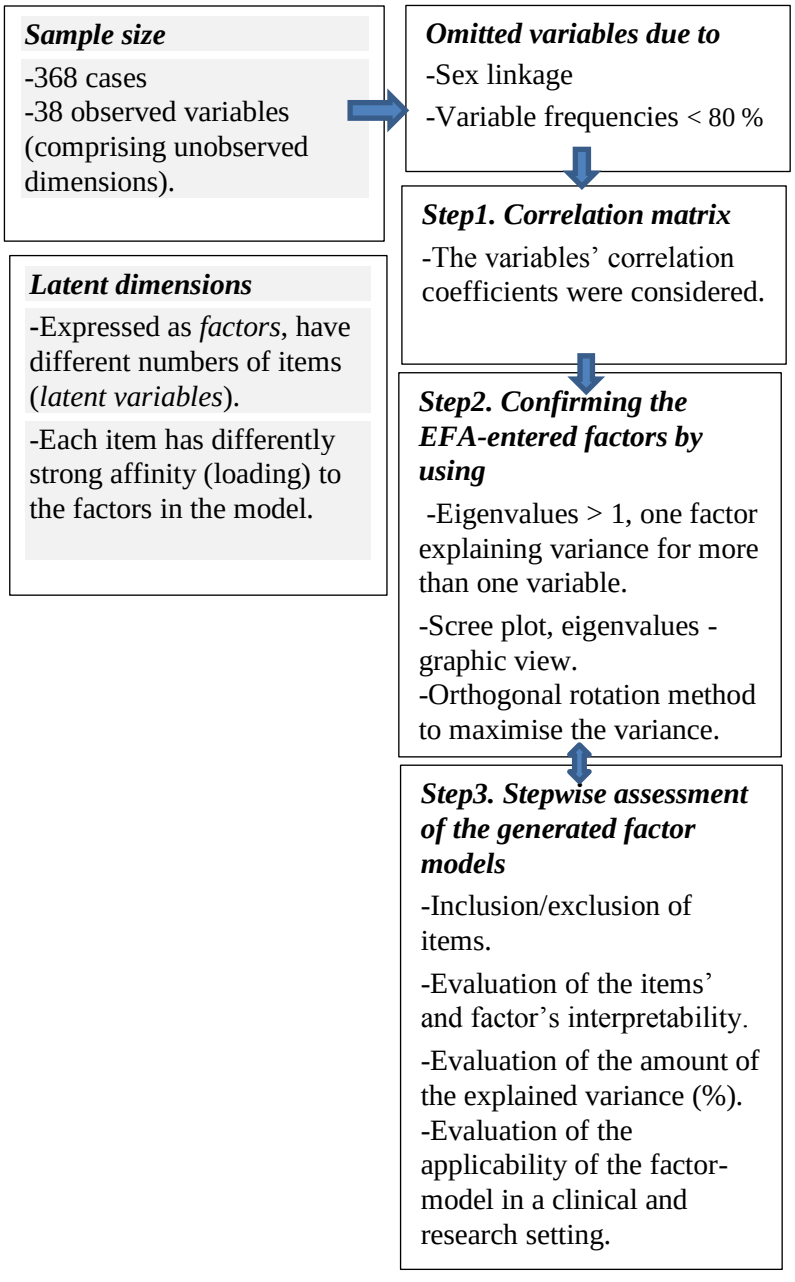

Figure 1. *The EFA as applied to the questionnaire variables.*

Paper III. The Mann-Whitney test was used for the unpaired comparison of the DDQ between the D and C groups, as well as between the D subgroups. The Mann-Whitney test was used to analyse the DDQ in relation to dietary and oral hygiene habits in the D group and C group, respectively. The McNemar test was used for the paired comparison regarding oral hygiene and dietary habits. The Wilcoxon signed-rank test was used for all paired comparisons between the D group and the C group regarding the DDQ and DMFT. The Kruskal-Wallis test and Mann-Whitney test were used to analyse the grouped DMFT in relation to the DDQ of the D group.

Paper IV. Descriptive statistics were used to present the main data for the 37 items, divided into categories A-E. Regarding these categories, the Chi-square test was performed to analyse relationships in the distribution of sex, age and years of professional experience, the proportion of working time devoted to treating children and adolescents and being a parent. One-way analysis of variance (ANOVA) was applied to the variables of sex, age, years of professional experience, proportion of working time devoted to treating children and adolescents and being a parent (regarding A-E). To adjust for multiple comparisons, a correction of the significant level was made according to the Bonferroni-Holm technique. Due to missing responses on different items, the numbers of analysed individuals vary in the tables (Paper IV).

3.5 Ethical approval and considerations (Paper I-IV)

Ethical consent was granted by the Ethical Research Committee at the University of Gothenburg, reference numbers: S 663-02 (Paper I, II) and 208-10 (Paper III). No ethical approval was required for Paper IV, as it was performed anonymously on dentists. Informed consent was obtained from all eligible groups of children and adolescents as well as from legal guardians (Paper I-III). Information on the aim and performance of the study was given individually, both verbally (Paper I, IV) and in writing (Paper I, III, IV). In addition, the informants were asked for their permission to access their electronic dental records at the PDS clinic responsible for their dental care (Paper III). The informants were guaranteed that only the researchers would be given access to names and addresses and questionnaire data (Paper I-IV).

