# Children's Oral Health Related Quality of Life

Determinants related to subjective and objective orthodontic measures

**Lea Kragt- de Roos**

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## Determinants related to subjective and objective orthodontic measures

## Mondgezondheid gerelateerde levenskwaliteit van kinderen
## Determinanten gerelateerd aan subjectieve en objectieve orthodontische metingen

**Thesis**

to obtain the degree of Doctor from the
Erasmus University Rotterdam
by command of the
Rector Magnificus

Prof.dr. H.A.P. Pols

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by

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**Erasmus University Rotterdam**

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Strahinja Vucic

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# Manuscripts that form the basis of this thesis

## Chapter 2

**Kragt L**, Tiemeier H, Wolvius EB, Ongkosuwito EM. Measuring oral health-related quality of life in orthodontic patients with a short version of the Child Oral Health Impact Profile (COHIP).
*Journal of Public Health Dentistry, 2016, 76, 105-112*

**Kragt L,** Hermus AM, Wolvius EB, Ongkosuwito EM. Three-dimensional photographs for determining the Index of Orthodontic Treatment Need in scientific studies.
*American Journal of orthodontics and dentofacial orthopedics, 2016, 150, 64-70*

## Chapter 3

**Kragt L,** Dhamo B, Wolvius EB, Ongkosuwito EM. The impact of malocclusions on oral health-related quality of life in children - a systematic review and meta-analysis.
*Clinical Oral Investigations, 2015, [Epub ahead of print]*

**Kragt L,** Jaddoe VWV, Wolvius EB, Ongkosuwito EM. The association of subjective orthodontic treatment need with oral health-related quality of life.
*Community Dent Oral Epidemiol, accepted*

**Kragt L,** van der Tas JT, Moll HA, Elfrink MEC, Jaddoe VWV, Wolvius EB, Ongkosuwito EM. Early caries predicts low oral health-related quality of life at a later age.
*Caries Research, 2016, 50, 471-479*

## Chapter 4

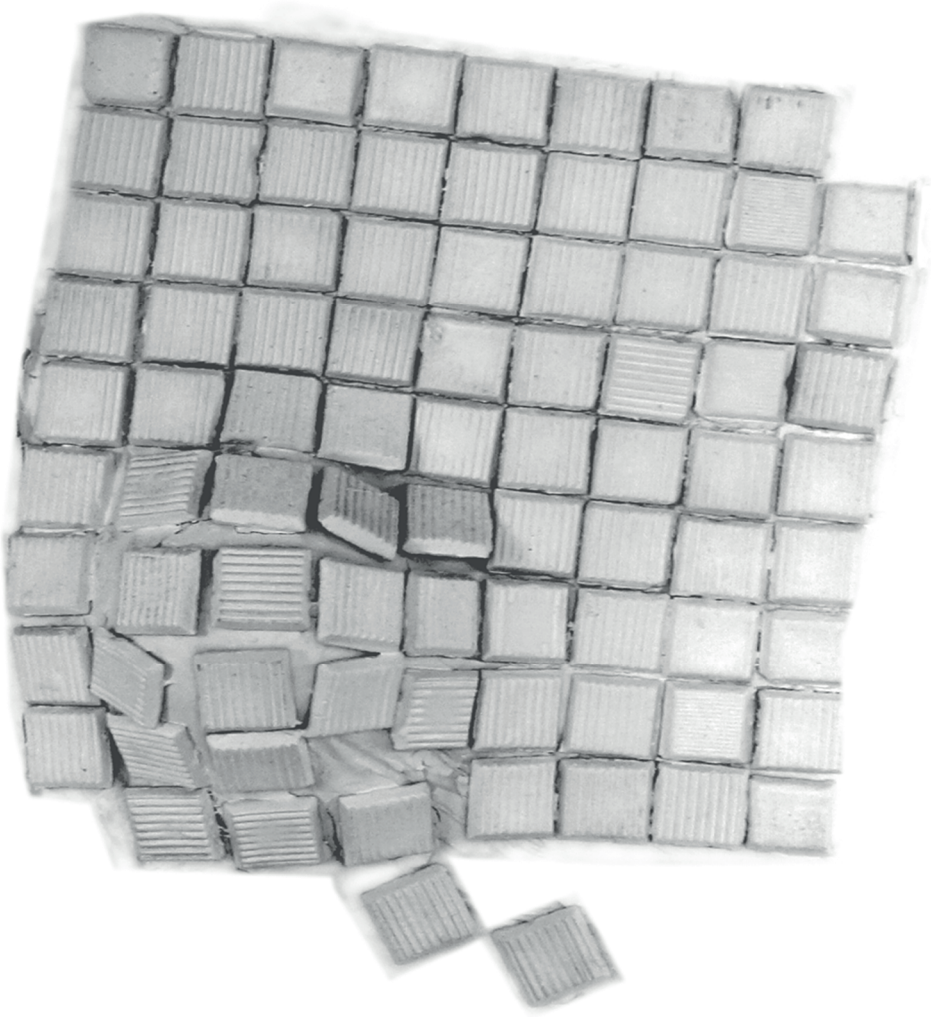
**Kragt L,** Wolvius EB, Raat H, Jaddoe VWV, Ongkosuwito EM. Social inequalities in children's oral health related quality of life: The Generation.
*Submitted for publication*

## Chapter 5

**Kragt L,** Wolvius EB, Jaddoe VWV, Ongkosuwito EM. Associations between objective and subjective orthodontic treatment need related to children's oral health related quality of life: Comparison between ethnic groups.
*Submitted for publication*

**Kragt L,** Wolvius EB, Jaddoe VWV, Tiemeier H, Ongkosuwito EM. Influence of self-esteem on self-perceived orthodontic treatment need and oral health-related quality of life.
*Submitted for publication*

# Chapter 1
# General introduction



## General introduction

### *Children's oral health related quality of life (OHRQoL)*

Reflecting the WHO definition and bio-psychosocial concepts of health, oral health is defined as: the "standard of health of the oral and related tissues which enables an individual to eat, speak and socialize without active disease, discomfort or embarrassment and which contributes to general well-being" (1). Oral health can be measured objectively and subjectively. The term objective oral health measure is used to describe the different methods clinicians, like dentist, orthodontist and other oral professionals use to determine oral health deviations, diseases and treatment needs. The term subjective oral health measure is used to describe the perception of patients with regard to their oral health deviations, diseases and treatment needs. Objective oral health measures and subjective oral health needs were shown to be weakly correlated in different fields of dentistry, but most profound in the orthodontic specialty (2, 3). To explain these differences, the concept of oral health-related quality of life (OHRQoL) was introduced in the orthodontic literature (4, 5). Basically, OHRQoL aims to capture subjective oral health in an objective way. However, it appeared that also OHRQoL cannot simply explain the inconclusive relation between objective orthodontic oral health measures and subjective orthodontic oral health needs.

Nowadays, OHRQoL is the most prominent patient-reported outcome measure in dentistry, and its use has enormously increased in the recent decade (1, 6, 7). OHRQoL measures the particular impact of oral health in terms of oral symptoms, functional limitations, emotional and social wellbeing on daily life (6). Thus, the concept of OHRQoL is very suitable to measure the perceived oral health of individuals. OHRQoL has several definitions, which differ in their complexity. The most general, but comprehensive one was given in the United States Surgeon General's report on oral health: "[OHRQoL is defined as] a multidimensional construct that reflects (among other things) people's comfort when eating, sleeping, and engaging in social interaction; their self-esteem; and their satisfaction with respect to their oral health" (7). The multidimensional character of OHRQoL suggests influences from various factors other than clinical status only.

The assessment of OHRQoL is of importance for theoretical as well as practical reasons. First, oral health is important for overall health and quality of life (6). Second, the assessment of OHRQoL links the relationship between traditional clinical variables and person centered self-reported measures, herewith influencing the decision-making process in clinical practice (4, 6). Third, OHRQoL is important to determine social inequalities and access to care related to oral health (6, 8, 9). Thus, potential uses of OHRQoL can be found in clinical (communication with patient, evaluation of care), public health (monitoring oral health, assessment of need, promoting a more active role of the patient), epidemiologic (evaluation of interventions, finding relationships), and political fields (planning of health policy and resource allocation).

### *Research on children's OHRQoL*

Because oral health during childhood predicts oral health in adulthood, it is particular important to conduct OHRQoL research in children (10, 11). This fact may have reached

the orthodontic discipline more than other pediatric oral health specialties, possibly because the majority of orthodontic patients are children and because orthodontic treatment during growth and development of the child's face, including dentition, is crucial for its later health of the oral and related tissues (12, 13).
OHRQoL is commonly assessed with multiple item questionnaires. Many different measures are developed for the assessment of children's OHRQoL and the most recent one, designed for use in research and practice as well as for clinical and community-based samples, is the Child Oral Health Impact Profile (COHIP). Figure 1 gives the common dimensions, i.e. oral symptoms, functional wellbeing, emotional wellbeing, peer interaction and school of children's OHRQoL, as proposed by the COHIP (14). This particular questionnaire covers the 5 domains of OHRQoL with 38 questions. The length of the COHIP contributes to a high response burden, which is a common problem with OHRQoL questionnaires.
Most studies on OHRQoL, especially in the orthodontic field, use small convenience samples, which limit their evidence. Furthermore, studies on OHRQoL in the orthodontic field often make use of clinical samples instead of population-based samples. Although this can enhance accurateness of the data collection, this also increases the chance for selection bias, meaning that the conclusions drawn are not representative for the general population (15).

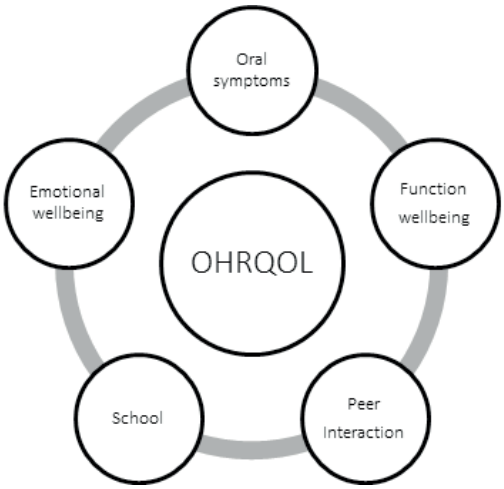



**Figure 1.** Dimensions comprising oral health-related quality of life (OHRQoL) based on the Child Oral Health Impact Profile (COHIP) (14).

Systematic reviews emphasize the impact of heterogeneous population groups and measurement tools on the conflicting evidence in OHRQoL research (16, 17). Large-scale population-based studies on OHRQoL are limited to two prospective cohort studies, the Cardiff Dental Health study and the Pelotas Birth Cohort (11, 18). One reason for this lack in large-scale (orthodontic) oral health research might be due to the ab-

sence of valid oral health assessment methods applicable in the extensive logistics of data collection within large samples.

A useful framework to guide research on OHRQoL is given by the Wilson and Cleary Model for health-related quality of life refined by Ferrans and Colleagues (19-22). This refined model is the most common used one in health-related quality of life research and can be applied to any health specialty; we applied it to oral health, particularly orthodontics (23). This multidimensional model relates objective oral health, called biological/physiological factors, with OHRQoL, via symptoms status, functional status, and general oral health perception. This taxonomy in turn is influenced, or modified, by individual and environmental factors. Wilson and Cleary defined each variable clearly, which enables an accurate measurement of all of them (19). Ferrans and Colleagues explained later the understanding of environmental and individual factors (24). While the refined Wilson and Cleary model helps clinicians for a broader view on OHRQoL than biological factors only and researchers to generate hypothesis to be tested, the model was never tested in its entirety (23). Moreover, to the best of our knowledge it was never applied to populations that are unable to rate their own OHRQoL, like young children. Therefore, the proposed relationships might not always hold true. Although the refined Wilson and Cleary model already simplifies the complexity of the concept of OHRQoL, a major problem to test the entire model is still formed by the overall complexity of the model with multiple relationships (23). Figure 2 depicts the Wilson and Cleary model of health-related quality of life adapted for OHRQoL.

*Biological and Physiological factors and children's OHRQoL*

Assessment of biological and physiological factors includes the occurrence and diagnosis of oral diseases as conceptualized in clinical practice like dental caries, gum disease, malocclusions and dental traumas.

Studies on the prevalence of oral diseases in children are very limited and suffer from heterogeneous assessment methods. However, based on the limited amount of studies it can be concluded that children's oral health problems are highly prevalent. Worldwide, caries prevalence in children varies from 13% to 60%, whereas Dutch children show an estimated prevalence of 19% to 62% (25, 26). The prevalence of periodontal disease is estimated to be 5% to 70% in children worldwide, however no specific data are available on periodontal health in Dutch children (26). Malocclusions are the most common cause for oral treatment among children worldwide with a prevalence of 17% to 90% (26). In the Netherlands, more than half of young adults had orthodontic treatment (25). Also traumatic dental injuries show a high prevalence all over the world up to 41 % in children (26). Unfortunately, there is no information about dental trauma in Dutch children.

All of these oral conditions were reported to have negative effects on OHRQoL. However, the strength of the relationships do vary enormously between but also within these conditions (5, 17, 27-30). The impact of caries on OHRQoL is exclusively seen on the oral symptoms domain (28), whereas periodontal disease was shown to affect OHRQoL mostly in the oral symptoms and the functional domain (27, 31). The impact of dental trauma and malocclusion, however, is mainly seen in the social and emotional domains of OHRQoL (17, 30).

*Characteristics of the environment and children's OHRQoL*

Environmental characteristics affecting the relationship between oral conditions and OHRQoL are categorized into social and physical (24). In relation to oral health, support of social networks and socio-economic status (SES) (social) as well as provision and access to dental care (physical) appear to be most relevant.

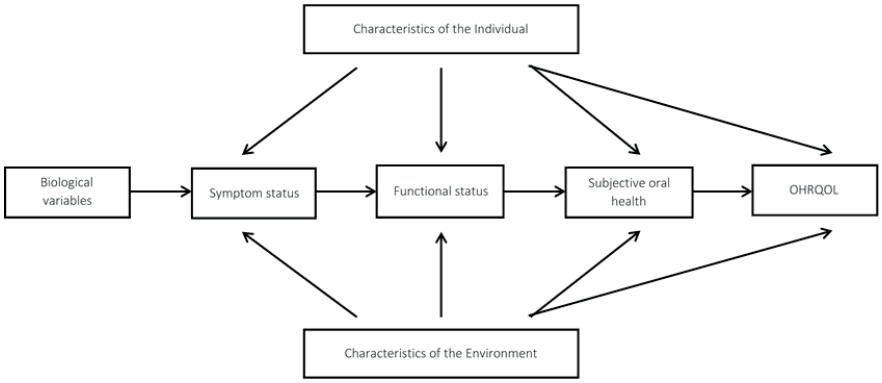



**Figure 2.** Conceptual Model of possible pathways through which subjective oral health measures and OHRQoL are determined. This model is adapted from the Wilson and Cleary model for health related quality of life (1)

SES in relation with OHRQoL has widely been investigated, but associations are inconsistent. As SES can be measured with many different indicators, this might be one of the reason why results on the relationship between SES and OHRQoL are mixed (32). In addition, as children do not have their own socio-economic status, they need to be classified according to indicators of their family socio-economic position (SEP). The most traditional indicators of family SEP are parental educational level, parental employment status and household income (33). Educational level as well as household income were both shown to be unrelated to OHRQoL as well as to be positively related (3, 34-39). Another reason why the results on the relationship between SES and OHRQoL are mixed is, that many studies do not take into account the potential mediating or modifying role of SEP indicators within the relationship between objective oral health measures and subjective oral health measures, but rather look at the direct effects on OHRQoL. Furthermore, social networks, like family and friend support, were shown to be positively related to oral health (40, 41). However, the association between social networks and OHRQoL is less investigated, only one study indicated that people with more social support perceive better OHRQoL (21).