4 RESULTS

In *Paper I,* questionnaires from 368/383 participants (8-19 years old) without disabilities, equally distributed between genders, were analysed.
In *Paper II,* variables from the questionnaire data gathered in *Paper I* were analysed.
In *Paper III*, 142/188 (57 girls and 85 boys) with disabilities, and 135/188 sex and age-matched controls (aged 12-18 years) were analysed.
In *Paper IV*, questionnaires from 387/567 general dentists (116 male and 271 female) were analysed (Table 3 in Method, p. 29).

4.1 Reliability and internal consistency

CPI (Paper I, II)

Fourteen children, aged 8-14 years, filled in the CPI twice, with one week apart. Spearman's correlation demonstrated good reliability, 0.72 ($P < 0.01$) in the test-retest setting regarding the item '*Fallen and scraped the skin'*, which all the children in this group had experienced (Paper I).
Thirty-two children and adolescents, 16-19 years old, filled in the questionnaire twice, with one week apart. The Cohen's Kappa coefficients for the everyday pain items ranged from 0.65-0.91, and for the dental pain items from 0.68-1.0. The test-retest with the Intraclass Correlation Coefficient (ICC) yielded a result of 0.95 (95 %, CI 0.85-0.98) (Paper I).
The internal consistency of the CPI items was established for the most frequently reported everyday pain (78 participants); Cronbach's alpha was 0.88 (Paper I).
For the short CPI, tested on 30 patients (18 boys, 12 girls), aged 14.4 years (SD 1.7), Cronbach's alpha was 0.51 (Paper II).

DDQ (Paper III)

Twenty-three parents of children without disabilities (aged 2-13 years, median age 8.4) filled in the DDQ questionnaire twice, with one week apart. Cohen's Kappa was 0.63. The ICC was 0.85 (95 %, CI 0.67-0.96).

Dentists' Knowledge and Attitudes to children's pain perception, DKA-CPP form (Paper IV)

A test-rest performed on ten general dentists showed the ICC for all items to vary between 0.97 and 0.99. The corresponding Spearman correlations varied between 0.7 and 0.96.

4.2 Everyday and dental treatment pain, and associated factors reported by children and adolescents without disabilities (Paper I, II)

4.2.1 Occurrence of everyday and dental treatment events

There were considerable individual differences between participants, with regard to whether the everyday and dental treatment pain events of the CPI had been experienced or not. The most frequently experienced CPI items were '*Fallen and scraped the skin*' (N = 356) '*Had a cold*' (N = 350) and '*Bitten tongue*' (N = 352). The most frequently experienced dental treatment items were '*Dental X-ray*' (N = 346; 94 %), and '*Had the teeth probe-checked*' (N = 307; 83 %). The least common item was '*Tooth extracted*' (N = 135; 37 %), (Table 4).

4.2.2 Pain experience by encountering everyday and dental treatment events

The children and adolescents reported differently if an event, when encountered, had been painfully experienced (Table 4). The most frequently reported pain (> 90 %) among the everyday events was '*Headache*' and '*Stomach/tummy ache*'. The most common everyday events were reported as painful equally often by girls and boys. The dental treatment event most often experienced as painful was '*Dental injection*' (Table 4).

TABELL 4

4.2.3 Influence of age on everyday and dental pain occurrence and intensity

There was large variation in pain experiences. When age was dichotomised (median split), the older group (14-19-year-olds) reported more pain experiences compared with the younger (8-13-year-olds) participants. The younger group commonly rated pain intensity higher than the older group.

4.2.4 Influence of gender on everyday and dental treatment pain occurrence and intensity

There was a general trend for girls to report pain more often when encountering the CPI events. For some of the less common everyday events, statistically significant differences were observed between boys and girls. Typically, these events were characterised by *blunt trauma*, or a *cut through the skin/mucosa injury/procedure*. '*Dental injection'* was the only dental treatment event that girls reported as painful more frequently than boys (Table 4).

In general, pain intensity was reported as higher, as measured on the VAS, for the everyday events than for the dental treatment events. Girls generally reported higher pain intensity for all events. For some of the events there was a statistically significant difference, such as, '*Headache*' and '*Vaccination*', as well as '*Bitten tongue*' (Table 5). The dental treatment events rated with the highest pain intensity were equally rated by girls and boys.

TABELL5

4.2.5 Influence of dental anxiety on the occurrence and intensity of everyday pain and dental treatment pain

A median split of the DAS scores (median DAS = 5.7) was used to create two groups: *Low* and *High dental anxiety*, respectively.
In general, children and adolescents in the *High dental anxiety* group reported higher levels of pain intensity.

4.3 Short CPI model (Paper II)

CPI items that were experienced by > 80 % of the respondents were chosen to enter the Exploratory Factor Analysis (EFA). These 18 items were reduced to twelve items, of which two were dental treatment-related. The twelve items formed four factors, each representing a distinctive topic area. Each factor was clearly defined and well held together by the clustered item-loadings with good affinity, well above 0.5 (Figure 2). After discussion and interpretation of the clustered items and their shared meaning, each factor 1-4 was termed as shown in Figure 2.