Of course household income can be strongly related to access to dental care, however also the availability of dental specialty practices might influence OHRQoL. Unfortunately, no data are available yet on the association between presence of dental care and OHRQoL. Next to this, having a dental health insurance might influence OHRQoL. Studies have shown that a lack of dental insurance is associated with various adverse oral health outcomes (42-44). However, no studies investigated the relation between dental insurance and OHRQoL.

*Characteristics of the individual and children's OHRQoL*

Individual factors influencing the relationship between oral conditions and OHRQoL are categorized into demographic, developmental, psychological and biological (24). In relation to dentistry, factors like family history (biological), sex and age (demographic), oral health behavior (developmental) or self-esteem, sense of coherence and coping beliefs (psychological) are those which generally emerge to be of interest. However, the associations of these factors with OHRQoL or other self-perceived oral health measures is largely unknown due to a lack of literature.

A limited amount of studies has investigated the associations between individual factors and OHRQoL, however this showed to be generally weak. For example, people performing unfavorable oral health behaviors, i.e. less dental visits and lower tooth brushing frequency, reported to have lower OHRQoL (45). Higher sense of coherence, which enables people to manage stress and to find solutions for health problems, as well as positive dental coping beliefs were associated with better OHRQoL (20-22, 45, 46). And lower self-esteem, defined as the perception of one's own to deal efficiently with the environment, was associated with lower OHRQoL in several studies, but seemed unrelated to clinical oral health status (20, 47). Biological and demographic factors have not been related to OHRQoL yet, except for age, which was consistently negatively associated with OHRQoL (48, 49). Based on these studies it is not clear yet, whether these individual factors have only direct relationships with OHRQoL or also modify the relationship between all oral diseases and subjective oral health measures and the way these relationships function.

*Research questions*

The overall aim of this thesis was to understand the relationships among measures of objective health outcomes, subjective oral health outcomes and OHRQoL related to the orthodontic field. Therefore, following research questions were formulated:

1. How can objective oral health measures, in particular orthodontic treatment need, and subjective oral health measures, in particular OHRQoL, be assessed in children within the scope of large scale epidemiological research?
2. What is the relation between children's objective oral health measures, like malocclusion and caries experience, as well as children's subjective oral health measures, like perceived orthodontic treatment need, with OHRQoL ?
3. How do non-clinical variables, like characteristics of the environment, i.e. socio-economic position, or characteristics of the individual, like gender and self-esteem, influence the relationship between objective oral health measures, subjective oral health measures and OHRQoL in children?

*Methods*

The studies conducted in this thesis have used different methods.

The first research question was addressed with validation studies, which made use of two clinical samples. The study sample for the validation study of the shortened OHRQoL measure comprised 241 12-year old children with their caregivers recruited from an orthodontic practice in Capelle a/d IJssel, The Netherlands as well as the Department of Orthodontics in the Erasmus University Medical Centre, Rotterdam, The

Netherlands. The study sample for the validation of photographic records for the assessment of orthodontic treatment need with the Index of Orthodontic Treatment Need (IOTN) comprised 91 12-year old children recruited from an orthodontic practice in Capelle a/d IJssel, The Netherlands. The data collection for both validation studies started after the first consultation at the practice but before treatment had started except for those with craniofacial malformations. Data for the assessment of children's OHRQoL were assessed in parental questionnaires with the COHIP. Data for the assessment of the IOTN included plaster casts, 2D and 3D photographs as well as radiographs. Informed consent was, of course, obtained from the parents before data collection started.

The first study addressing the influence of objective oral health, to be precise, malocclusions on OHRQoL is a systematic literature review and meta-analysis. Relevant studies were identified in Medline OvidSP, Embase, Web-of-sciences, Cochrane central, PsycINFO, OvidSP, Scopus, PsycINFO, Cinahl and finally Google Scholar. The systematic literature search was performed with an extended search strategy and the selection of studies was done by two reviewers independently based on pre-defined inclusion and exclusion criteria. In total, 57 studies were included in the narrative part of the study and of those 40 studies were included in the meta-analysis.

The remaining studies were embedded in the Generation R Study, a population based prospective cohort study from fetal life until young adulthood conducted in Rotterdam, The Netherlands (50). Originally, this study has been designed for the identification of early environmental and genetic determinants of normal or abnormal growth, development and health. All pregnant women who had an expected delivery date between April 2002 and January 2006 were invited to participate in the study resulting in an enrolment of 9778 women living in the study area. In the school-aged period of the Generation R Study, still 8305 children participated, of which 6690 children visited the dedicated research center at which photographic records were taken for the assessment of caries. The study population for the study period of Generation R at children's age of 10 still comprised 7393 children, of which 5862 children visited the dedicated research center at which photographic and radiographic pictures were taken for the assessment of objective orthodontic treatment need. The main measures of this thesis, OHRQoL and subjective orthodontic treatment need, were assessed in questionnaires, which were send to the mother and completed for 4141 children. Objective orthodontic treatment need and OHRQoL were assessed following the methods validated in the previous mentioned validation studies.

### *Outline of the thesis*

Following the general introduction in *chapter 1*, *chapter 2* describes two methods for large-scale epidemiological research about children's OHRQoL, in particular for the orthodontic field. We validate the use of a shortened OHRQoL measure, which was derived from the COHIP and we validate the use of photographic records, i.e. 3D extra-oral pictures, 2D extra-oral photos, lateral cephalograms and orthopantomograms, for the assessment of the Index of Orthodontic treatment need (IOTN). *Chapter 3* is devoted to oral health factors related to OHRQoL.

**Table 1.** Studies presented in the thesis

| Chapter | Study sample | Age | N | Study design | Main exposure | Main outcome |
|---|---|---|---|---|---|---|
| **2. Methods to conduct large scale orthodontic and OHRQoL research** | | | | | | |
| **2.1** | Clinical sample | 12 years | 243 | Cross-sectional/ validation study | Not applicable | OHRQoL |
| **2.2** | Clinical sample | 12 years | 91 | Cross-sectional/ validation study | Not applicable | Orthodontic treatment need |
| **3. Biological and Physiological factors influencing OHRQoL** | | | | | | |
| **3.1** | Systematic literature review | 5 - 17 years | 57 (40) | Not applicable | Malocclusions | OHRQoL |
| **3.2** | Generation R | 10 years | 3774 | Cross-sectional | Subjective orthodontic treatment need | OHRQoL |
| **3.3** | Generation R | 6-years/ 10years | 2833 | Longitudinal | Caries experience | OHRQoL |
| **4. Characteristics of the environment influencing OHRQoL** | | | | | | |
| 4.1 | Generation R | 10 years | 3796 | Cross-sectional | Different SEP indicators | OHRQoL |
| **5. Characteristics of the individual influencing OHRQoL** | | | | | | |
| **5.1** | Generation R | 10 years | 3040 | Cross-sectional | Objective orthodontic treatment need, Ethnic background | Subjective orthodontic treatment need, OHRQoL |
| **5.2** | Generation R | 10 years | 3796 | Cross-sectional | Subjective orthodontic treatment need, Self-esteem | OHRQoL |

OHRQoL = oral health-related quality of life
SEP = socio-economic position

Here, we investigate the impact of malocclusion on OHRQoL and potential age and cultural differences in this association are examined. Also, the association of subjective orthodontic treatment need with OHRQoL is investigated. Within this study, gender differences are studied. Furthermore, we investigate the influence of caries experiences in early childhood on OHRQoL at later age. *Chapter 4* focusses on the influence of environmental characteristics on subjective orthodontic treatment need and OHRQoL. For this, we describe social inequalities in OHRQoL. *Chapter 5* concentrates on individual characteristics, i.e. the influence of self-esteem on the relationship between subjective orthodontic treatment need and OHRQoL. Also, we investigated differences among ethnic groups in the associations between OHRQoL, subjective and objective orthodontic treatment need. Finally, the overall discussion of the thesis is presented in *chapter 6* with a statement of the principal findings, strength and weaknesses of the included studies, possible mechanisms, implications for clinicians as well as policymakers and unanswered questions for future research. Table 1 gives an overview of the studies included in this thesis.

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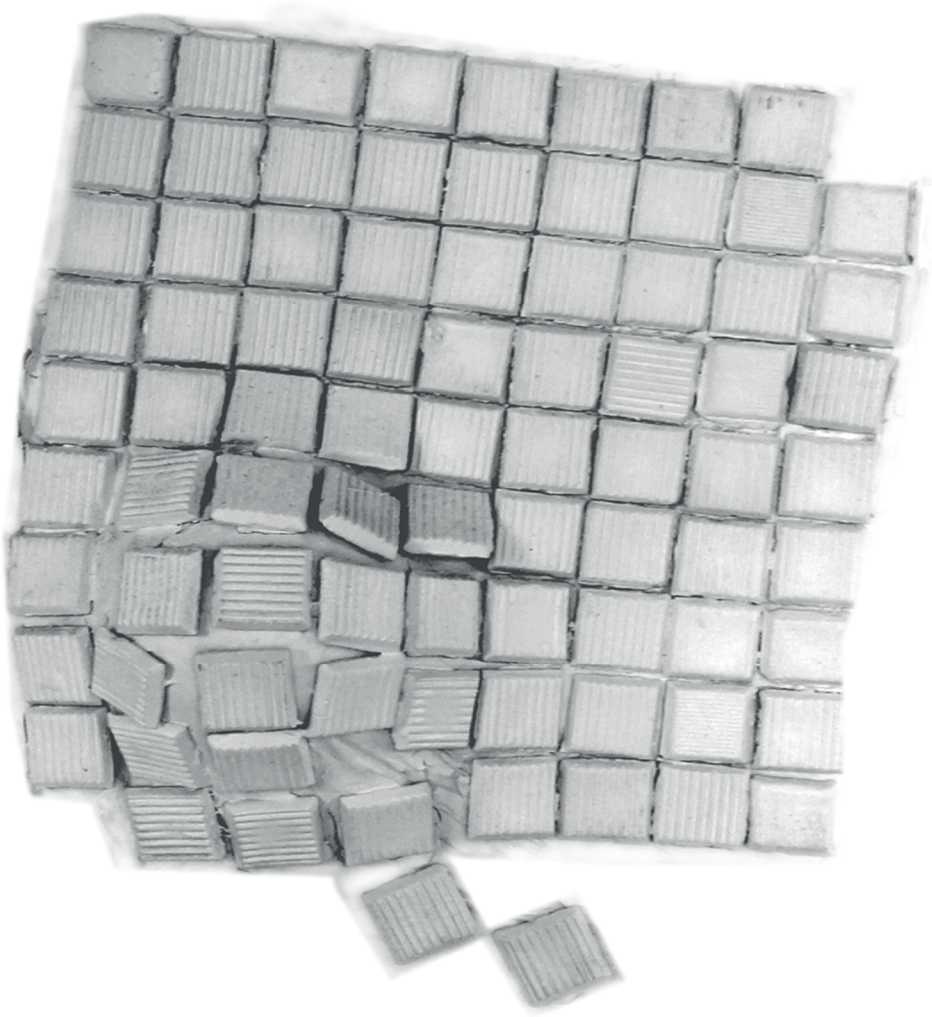
# Chapter 2
# Methods to conduct large scale orthodontic and OHRQoL research



# Chapter 2.1

## Measuring oral health-related quality of life in orthodontic patients with a short version of the Child Oral Health Impact Profile (COHIP)

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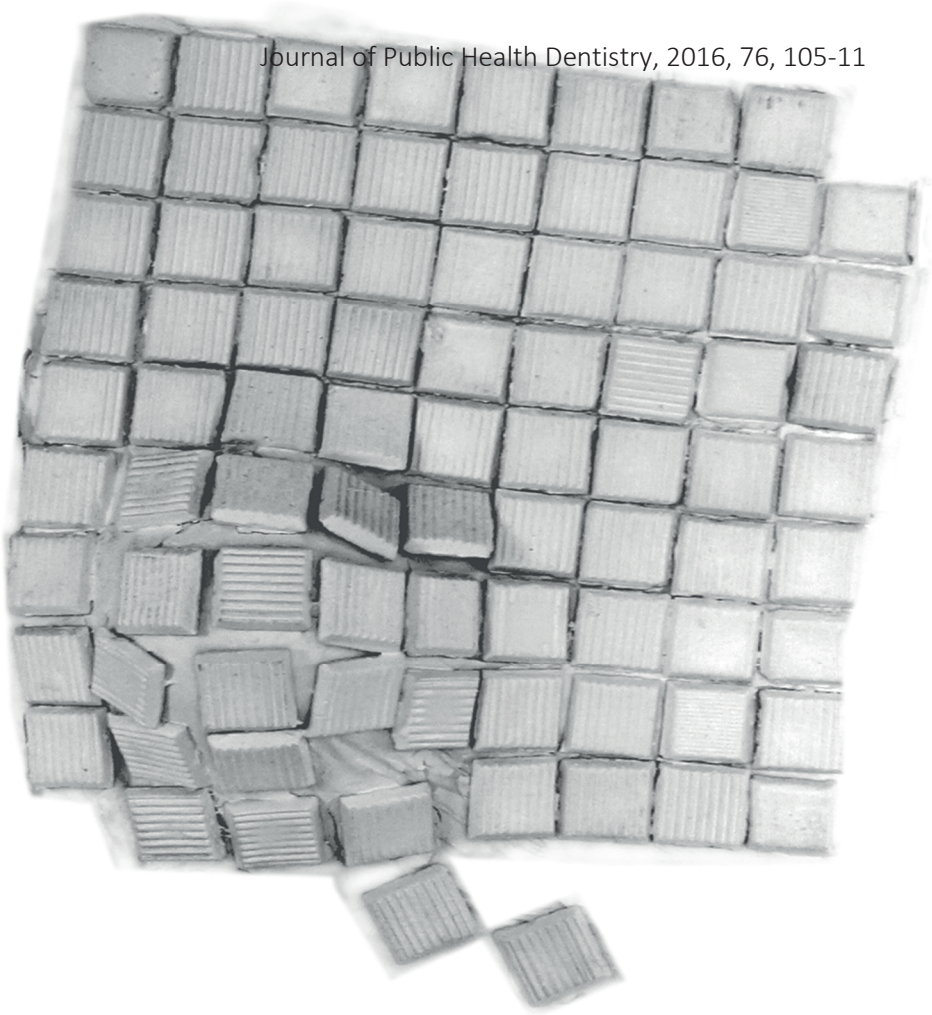

Abstract

***Objectives*** Oral health-related quality of life (OHRQoL) is currently assessed by long questionnaires, which limits its applicability as an outcome measure in orthodontic practice and research. The aim of the study was to evaluate a shortened measure with a low response burden for assessing OHRQoL in children.

***Methods*** A cross-sectional study of 243 prospective orthodontically treated children (12% cleft lip, 68% with definite objective treatment need, 20% borderline objective treatment need) collected data on OHRQoL using the 38-item version of the children's oral health impact profile (COHIP-38), general health perception (GHP), and subjectively and objectively measured orthodontic treatment need. Eleven items of the COHIP-38 were selected for the short version of the questionnaire (COHIP-ortho). Score distributions, internal consistency, construct and criterion validity as well as subgroup analysis were used to evaluate the psychometric properties of both questionnaires. The performances of COHIP-ortho and COHIP-38 were compared.

***Results*** The internal consistency was somewhat lower for the COHIP-ortho compared to the COHIP-38, but on an acceptable level for both questionnaires. The correlations between COHIP-ortho scores and COHIP-38 overall as well as subscale scores were excellent. COHIP-ortho performed adequately regarding construct and criterion validity related to most sample characteristics compared to the performance of the COHIP-38.