Factor 1 Cutting trauma to skin/mucosal pain ***Item/loading***	**Factor 2** Head/neck pain ***Item/loading***	**Factor 3** Tenderness/blunt trauma pain ***Item/loading***	**Factor 4** Oral/dental treatment pain ***Item/loading***
Scraped the skin **0.894**	*Had a sore throat* **0.865**	*Got a bruise* **0.939**	*Dental injection* **0.799**
Got a sliver **0.846**	*Had a cold* **0.851**	*Stomach/tummy ache* **0.695**	*Dental X-ray* **0.795**
Bitten tongue **0.759**	*Had a headache* **0.751**	*Stubbed toe* **0.526**	
Got a paper cut **0.692**			

Figure 2*. A short CPI model, resulting from the Exploratory Factor Analysis, with four factors and twelve items. The items building each factor are shown together with their loadings (affinity to a factor*).

4.4 Oral pain and discomfort in children with disabilities versus children without disabilities (Paper III)

A total of 188 children and adolescents, 12-18 years old (mean age 14.6 years) with a disability (D group) agreed to participate in the survey. Due to incomplete questionnaires or dental records, 142 could be analysed. For the same reasons, 135/188 of the sex and age-matched controls without a disability (C group) could be analysed. The main oral problems in the D group were clustered around items representing 'C*hewing*', '*Biting off'* and '*Brushing the teeth'* (Table 6).

TABELL 6

Dental discomfort and pain, as measured by the total mean DDQ, was statistically significantly higher in the D group (3.2, SD 2.9, range 0-14) compared with the C group (1.6, SD 2.0, range 0-10), using the Mann-Whitney U test (P = 0.001). The same difference was found in the paired analysis using the Wilcoxon signed-rank test (P = 0.000) (Table 7).

Among the D subgroups, the highest mean DDQ score of 4.8 (SD 4.2) was observed for the children with severe intellectual difficulties. This was statistically significantly higher compared with the group with a physical disability 2.2 (SD 2.1), using the Mann-Whitney test (P = 0.012) (Table 7).

A similar difference was found for the mean DDQ score of 4.8 (SD 4.2) for the children with the most severe intellectual disability compared with the youngest children with a mild intellectual disability 2.4 (SD 2.9), using the Mann-Whitney test (P = 0.034) (Table 7).

TABELL 7

4.4.1 Dental health, DMFT in relation to DDQ

Both the D group and the C group had a similarly low DMFT mean of 1.6 (SD 2.1) and 1.5 (SD 2.1), respectively, without a statistically significant difference when the Wilcoxon signed-rank test was applied to the 135 pairs. No differences in DMFT means were observed across the D subgroups.

The DDQ mean for DMFT = 0 did not differ significantly in comparison with the DDQ mean for DMFT ≥ 1. DMFT = 0: DDQ mean 3.0 (SD 3.4), median 2; DMFT ≥ 1: DDQ mean 3.4 (SD 2.7), median 3) with the Mann-Whitney U test.

4.4.2 Oral hygiene and dietary habits

Differences were observed for the D group compared with the C group regarding oral hygiene and dietary habits.

The D group reported not brushing the teeth twice/day (27 %) as often as the C group (62 %) (P = 0.000, paired analyses). More participants in the D group preferred to brush their teeth in the morning (77 %) compared with the C (65 %) group (P = 0.038).

Both the D group and the C group had a similar frequency of intake of sweet snacks/soft drinks between meals (paired analyses), with no statistically significant difference.

The D group reported eating and drinking > 5 times/day less frequently than the C group; 27 % and 52 %, respectively (P < 0.0002).

4.5 Dentists' attitudes to and knowledge about pain and pain management in children and adolescents (Paper IV)

Of the dentists, 50.1 % were < 46 years of age. The mean number of professional years of experience for the dentists was 18.3 years (SD 13.4, median 17.0 years). More than 50 % of the male dentists had 0-25 % of their working time allocated to treating young patients. The corresponding figure for the female dentists was 26-50 %.

4.5.1 Factors influencing the attitudes and knowledge of dentists

There were differences in attitudes and knowledge related to the dentists' age, gender and years of professional experience, measured as Knowledge and Attitudes of Dentists to Child Pain Perception (DKA-CPP). A dentist

who was either ≥ 46 years, female, or had ≥ 17 years of experience was more likely to prevent or treat the young patient's pain. Examples of items with statistically significant differences due to the age, gender or professional experience of the dentist are presented in Table 8.

Table 8. *Knowledge and Attitudes of Dentists to Child Pain Perception (DKA-CPP). Examples of items with statistically significant differences, p < 0.05, based on the dentist's age < 46 / ≥46 (years), professional experience < 17 / ≥ 17 (years in practice) and gender, F = female, M = Male. Means, SD, and 95 % confidence intervals (CI) are presented. ANOVA, original P values (P-o) and P values after Bonferroni-Holm adjustment (P-a). Score 1 = low level of knowledge, score 5 = high level of knowledge.*

Item A1. *Children usually tolerate pain better than adults*

Age	n	Mean	SD	95 % CI	P-o	P-a
< 46 years	192	4.3	1.0	4.2; 4.5	0.0001	0.002
≥ 46 years	190	4.6	0.8	4.5; 4.8		
Total	382	4.5	0.9	4.4; 4.6		

Item B1. *Untreated pain prolongs the time of healing/recovery until the onset of pain relief*

Experience	n	Mean	SD	95 % CI	P-o	P-a
< 17 years	188	4.1	0.9	4.0; 4.2	0.001	0.03
≥ 17 years	191	4.4	0.8	4.3; 4.5		
Total	379	4.3	0.9	4.2; 4.3		

Item C4. *Usually, the child's pain experience diminishes when a parent is present*

Gender	n	Mean	SD	95%CI	P-o	P-a
F	268	3.7	1.1	3.5; 3.8	0.00001	0.0004
M	114	3.1	1.1	2.9; 3.3		
Total	382	3.5	1.1	3.4; 3.6		

5 DISCUSSION

This thesis found that the younger children, children with Dental Fear and Anxiety (DFA) and girls reported everyday pain and pain from dental treatments more frequently. Children with disabilities (D) had statistically significantly more oral discomfort and pain than their sex and age-matched controls (C). The difference could not be explained by the children's caries prevalence.

The thesis also found that dentists with long professional experience and/or female dentists had more knowledge on pain and pain management. It seems plausible that these dentists have a better understanding of and can offer better support for the management of pain in children.

The studies, taken together, demonstrate the significance of recognising pain in dental settings, from the perspective of the child and the dentist. The results suggest that dentists should routinely seek to identify the most vulnerable children with regard to pain experiences, and practice pain prevention.

5.1 Methodological discussion

Before the performance of each survey, possible methods were identified for the data collection on oral and general pain occurrence and pain intensity (Paper I, III) (45, 64, 132). A reasonable option would have been a qualitative study design; the in-depth interview. This method has shown good results when used on adolescents and adults for deeper exploration and understanding of the field of study (133, 134). If such a methodology had been selected, a small number of patients would have been enough to provide extensive knowledge of children's pain experiences.

Another possible study design would have been the structured interview method, as practiced by McGrath et al., 2000, on the pain experiences of 5-16-year-olds (with/without medical conditions) (89).

However, to explore the occurrence and intensity of pain and oral discomfort variables, the questionnaire was considered the most suitable method for the collection of data (Paper I, III). In this way, it was expected that larger groups of children (with or without a disability) could be reached, allowing for the generation of normative data for the studied variables. A further reason to use questionnaires was that there were already validated forms for these purposes; the Children's Pain Inventory (CPI) and the Dental Discomfort Questionnaire (DDQ) (52, 89). There was also a questionnaire for the study of the knowledge and attitudes to pain

and pain management in children among medical professionals (135). This questionnaire could be modified for use among dentists (DKA-CPP) (Paper IV).
Each questionnaire was a part of a cross-sectional study design; that is, the respondents provided their answers at a single point of time (Paper I, III and IV). The surveys were furthermore descriptive and could point out relationships, and were indicative of influential factors (on the studied variables) (Paper I, III, IV).

The statistical method Explorative Factor Analysis was considered the only instrument able to reduce the number of CPI questions and enhance the form's quantitative and qualitative properties (Paper II).

All questionnaires used in this thesis revealed solid psychometric properties, such as face and construct validity and reliability, as supported by previous studies (I, III, IV) (99, 101, 135, 136). Furthermore, the child's linguistic understanding was assured by translation back and forth of each pilot study (Paper I, III).

5.2 General discussion

Dental professionals working with children should be well informed about children's most common pain experiences and offer them up-to-date help when needed. The CPI questionnaire provided significant information on the occurrence of everyday and dental pain experiences among the studied children and adolescents without disabilities. The frequencies and intensity of pain were in accordance with those of other populations, which confirmed the hypothesis (49, 52, 89, 107). However, at an individual level there was a wide variety of experienced CPI events. The results most probably reflected the respondents' age range of 11 years (8-19 years).
More than 90 % of the studied children reported having experienced '*Headache'* and '*Stomach ache'*. The outcome was supported by McGrath's CPI reports of > 80 % for both events (89). Also, studies into children's health issues presented a high occurrence of headache and stomach ache (37, 89, 137-141). Some of the frequently occurring conditions might explain the increasing psychosomatic problems among children and adolescents (manifesting as tiredness, loss of appetite, emotional distress or depression) observed in the last decades (37, 51, 137-139). The prevalence in the reports fluctuates in accordance with the design of the questions. For example, having headache and/or abdominal pain 'a few times a month' was reported by > 40 % of schoolchildren in

grade 3-9 (37). Furthermore, the risk of acute pain turning into long-standing pain has been highlighted. Perquin et al., 2000, described the prevalence of long-standing pain (headache, limb pain and abdominal pain) as increasing with age among 4-18-year-olds (51). This was especially observed in girls 12-14 years old (51). How these children will be affected in the long term is not known. However, it is important that dentists are informed about children's health circumstances in order to approach therapy planning holistically.