***Conclusion*** The COHIP-ortho is as valid as the COHIP-38 for assessing OHRQoL in children. With a low response burden for patients or study participants, reduced cost, and less time needed for administration, the COHIP-ortho simplifies the investigation of OHRQoL in orthodontics.

Keywords Quality of life, Short form, Orthodontics, Validation, Oral health

## Introduction

Orthodontics has been a well-established dental specialty in the Netherlands for more than 50 years. In 2011, 60% of Dutch adolescents were reported to have had orthodontic treatment during childhood (1). However, how orthodontic treatment need is defined and the general health benefits of treatment are still ambiguous (2). Inconclusive results may be due to a general lack of orthodontic research on this topic or the use of improper outcome measures (3).

Orthodontic treatment need is often assessed with objective measures, such as the Index of Orthodontic Treatment Need (IOTN) (4). However, the subjective psychological impact of malocclusions is an important determinant of treatment need and satisfaction (5). Although patient-reported outcome measures provide a complementary or alternative measure to objectively assess orthodontic problems in research and practice, patient reported outcomes have not yet been established in this area of dentistry. Evidence for the burden of malocclusions, including dental crowding, from the patient's perspective is scarce (5).

In the dental literature the term 'oral health-related quality of life' (OHRQoL) is commonly used to describe a conceptualized patient-reported outcome measures on oral health (6). OHRQoL is assessed with questionnaires covering oral health in the functional, emotional, and social well-being domains to measure the influence of oral health on quality of life in general (7, 8). The relationship between OHRQoL and malocclusions is not clear yet (9, 10).

Some of the existing OHRQoL measures are usable in an orthodontic setting (11). However, these questionnaires are generally very long and burdensome to respondents, which is one reason why they are not used. The length of the OHRQoL measures makes administration difficult and the orthodontic literature recommends designing shorter OHRQoL measures (3, 12, 13). In order to make the OHRQoL measure more specific for orthodontics, redundant items less related to malocclusion, crowding, or crooked teeth can be removed from the instruments. The OHRQoL measure needs to be applicable in 8-13 year old children, because orthodontic treatment commonly starts at this age. A short and specific OHRQoL measure is not yet available to this extent. Therefore, the aim of this study was to validate the psychometric properties of a shortened questionnaire for assessing OHRQoL in children considered for orthodontic treatment. This measure should simplify the assessment of children's OHRQoL in dental practice and enhance its applicability as an outcome measure in large epidemiological studies.

## Material and Method

The protocol of the study was reviewed and approved by the METC of Erasmus MC Rotterdam, the Netherlands (METC-2013-098). Informed consent was obtained from the participants prior to administering the questionnaire.

### *Procedure*

The original long version of an OHRQoL measure was distributed to eligible participants. When the data collection was complete, OHRQoL scores for the participants were obtained in two ways. First, scores were calculated based on all questions on the original questionnaire. Second, scores were calculated separately based on a selection of 11

questions. The two scores were then compared in regards to psychometric tests and sample characteristics.

*The questionnaire*

The basis for the short and practical OHRQoL questionnaire was the 38-item Child Oral Health Impact Profile (COHIP-38) developed by Broder et al (14). This questionnaire was chosen for several reasons. The COHIP-38 is the most recently developed OHRQoL measure for children and a refinement of the Child Perception Questionnaire (CPQ), which is well known in dental research (11). The COHIP-38 is designed for use in research and practice with different clinical groups, such as pediatric, orthodontic, and craniofacial, as well as children from a community-based sample (14). Children and parents were closely involved in the development of the COHIP-38 (14). The COHIP-38 is the only OHRQoL measure that has both positively and negatively formulated items (11). Furthermore, a Dutch translation of the COHIP-38 is available, which is designed for the parent and child (15). In the present study the parent form was used as the primary caregivers were considered suitable proxies for measuring OHRQoL in children between 8 and 15 years of age (16, 17). The Dutch COHIP-38 contains 38 questions comprising five subscales: oral symptoms (10 items), functional well-being (8 items), emotional well-being (10 items), school (4 items), and peer interaction (6 items) (18). The questions inquire about the frequency of oral health impacts on daily life and are answered on a 5-point Likert scale: never (5 points), almost never (4 points), sometimes (3 points), often (2 points), and always (1 point). We included an additional "I don't know" (DK) response for validation purposes. The majority of items are negatively formulated, and a higher score indicates better OHRQoL.

The items for the short form of the COHIP-38 were selected prior to the start of the study using an expert-based approach. Two of the investigators, one professional in orthodontics (last author) and one psychiatric epidemiologist (second author), selected 11 items for the COHIP-ortho. Their choices were guided by the factor analysis described by Broder et al (19) and consideration of the OHRQoL model developed by Locker and Allen (7).

*Participants*

A convenience sample of children seeking orthodontic treatment between June 2013 and March 2014 was used to collect the data for this validation study. The participants were children and their caregiver visiting an orthodontic practice for the second time. For this study, a total of 315 parents were asked to complete the questionnaire. In addition, 30 parents of children with a cleft lip or palate were asked to fill out a questionnaire. Data collection started after the first consultation but before treatment for all patients except those with craniofacial malformation. Children who neither wanted nor needed treatment at the first visit, children whose parents did not have sufficient command of the Dutch language or children with more than 30% of missing answers were excluded from the study.

*Measures*

OHRQoL was scored for the children based on the COHIP questions, with each question having the same weight. Overall and subscale scores were calculated the same way. Four positively worded items were reversed to ensure that the highest score reflected the best possible OHRQoL. In line with other studies, DK responses were treated as missing values and replaced by the mean score for that relevant subscale (18). This method is similar to a multiple imputation method but more reasonable for clinicians (20). The possible overall score for the COHIP-38 ranged from 38 to 190. The OHRQoL scores based on the COHIP-ortho were calculated the same way, but with a possible range of 11-55.

Subjective orthodontic treatment need was measured by the question *'Do you think your child needs orthodontic treatment?'* Responses to the question were on a 5-point Likert scale from "disagree" (5 points) to "agree" (1 point). For the analysis, the response options were re-categorized into a yes –no format as "agree" (1-2 points) and "do not agree" (3-5points).

General health perception (GHP) was assessed with six additional questions (*Appendix 1*). These questions were answered on a 5-point Likert scale with a high score indicating a good GHP. The overall score for GHP was calculated the same way as the COHIP score and then back transformed to a 5-point Likert scale ranging from bad (1 point) to excellent (5 points). One question about perceived happiness, which was also answered on a 5-point Likert scale, completed the questionnaire.

Objective orthodontic treatment need was measured using the IOTN (4). The IOTN has two components, a dental health component (DHC) and an aesthetic component (AC). The IOTN-DHC rates treatment need from a dental health point of view on a scale from 1 to 5, with a score higher than 3 indicating definite clinical need. The index for the IOTN-AC ranges from 1 to 10, with 10 indicating the most severe aesthetic impact of malocclusion. Treatment need is commonly defined at IOTN-AC > 5. The investigator was calibrated in using the IOTN on dental casts (22).

*Statistical analysis*

Statistical analyses were performed using Statistical Package for Social Sciences (SPSS) version 20.0 for Windows (2011, SPSS Inc., Chicago, IL, USA). All variables were summarized as mean scores and standard deviation (SD). The overall scores for the COHIP-38 and COHIP-ortho, as well as the subscale scores, were tested for normality using the Shapiro-Wilk-Test ($\alpha > 0.05$) (*Results in Appendix 2*).

Floor and ceiling effects (> 25% of the respondents having the lowest/highest value) were evaluated per question and per overall score by looking at the distributions of scores.