The CPI furthermore indicated that 66 % of the 8-19-year-olds had at some point experienced '*Toothache*'. Various conditions originating from, for example, the shedding of teeth, sensitive teeth or dentine caries might be the explanation. However, there are no available data on untreated carious and consequent pain conditions among young Swedish children. Levine et al., 2002 studied 1,409 unrestored carious deciduous teeth retrospectively. Eighteen per cent of these were recognised as provoking pain sensations. The carious teeth most likely to trigger pain were the molars with larger cavities presenting shortly after eruption (142). Colak et al., 2013, summarised the longstanding consequences of untreated caries as pain, disturbed sleep, infections, and the child not thriving, among other problems (143). Early detection of dentine caries in young children should aim at preventing the development of painful conditions and the need for invasive treatment with the potential to inflict pain.

Dental treatment procedures

Children often fear pain experiences in connection with dental conditions and dental treatment. However, there are very few studies with the immediate aim of observing which dental treatments children and adolescents perceive as painful. The most substantial knowledge on painful dental treatment originates from studies on the origin of DFA (64, 65, 144).

Between 56 % and 69 % of the studied children and adolescents reported that they had experienced the CPI dental treatment items of '*Tooth extracted*', '*Tooth drilled*', *Tooth restored*', '*Dental injection*' as painful. Accordingly, in a longitudinal Swedish study, BITA, performed over five years, 60 % of the children reported *extraction*s to be painful. Forty per cent had perceived pain during *tooth restoration* (Ghanei & Robertson in manuscript). Bergius et al., 1997, also found that 59 % of a group of 13-18-year-olds had painful experiences from the corresponding dental procedures (107, 145).

The child that experiences pain in the dental setting may subsequently develop DFA, begin to avoid dental appointments, and eventually develop poorer dental health (65, 146, 147). The risk of this negative development indicates the importance of painless dental treatment. To ensure such an outcome, the use of topical analgesia is mandatory before administering anaesthesia. However, Wondimu et al., 2005, found that only 48 % of the dentists used topical analgesia before an injection (79). The fact that children reported restorative treatment as painful could indicate insufficient administration of anaesthesia (with regard to technical performance, onset, or amount).

Children with already developed DFA or behaviour management problems (BMP) due to painful dental treatment experiences would need to relearn dental procedures as more positive experiences (68). Jälevik et al., 2002, found that children with hypomineralised first molars had undergone repeated painful dental restorations, often without being given anaesthesia (148). These children had significantly more DFA than the controls (148).

To help children with DFA rebuild their trust in dental care, it is necessary to ensure proper pain management and comfort. An additional measure is to offer sedatives and administration of analgesics in connection, for example, with tooth extractions. However, a Cochrane review carried out in 2016 could not determine 'whether or not preoperative analgesics are of benefit in paediatric dentistry for procedures under local anaesthetic' (149). The uncertainties of the review were due to methodological study differences. However, the dentist should use all available means to achieve optimum pain management in every child.

Non-invasive dental procedures are occasionally also reported to be painful. Thirty per cent of the studied children who had experienced the oral CPI item '*X-ray*' reported the procedure as painful. The explanation could be that radiographic plates could be perceived as being too big and hard, creating discomfort or pain of the palate or the floor of the mouth. In a sample of 289 five-year-olds, eleven children could not cooperate sufficiently in connection with bitewing radiography and were excluded from further participation (150). It is not known whether the cooperative five-year-olds perceived discomfort or pain.

With regard to the studied CPI dental treatment procedures, the child may perceive any oral intervention as painful in the wrong situational circumstances. For this reason, dentists need to perform well-practiced invasive and non-invasive treatments.

Age and sex as influential factors

Dentists treating young patients generally take care of both preschool children and adolescents. They thus need to relate to the characteristics of children's various cognitional and socio-emotional developmental levels (151, 152). The dentist also has to be familiar with how to manage children's stress, such as DFA, discomfort or pain experiences (151-153). Altogether, age has been shown to be naturally associated with human abilities and experiences and is therefore a frequently studied variable.

The CPI reports also show that age influences the occurrence and intensity of pain perception, which was supported by previous findings (49, 52, 89).The children younger than 14 years had experienced fewer CPI events; however, these children reported CPI events as more painful than the older children. The children's different levels of intellectual maturity and ability to classify and express pain may explain this difference (154). For example, Vlok et al., 2011 found that 6-10-year-olds had very different perceptions from 18-24-year-olds as to which dental injury would hurt the most, when viewing dental trauma pictures (154). McGrath has described children's pain experiences as matching their age and corresponding intellectual and physical skills (49). The CPI outcomes indicate that dentists should strive to make a correct assessment of the child's intellectual and emotional level and meet his/her individual capability when introducing painful dental procedures. The CPI findings indirectly also require an accurate assessment of pain in children. Such an assessment would warrant adequate pain management, which is also a part of treatment success (155).

On the whole, the studied girls reported the everyday CPI events as painful more often than the boys (even if not always statistically significantly higher). In a review of 14 populations, Klingberg & Broberg, 2007, described ten populations that reported more dental anxiety in girls than in boys. (68). In general, studies indicate that girls more frequently display DFA and BMP. The relationship between DFA and BMP and girls' general fear (and internalisation and externalisation of behaviour problems) might explain, to some extent, the differences between the sexes.

Interestingly, the studied girls rated the pain intensity as higher for some events. It appeared that items related to 'sharp objects' ('probe', 'bee⁄wasp sting', 'knife', and 'vaccination') were prevalent among the items scored higher by girls. This result also points to greater vulnerability to pain among girls.

Regarding the dental treatment events, '*Dental injection*' was statistically significantly more often reported as painful by the girls. Many studies have concluded that girls and boys experience pain differently, although there are also contradictory results. Biological and physiological differences have been shown (56, 156-158). A meta-analysis of pooled data on experimental pain among healthy boys and girls found that the girls reported significantly higher cold pressor pain intensity compared with the boys (156). The meta-analysis of heat pain also concluded that boys had significantly higher tolerance than girls (156). It also suggested that girls are indirectly expected to express their experiences more than the boys in the psychosocial communication (159). It is, however, important that the dentist does not interpret the results as boys being in less need of pain-preventive efforts than girls. There may be many intervening processes in play that result in the individual's willingness to report or not report pain.

The time factor as such has not been studied in this thesis. However, the dental treatment CPI results suggest that time should be allocated for communication between the child and the dentist. This would probably help the child to build interpersonal trust and to cooperate better in the dental setting (160-165). A current Swedish survey analysed the methods and needs of paediatric nurses during their interaction with the children (166). The main conclusion was that 'Health care organisations must earmark time to allow important communication to take place between staff and paediatric patients so that children and families feel safe when being treated' (166). This reasoning could be transposed to the dental care system, in order to give children the right prerequisites to understand and manage pain experiences (80).

Still, even now, acute and postoperative pain constitutes a problem for children in the Western world (167, 168). Surveys have shown that one third of surgical and medical in-patients usually had pain before their admission, and that 77 % experienced pain during the admission (167). Since the 1990s, studies have also indicated that not all children received enough pain alleviation or any at all in the dental setting (77-80).

The amount of treatment time has also been recognised as significant for the approach to pain alleviation in children (167). The dentist's systematic documentation of pain gives an overview of how the child's pain emerges. The documentation is an attempt to improve the planning for the alleviation of unnecessary pain and suffering (127, 169).

Dental Fear and Anxiety as influential factors

Invasive dental procedures, such as injections and tooth extractions, have been considered major risks for the development of DFA and consequent treatment failure (64, 107, 132, 170). For this reason, it seemed obvious to look at how the DFA varied with the CPI. Children with higher DFA reported statistically significantly higher pain intensity regarding many everyday events such as '*Toothache*' and '*Vaccination*'. All invasive dental treatments: '*Tooth extracted*', '*Tooth drilled*', *Tooth restored*', and '*Dental injection*', were also statistically significantly rated as more painful by the group with higher DFA. Bergius et al., 1997, also found children (13-18 years of age) with higher DFA to have had more painful dental treatments (107).

Because of the relationship found between DFA, pain and pain intensity, it is suggested that dentists should be more observant and communicative on these matters. As proposed by Rantavuori et al., 2008, pain and DFA could be regarded as an entity (in the individual case), until otherwise proven (171).

The CPI event, '*Braces tightened,*' was given similar scores by the children with lower and higher DFA. This might indicate that the children undergoing orthodontic treatment are often highly motivated to endure discomfort. However, Campos et al., 2013, (172) found no relationship between the motivation of a child to undergo orthodontic treatment and the reported intensity of pain. It has furthermore been shown that the greatest concern of children and parents with regard to orthodontic treatment was the anticipation of pain (173).

Disability and oral discomfort and pain

The participating children with disabilities (D) had various individual functioning difficulties. To group the children into intellectual and physical disability groups was one way to handle the research methodology and make comparisons with controls (Paper III). The D children experienced statistically significantly more oral discomfort and pain compared with their sex and age-matched controls, which confirmed the hypothesis (C). However, a direct explanation could not be found.

Also, the D children's most frequently reported oral problems were concentrated to the areas of chewing, bite-off and tooth-brushing, as shown by the Dental Discomfort Questionnaire (DDQ) (Paper III). The results of Versloot et al., 2006, highlighted the same problems as the most challenging for young children (without disabilities) with toothache (101).

Daher et al., 2014, also found children with caries and toothache to have problems with eating and tooth-brushing (136).

The D children with the lowest intellectual capacity showed a higher DDQ score than the children with a higher intellectual ability (and those with a physical disability). The DDQ score difference may be a result of oral motor difficulties among some D children, resulting in naturally altered chewing patterns. However, children with disabilities may also have complex medical problems, for instance, recurrent reflux (originating from gastrointestinal conditions) (174). This could trigger greater oral sensitivity (174). The oral and the medical condition could influence each other in a negative way.

Some additional medical conditions could probably further explain why the children with the lowest intellectual capacity had the highest DDQ score. However, there are probably also more complex explanations. It has been reported, for instance, that families taking care of a child with a disability have more frequent additional medical and other professional contacts concerning the care of the child (175). A vicious circle is perhaps more readily initiated in children with a very low intellectual functioning level and complex medical conditions. They may have more frequent oral motor dysfunctions and suffer greater oral sensitivity, and underlying environmental challenges may also play a role. Such circumstances could be the cause of enhanced irritability and vulnerability throughout repeated incidents during the day (33). Furthermore, the parents of the D children may have interpreted this as oral discomfort or pain.