The reliability of the COHIP-38 and the subscale scores of the COHIP-38 and COHIP-ortho were investigated using Cronbach's alpha ($\alpha$) and corrected item total correlations. For the COHIP-38, $\alpha \geq 0.7$ was considered to provide acceptable internal consistency (23). For the COHIP-ortho, the acceptable level was set to $\alpha \geq 0.6$ because it has fewer items, which typically reduces the $\alpha$ levels and the minimum for the item total correlation was set to 0.2 in order to test for sufficient consistency between items and the overall COHIP score (24)

Concurrent criterion validity is how well the COHIP-ortho assesses OHRQoL compared to a gold standard, which is the COHIP-38 in this study. Spearman's correlation coefficients were used to test for criterion validity. Correlations between subscale scores, overall scores, and COHIP-ortho scores were investigated. Also kappa correlations between COHIP-ortho and COHIP-38 scores reverse transformed to a 5-point scale were investigated and judged following the guidelines of Landis and Koch (25).
The construct validity of a measure describes the way the measure relates to other known concepts and can be divided into convergent and discriminant validity (26). Convergent validity is how well the COHIP-ortho corresponds to measures with which it should correlate. Discriminant validity describes whether the measure is unrelated to concepts to which it should be unrelated. Convergent and discriminant validity were addressed by age-adjusted partial Spearman correlations between COHIP-ortho or COHIP-38 scores and perceived treatment need, DHC, AC, GHP, and happiness.
Furthermore, the discrimination of the COHIP-ortho among different sample characteristics (gender, dichotomized perceived treatment need, DHC, AC, GHP, and happiness) was assessed. Mean scores were summarized and evaluated by the Mann-Whitney-U test. The differences were compared to the differences in mean COHIP-38 scores among the sample characteristics.

## Results

### *Sample characteristics*

A total of 315 questionnaires were distributed to parents, of which 47 refused to give informed consent and 25 did not return the questionnaire or returned it uncompleted (response rate 77%). Therefore, 243 questionnaires remained for the validation study. A similar number of boys and girls participated, with an overall mean age (SD) of 11.86 (1.73) years. All children, except one, had an IOTN-DHC score of 3 or higher. Also, the GHP of the sample was rated as 'good' or better, and the majority of the participants were 'very often' happy over the last 3 months. However, almost the whole range of the IOTN-AC scale was represented in the sample. Most of the parents perceived an orthodontic treatment need for their children, and 13% of parents asserted that they did not know whether their children were in need of treatment. The sample characteristics of participating children are provided in Table 1.

### *Score distributions*

The mean (SD) COHIP-ortho score was 46.72 (4.86), with a minimum score of 26 and maximum score of 55. The mean (SD) COHIP-38 score was 166.24 (14.06), with a range of 116 to 189. OHRQoL was generally high in the orthodontic sample. All COHIP-ortho items exhibited ceiling effects (> 25% of the participants had the maximum score), and six questions had profound ceiling effects (> 50% of the participants had the maximum score). For the overall COHIP-ortho score, 37.4 % of the participants scored in the highest range (49-55 points) and 1.2% had the highest score (55 points, Table 2). For the COHIP-38, 55.9% of the participants scored in the highest range (168-190 points) and 0.8% had the highest obtained score of 189 (*Appendix 3*).

The only positively formulated question in the COHIP-ortho (i.e. *'During the past 3 months, how often did your child feel that he/she is attractive (good looking) because of his/her teeth, mouth, or face')* had the highest percentage of DK responses (30.0%). Two more questions in the COHIP-38 had a high DK response rate: the question about *'confidence'* (27.0%) and the question about *'felt worried about the teeth'* (12.0%)

**Table 1.** Sample characteristics of the children participating in the validation study

| | | Female | Male |
|---|---|---|---|
| **N** | | **122** | **121** |
| **Mean Age (SD)** | | 11.6 (1.58) | 12.1 (1.84) |
| **Subjective Treatment Need**[a] | Don't know | 17 | 12 |
| | Totally disagree | 1 | 4 |
| | Disagree | 4 | 3 |
| | Agree | 57 | 52 |
| | Totally agree | 35 | 28 |
| **Objective Treatment Need**[b] | Grade 1 | 0 | 0 |
| | Grade 2 | 0 | 1 |
| | Grade 3 | 28 | 18 |
| | Grade 4 | 57 | 62 |
| | Grade 5 (+ cleft lip/palate) | 28(+8) | 18 (+22) |
| **Aesthetic Treatment Need**[a,c] | 2 | 4 | 0 |
| | 3 | 11 | 11 |
| | 4 | 13 | 13 |
| | 5 | 17 | 9 |
| | 6 | 18 | 16 |
| | 7 | 18 | 15 |
| | 8 | 15 | 15 |
| | 9 | 11 | 16 |
| | 10 | 5 | 3 |
| **General Health Perception**[d] | Bad | 0 | 0 |
| (one missing) | Moderate | 0 | 0 |
| | Good | 14 | 20 |
| | Very good | 2 | 0 |
| | Excellent | 106 | 100 |
| **General Happiness** | Never | 1 | 0 |
| (one missing) | Almost never | 3 | 2 |
| | Sometimes | 7 | 8 |
| | Very often | 71 | 65 |
| | Always | 40 | 45 |

[a] cleft lip/palate received no AC-score and were not asked whether they perceive treatment need, for analysis added to group 'agree'; [b] based on IOTN-DHC; [c] based on IOTN-AC; [d] based on 6 Questions from CHQ

### *Psychometric testing of the COHIP-ORTHO*

The COHIP-38 had a good Cronbach's α of 0.87 in the orthodontic sample. For the COHIP-38 subscales, the values exhibited less reliability, with the smallest Cronbach's α for the functional well-being scale (α = 0.57, 8 items) and the highest for the social emotional well-being scale (α = 0.87, 10 items, *Appendix 3*). The Cronbach's α for the COHIP-ortho was reduced to a marginally acceptable level (α = 0.63). Cronbach's α did not increase after excluding any one of the 11 items, with the exception of the positively formulated item which also showed a low corrected item-correlation (r = 0.18). The results of the reliability analysis are shown in Table 2.

A significant positive correlation (r = 0.87, p< 0.01) was found between the COHIP-ortho and COHIP-38. The COHIP-ortho and subscales of the COHIP-38 were significantly positively correlated (r = 0.38-0.74, all p<0.01, *all correlations are presented Appendix 4*).

Also after reverse transformation of COHIP-ortho scores and COHIP-38 scores to a 5-point Likert scale, both measures showed substantial agreement in assessment of OHRQoL levels (K = 0.605, p = 0.0005, *Appendix 5*). Thus, the COHIP-ortho has high criterion validity.

The similarity in correlations of both COHIP scores with other concepts suggested construct validity of the COHIP-ortho. The COHIP-ortho scores correlated significantly with GHP scores (r = 0.210, p < 0.05), the happiness score (r = 0.230, p < 0.05, Table 3) and the DHC score (r = - 0.115, p = 0.037, Table 3). Similar the COHIP-38 correlated significantly with these concepts (GHP score: r = 0.286, p < 0.05; happiness: r = 0.267, p < 0.05; DHC (r = - 0.163, p = 0.006). The convergent validity of the COHIP-ortho was supported.

No correlations were found between the COHIP-ortho scores and perceived treatment need (r = 0.052, p = 0.212) or AC scores (r = -0.083, p = 0.115). Similar no substantial correlation was found between COHIP-38 scores and perceived treatment need (r = 0.017, p = 0.398) or AC scores (r = - 0.11, p =0.056, Table 3). The discriminant validity of the COHIP-ortho in the orthodontic sample was supported.

The differences in mean COHIP-ortho and COHIP-38 scores between subgroups, including gender (female-male), perceived treatment need (yes-no), DHC (≤3 vs. >3), AC (≤5 vs. >5), GHP, and happiness (lower vs. higher), are presented in table 3. Significant differences in COHIP-ortho scores were found only for GHP (mean difference: 3.32; p = 0.002) and general happiness (mean difference: 4.35, p = 0.001); no significant difference was found between DHC subgroups (mean difference: 1.02, p = 0.210). The same was observed for the COHIP-38 score (GHP mean difference: 12.24, p = 0.00; happiness mean difference: 14.06, p = 0.00; objective treatment need mean difference: 4.07, p = 0.087).