Underreporting of oral problems (such as teeth eruption or exfoliation as well as sensitive teeth) could also be suspected among the C group participants. They may have interpreted their oral problems as being too trivial or transitional, and not significant enough to be communicated.

Oral discomfort and oral health

The second hypothesis put forward in Paper III was that children and adolescents with intellectual or physical disabilities are expected to have more dental caries. Commonly, these groups of children present a more severe oral pathology related to different intrinsic and extrinsic circumstances (176).

In the D group, dental caries DMFT received a low score, similar to that of the C group. The higher DDQ in the D group could thus not be explained by caries-related discomfort or pain symptoms.

Reports on dental caries in children with disabilities have been inconsistent regarding different physical and intellectual disabilities. Dieguez-Perez et al., 2016, evaluated 14 articles in order to analyse oral health in children with an intellectual or physical disability (176). The authors looked for dental caries, oral hygiene, gingival health, malocclusion and habits (concerning children with cerebral palsy (CP), representing a physical disability, and Down syndrome (DS), representing an intellectual disability), compared with controls (176). It was subsequently recognised that there was no consensus among the revised authors regarding dental caries, oral hygiene and gingival health (often due to methodological differences). These data support the low DMFT score in the studied children with intellectual and physical disabilities.

In the same review, Dieguez-Perez et al., 2016, further concluded that oral hygiene was generally poorer among DS and CP children than among controls. Gingival health was also found to be worse in both the DS and CP children compared with the controls. This finding might be consistent with the fact that the oral hygiene habits in the studied D group were poorer than in the C group. The DDQ score of the D group did not show a relationship with the DMFT score, nor with the frequency of oral hygiene performance. However, the D group did not undergo a clinical examination and for this reason, no comparison with clinical data can be made.

The high DDQ score in children with physical and intellectual disabilities indicates that these groups are more exposed to aggravating intrinsic and extrinsic factors. They therefore need early, regular and customised observations by the dental health service. In order to enhance the oral wellbeing among children with a disability, it is desirable to adopt a holistic approach and to increase the oral health efforts for these children.

Knowledge and Attitudes of Dentists to pain

Dentists are responsible for helping a child in stress; for example, when an invasive procedure such as a dental injection is performed. Relatively few studies have looked at the knowledge among dentists about pain and pain management in children. The responses to the self-report questionnaire, Dentists' Knowledge and Attitudes to Children's Pain Perception (DKA-CPP), showed that ≥ 17 years of professional experience indicated better knowledge about pain and pain management. This was shown, for example, in the response to the statement: '*It is acceptable to treat a permanent tooth without any pain alleviation'*. Correspondingly, Ronneberg et al., 2015, observed that dentists who had worked less than

ten years reported more difficulties performing restorative treatment. These dentists also used local anaesthesia and conscious sedation less frequently in children below ten years of age (177). Accordingly, Wondimu et al., 2005, found that only 36 % of the dentists always used local anaesthesia during restorative treatment (79).

It has been discussed whether the dentist's choice not to give local anaesthesia could be related to insecurity when dealing with a demanding situation (178). Davidovich et al., 2015, analysed self-reported stress experienced by general and paediatric dentists when treating children. A main conclusion was that the professionals experienced the most stress when performing dental injections on anxious children (178). The dentist's age, sex, or years of professional experience were not significant for the amount of stress they felt in a given situation (178). Additionally, Ronneberg et al., 2015, found that 60 % of dentists never, rarely or sometimes used local anaesthesia in children aged 3–5 years, whereas 30 % of dentists used no local anaesthesia when performing restorative treatment on patients in the group 6–9 years old (177).

The DKA-CPP indicated that the responding dentist's age was related to better knowledge about children's pain. To some degree, this must be due to the time allocated to enhancing their experience and knowledge through practice. Similarly, Enskär et al., 2007, showed that nurses with longer work experience made greater contributions to the child in pain (although this was contradicted by other studies) (135, 179).

The present study suggested that female dentists had greater knowledge about pain and pain management in children (compared with male dentists). In contrast, Wondimu et al., 2005, found a significant association between female dentists (41-50 years old) and a tendency to attach less importance to pain-free dental treatment (79). Most likely, both results describe the studied dentists' characteristics. As knowledge and attitudes are compound and dynamic phenomena, future studies will probably also show varying outcomes. However, in order to ensure the best possible quality of dental care for children, recurrent surveys on knowledge and attitudes are significant.

Finally, dentists treating children face a work situation that is very complex and demanding. It requires, for example, adequate knowledge and attitudes to pain management, as well as a pedagogical and psychological approach to the individual child's needs.

Advantages of the extended and reduced CPI

The extensive CPI questionnaire had the advantage of covering a subject area and also producing normative data when applied to children without disabilities (Paper I). However, the 38-question form could be challenging for children to respond to and a short CPI form with twelve items was therefore developed (Paper II). Through the data processing of the most frequent CPI events, the aim to identify and retain the most representative CPI items among the studied children was achieved. The results indicate that processing with Exploratory Factor Analysis (EFA) can ensure the quantitative and qualitative information of the reduced form.