**Table 2.** Floor and ceiling responses, descriptive statistics of the items of the COHIP-ortho, and overall reliability and item analysis of the COHIP-ortho

| COHIP-ortho | Responses | | Descriptives | | | Scale reliability | | |
|---|---|---|---|---|---|---|---|---|
| Item | Always | Never | Mean | SD | Sk[a] | α[b] | Mean[c] | r[d] |
| 1. Toothache | 0 | 75.3 | 4.63 | 0.706 | -1.75 | 0.613 | 42.08 | 0.236 |
| 3. Crooked teeth/spaces | 11.1 | 27.6 | 3.36 | 1.318 | -0.26 | 0.583 | 43.35 | 0.370 |
| 5. Discolored teeth | 2.5 | 52.3 | 4.14 | 1.056 | -1.01 | 0.585 | 42.57 | 0.363 |
| 6. Had bad breath | 1.2 | 37.9 | 3.98 | 0.987 | -0.63 | 0.623 | 42.74 | 0.188 |
| 7. Bleeding gums | 0.4 | 43.2 | 4.13 | 0.902 | -0.66 | 0.607 | 42.58 | 0.263 |
| 15. Difficulty saying words | 0.8 | 75.7 | 4.56 | 0.866 | -1.98 | 0.612 | 42.15 | 0.236 |
| 16. Difficulty eating foods | 0 | 83.1 | 4.78 | 0.510 | -2.36 | 0.596 | 41.93 | 0.417 |
| 17. Felt worried or anxious | 1.2 | 49.8 | 4.15 | 1.00 | -0.88 | 0.573 | 42.56 | 0.423 |
| 25. Avoid to speak out loud | 0 | 90.9 | 4.86 | 0.475 | -3.90 | 0.594 | 41.85 | 0.457 |
| 26. Been teased or bullied | 0 | 82.3 | 4.75 | 0.602 | -2.58 | 0.597 | 41.96 | 0.372 |
| *31. Felt attractive*[e] | 21.0 | 37.0 | 3.36 | 1.570 | -0.36 | 0.652 | 43.35 | 0.181 |
| **Total** | | | 46.71 | 4.86 | -0.93 | 0.626 | | |
| **Total**[f] | 0 | 37.4 | 4.31 | 0.59 | -0.33 | | | |

[a] Skewness Standard Error 0.1156; [b] Cronbach's alpha if item deleted; [c] Scale mean if item deleted; [d] Corrected item – total correlation; [e] positive formulated items, answer scale reversed; [f] back transformed to Likert scale; SD = Standard deviation

**Table 3.** Comparison of COHIP-ortho and COHIP-38 scores among subgroups and correlation of COHIP-ortho scores with other concepts to evaluate construct validity

| | | | COHIP-ortho | | | COHIP-38 | |
|---|---|---|---|---|---|---|---|
| | | N | Mean (SD) | ρ[a] (p-value) | | Mean (SD) | ρ[a] (p-value) |
| **Gender** | Male | 121 | 46.76 (5.20) | | | 166.17 (14.89) | |
| | Female | 122 | 46.65 (4.52) | | | 166.32 (13.24) | |
| | p-value[b] | | 0.670 | - | | 0.583 | - |
| **Subjective Need** | No | 41 | 47.29 (4.17) | | | 166.90 (14.42) | |
| | Yes (incl cleftlip) | 202 | 46.59 (4.99) | | | 166.11 (12.25) | |
| | p-value[b] | | 0.574 | 0.052 (0.212) | | 0.997 | 0.017 (0.398) |
| **IOTN DHC[c]** | ≤ 3 | 47 | 47.51 (4.32) | | | 169.51 (11.03) | |
| | > 3 | 195 | 46.49 (4.97) | | | 165.44 (14.65) | |
| | p-value[b] | | 0.210 | -0.115[f] (0.037) | | 0.087 | -0.163[f] (0.006) |
| **Clinical group** | Malocclusion | 213 | 46.92 (4.71) | | | 167.26 (13.25) | |
| | Cleft lip/palate | 30 | 45.27 (5.71) | | | 159.03 (17.47) | |
| | p-value[b] | | 0.099 | - | | 0.013 | - |
| **IOTN AC[d]** | ≤ 5 | 78 | 47.27 (4.33) | | | 168.27 (12.05) | |
| | >5 | 132 | 46.70 (4.96) | | | 166.64 (14.04) | |
| | p-value[b] | | 0.441 | -0.081 (0.123) | | 0.549 | -0.110 (0.056) |
| **Health perception[e]** | Lower | 36 | 43.86 (5.94) | | | 155.75 (17.49) | |
| | Higher | 206 | 47.18 (4.48) | | | 167.99 (12.54) | |
| | p-value[b] | | 0.002[f] | 0.230[f] (0.000) | | 0.000[f] | 0.286[f] (0.000) |
| **Happiness[e]** | Lower | 21 | 42.71 (5.90) | | | 153.33 (16.94) | |
| | Higher | 221 | 47.06 (4.58) | | | 167.39 (13.15) | |
| | p-value[b] | | 0.001[f] | 0.210[f] (0.001) | | 0.000[f] | 0.267[f] (0.000) |

[a] partial Spearman correlation adjusted for age (df =240); [b] Mann Whitney U test, 2-sided p-value, significant values are bolded; [c] one missing because of no study cast; [d] based on N = 210 because of missing values; [e] one missing because incomplete questionnaire for GHP and happiness; [f]significant spearman correlations and p values are bold; SD = Standard deviation

## Discussion

The study validated a short practical measure to assess OHRQoL in an orthodontic sample. The performance of the short measure, the COHIP-ortho, was compared to that of the validated original one, the COHIP-38. To our knowledge, this study is the first that developed a short and condition-specific item-list that can be used in large scale epidemiologic studies as well as in clinical settings to assess children's OHRQoL.

In this study, the sample was restricted to future orthodontic patients recruited at one practice, because variability in orthodontic treatment need and malocclusion severity was assumed in this group. Finally, the clinical sample had generally high objective orthodontic treatment need (IOTN-DHC), but the distributions of the COHIP-ortho and COHIP-38 scores were skewed towards good OHRQoL. Other studies have likewise reported rather high OHRQoL scores in orthodontic samples (12, 14, 16-19, 27). The tendency towards high scores is a common phenomenon in the quality of life literature, but validation of measures, for example by detecting variation in generally healthy children seeking orthodontic treatment, is then complicated (21). The ability to detect changes in OHRQoL was not evaluated in this study because the data collection was restricted to one time point. OHRQoL was measured once with the COHIP-38; the COHIP-ortho was not distributed separately, but analyzed in a secondary analysis of the full version. This approach could influence the answers of the participants, but item scores on short