When comparing the original CPI form with the reduced form, it appears that both have advantages and disadvantages. The research question and the aim of the study should therefore be reflected on before choosing the most appropriate measuring instrument of the two. It was concluded that the new CPI model, arranged into four factors, including a dental factor that consisted of the items '*Dental X-ray*' and '*Dental injection*', was the most logical.

Clinical experience has recognised the importance of discomfort and pain inflicted by '*Dental X-ray*' or '*Dental injection*', and the risk of initiating DFA (89, 107, 180, 181). Another important finding that justified the specific EFA model was the high percentage (79 %) of explained variance among the processed items. In conclusion, it could be argued that both the CPI (Paper I) and the reduced CPI form (Paper II) could serve different purposes in the future, when applied to different clinical and research settings.

Strengths and limitations

The strength of the present studies was that they together form a coherent study into children's general, oral and dental pain experiences. The studies recognised influential internal and external factors, such as age, sex, DFA and disability. The dentist's knowledge and attitudes to pain in children were highlighted, which is significant for a successful treatment outcome. A possible methodological challenge was that the CPI, DDQ and DKA-CPP forms were applied for the first time to a young Swedish population and general dentists. However, they contributed to greater knowledge and were supported by previous outcomes (89, 101, 135).

In *Paper I,* the strength was that the CPI applies a holistic view to children's everyday and dental treatment experiences (89).

In *Paper II*, the strength was the development of a short and more user-friendly CPI form, holding only twelve items. A further advantage was that a dental factor including '*Dental injection*' and '*Dental X-ray',* occurred.

In *Paper III*, a limitation was that proxy reports, as opposed to self-reporting, were used. A strength was the focus on the children and adolescents with disabilities, addressing their oral discomfort and pain, as well as their status and extended needs.

In *Paper IV*, a strength was the multidimensional nature of the DKA-CPP instrument, with several subject categories. A limitation was that the DKA-CPP comprised 56 items, which may have been experienced as too long by the respondents.

Ethical considerations

The children's situation, the fact that they are exposed to the knowledge and ability of their surroundings was recognised throughout the research process (Paper I, III, IV). However, because of their dependent position in the adult domain, it was considered important to let them speak about their own situation.

To involve children in research is ethically and methodologically challenging and includes various areas for reflection. In particular, it should be emphasised that children are not capable of making decisions on their own. However, as all efforts were made to comply with the existing ethical standards, the studies were considered justified, especially since they have promoted the children's voices (through the extended knowledge gained about children's oral pain and discomfort experiences) (Paper I, III).

The methodological processing of the CPI form was regarded as suitable for future clinical studies and may promote further knowledge on children's pain experiences (Paper II).

The survey of the general dentists may lead to enhanced awareness at an organisational level and to extended education being offered (Paper IV).

6 CONCLUSION

General and dental treatment pain and oral discomfort were considered from the point of view of the child (with or without a disability), the legal guardian and the dentist.

This thesis contributes to a deeper understanding of children's pain experience with regard to frequency and intensity. The results indicate intrinsic and extrinsic factors that may influence the pain experience and also highlight relationships. This thesis may be useful for dentists treating children and adolescents.

The main conclusions from the four articles are:
Younger children, girls and children with higher dental anxiety (8-19-year-olds) experienced everyday and dental treatment-related pain more frequently and occasionally with greater intensity (as measured by the CPI).

The original CPI with 38 questions and the reduced CPI with twelve questions were both found to have robust psychometric properties. In the future, they may be used for different purposes, based on the research question and the aim.

Children with an intellectual or physical disability (D) experienced oral discomfort and pain (DDQ) more frequently than their matched controls (C) (12-18-year-olds), despite the good dental health in both the D and C groups.

Dentists with more professional practice, older dentists, and female dentists applied more pain management strategies.

7 FUTURE PERSPECTIVES

The findings suggest that dentists should attempt to identify children who can be expected to undergo frequent invasive dental procedures. Furthermore, dentists should always aim to offer all children painless dental experiences.

Children with a limited performance capacity and communication difficulties should also be recognised, because of the increased risk of a negative dental treatment experience.

Furthermore, the findings suggest that dentists should routinely include an overview of the child's prior and current pain experiences in the anamnesis. Adequate information could include hospitalisations, experiences of invasive medical and dental procedures, vaccinations and the child's own idea of being susceptible to pain. Simple questions could be put to the child: *'Are you worried that the treatment will hurt'*, and *'Does it hurt?'*

The findings further suggest that dentists should pay attention to the technical performance of the treatment procedures, such as dental anesthesia, dental extraction and restorative therapies.

Also, the findings indirectly point to the significance of continuous preventive dental health measures from a very young age. With this approach, children will not be subjected to unnecessary invasive dental treatment, resulting in a risk of experiencing dental pain. To support such a development, efforts could be made to provide education for dentists who treat children. The updates should aim to deepen the insight into the child's different functions, reduced autonomy and distinctive needs.

A more comprehensive effort to convert the results into fruitful development would be the setting up of a national network, including dental educational entities, to create an on-going dialogue/consensus regarding acute dental-related pain issues in the young patient.

Finally, this thesis also shows that insufficient research has been carried out into pain and pain management in children and that extensive efforts should be made to acquire more knowledge.

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APPENDIX