versions of questionnaires are not necessarily different when imbedded in long forms or administered separately (28).
A limitation of this study was that the questionnaire was administered to one of the caregivers of the children. This approach was chosen, because the orthodontic literature describes parents as valid proxies for the assessment of their children's' OHRQoL (16, 17). Parents play an important role in the relationship of orthodontists with their patients as they are into the wellbeing of the child but burdened with the costs of the treatment. However, the COHIP-38 and the COHIP-ortho can be used in the children themselves, when the age, literacy level and other sample characteristics allow this.
One item of the COHIP-ortho behaved conspicuously compared to the other items, the question '*How often did your child feel that he/she was attractive (good looking) because of his/her teeth, mouth, or face?".* In general, the interpretation and analysis of positive formulated items needs extra thoughtfulness due to psychometric issues (13). Based on the reliability analysis, this question would not belong to the concept of OHRQoL (23). However, it is the only positively formulated item in the COHIP-ortho and enables measurement of the eventual positive impacts of orthodontic treatment. Furthermore, this question represents the peer interaction scale, maintaining COHIP-ortho face and content validity for the theoretical OHRQoL concept. Finally, the question addresses a topic that is very important to orthodontics. For a long time, aesthetics has been suggested to be one of the major reasons to seek and provide orthodontic treatment in addition to oral function (2, 9). When the COHIP-ortho is administered to the children themselves, the reliability of the item might increase because they simply do know themselves better. The main aim of the study was to evaluate a short and easy instrument for measuring OHRQoL in children. The COHIP-ortho is short and easy and performed as good as the COHIP-38 in the orthodontic sample. The slightly lowered Cronbach's α seems no direct threat to the internal consistency of the COHIP-ortho, because this measure depends on the number of items in the questionnaire and the variance of COHIP scores in the sample (23, 29). Reliability as well as internal consistency are likely to improve after removing the DK-response from the final COHIP-ortho scale. The DK-response option should not be used in empirical research anymore, because children as well as parents might be encouraged to use this redundantly when it is presented. Correlation and subgroup analysis revealed satisfactory criterion, convergent and discriminant validity of the COHIP-ortho in the orthodontic sample.
Research on the patient perspective in orthodontics is important because of the high proportion of the population that is orthodontically treated and the high costs involved. With a low response burden for patients and study participants, reduced costs and less time needed for administration, the COHIP-ortho stimulates the investigation and clarification of the relationship between OHRQoL, aesthetics, and orthodontics. Future studies may focus on the concordance between children and parent reports on OHRQoL, for example among children of different age groups or to evaluate DK-responses from parents. In general, research should focus more on determinants of OHRQoL independent of clinical health status, which can explain unexpected OHRQoL levels in orthodontic and other samples. This can now easily be done with the COHIP-ortho.
In conclusion, the COHIP-ortho is a good questionnaire to be used in research on OHRQoL, especially related to orthodontics. In dental practice, this instrument can easi-

ly be combined with more objective indices, and in epidemiological studies it can be integrated into health surveys. Subsequently, the COHIP-ortho will contribute to the knowledge of patient's perspective and wellbeing in regard to orthodontics.

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## Supplemental Material

1. Questions to asses general health perception, selected from the Child Health Questionnaire (CHQ-PF28)

a. How would you describe your childs general health?
b. To what extend are the following statements true for your child?
    i. My child appears less healthy than other children.
    ii. My child is never seriously ill.
    iii. If there is an illness going around, my child usally gets it as well
    iv. I expect my child to have a very healthy life
    v. I am more concerned about my child's health than other people are about their child

Answers were given on a 5-point Likert scale (a: bad to excellent; b: not true to true)

Reference: Raat H, Botterweck A, Landgraf J, Hogenveen W, Essink-Bot M. Reliability and validity of the short form of the child health questionnaire for parents (CHQ-PF28) in large random school based and general population samples. J Epidemiol Community Health 2005;**59**(1):75-82.

**Table S1.** Test for normality of COHIP-38 and COHIP-ortho overall and subscale scores

| | Kolmogorov – Smirnov | | | Shapiro-Wilk | | |
|---|---|---|---|---|---|---|
| | Statistic | df | p-value | Statistic | df | p-value |
| COHIP-38 | 0.119 | 243 | 0.000 | 0.915 | 243 | 0.000 |
| COHIP-ortho | 0.108 | 243 | 0.000 | 0.948 | 243 | 0.000 |
| Oral Health | 0.066 | 243 | 0.012 | 0.984 | 243 | 0.009 |
| School | 0.422 | 243 | 0.000 | 0.478 | 243 | 0.000 |
| Emotional | 0.166 | 243 | 0.000 | 0.846 | 243 | 0.000 |
| Functional | 0.114 | 243 | 0.000 | 0.935 | 243 | 0.000 |
| Peer | 0.173 | 243 | 0.000 | 0.837 | 243 | 0.000 |

**Table S2a.** Floor and ceiling effects, descriptive statistics of the items of the COHIP-38, descriptive statistics of OHRQoL scores and subscale scores, and reliability analysis

| (Sub)scale | Item | Responses | | Descriptives | | | (Sub)scale reliability | | |
|---|---|---|---|---|---|---|---|---|---|
| | | Always (%) | Never () | Mean | SD | Skew | α[a] | Mean[b] | r[c] |
| **Oral health** | 1. Had pain in your teeth/toothache | 0 | 75.3 | 4.63 | 0.706 | -1.75 | 0.673 | 35.95 | 0.223 |
| | 2. Breathing through your mouth/snoring | 2.1 | 42.0 | 3.95 | 1.099 | -0.71 | 0.671 | 36.63 | 0.265 |
| | 3. Crooked teeth or spaces between | 11.1 | 27.6 | 3.36 | 1.318 | -0.26 | 0.675 | 37.22 | 0.282 |
| | 4. Sores or sore spots in or around mouth | 0.4 | 53.9 | 4.35 | 0.811 | -1.05 | 0.653 | 36.23 | 0.359 |
| | 5. Discolored teeth or spots on teeth | 2.5 | 52.3 | 4.14 | 1.056 | -1.01 | 0.656 | 36.44 | 0.337 |
| | 6. Had bad breath | 1.2 | 37.9 | 3.98 | 0.987 | -0.63 | 0.65 | 36.6 | 0.367 |
| | 7. Bleeding gums | 0.4 | 43.2 | 4.13 | 0.902 | -0.66 | 0.639 | 36.45 | 0.432 |
| | 8. Food sticking in or between your teeth | 0 | 27.2 | 3.91 | 0.853 | -0.31 | 0.642 | 36.67 | 0.418 |
| | 9. Pain or sensitivity in teeth (hot/cold) | 0 | 51.9 | 4.27 | 0.867 | -0.83 | 0.662 | 36.31 | 0.301 |
| | 10. Dry mouth or lips | 1.2 | 32.9 | 3.86 | 0.996 | -0.42 | 0.629 | 36.72 | 0.47 |
| **Total** | | | | 40.58 | 4.93 | -0.12 | 0.679 | | |
| **Total[d]** | | 0 | 25.1 | 4.14 | 0.59 | -0.03 | | | |
| **School** | 14. Missed school for any reason | 0 | 85.2 | 4.78 | 0.589 | -2.84 | 0.816 | 14.72 | 0.147 |
| | 20. Difficulty paining attention in school | 0 | 91.8 | 4.89 | 0.403 | -4.67 | 0.445 | 14.6 | 0.571 |
| | 25. Not wanted to speak out loud in class | 0 | 90.9 | 4.86 | 0.475 | -3.90 | 0.435 | 14.63 | 0.554 |
| | 36. Not wanted to go to school | 0 | 96.7 | 4.96 | 0.237 | -6.36 | 0.507 | 14.53 | 0.652 |
| **Total** | | | | 19.49 | 1.22 | -3.76 | 0.626 | | |
| **Total[d]** | | 0 | 93.4 | 4.9 | 0.27 | -4.0 | | | |
| **Emotional** | 12. Been unhappy or sad | 1.2 | 71.6 | 4.52 | 0.87 | -1.88 | 0.859 | 39.09 | 0.72 |
| | *13. Felt confident* [e] | 28.0 | 33.7 | 3.16 | 1.65 | -0.17 | 0.925 | 40.46 | 0.261 |
| | 17. Felt worried or anxious | 1.2 | 49.8 | 4.15 | 1.00 | -0.88 | 0.865 | 39.47 | 0.628 |
| | 19. Felt shy or withdrawn | 0.8 | 64.6 | 4.44 | 0.872 | -1.56 | 0.854 | 39.17 | 0.791 |
| | 21. Avoided smiling or laughing | 0.4 | 78.6 | 4.66 | 0.746 | -2.36 | 0.861 | 38.95 | 0.714 |
| | 26. Been teased or bullied | 0 | 82.3 | 4.75 | 0.602 | -2.58 | 0.871 | 38.86 | 0.585 |
| | 27. Felt that he/she looked different | 0.8 | 74.5 | 4.59 | 0.789 | -2.04 | 0.857 | 39.02 | 0.771 |
| | 33. Been worried about what others think | 0 | 63.4 | 4.44 | 0.838 | -1.31 | 0.855 | 39.17 | 0.776 |
| | 34. Got questions about teeth | 0.4 | 57.6 | 4.29 | 0.928 | -0.96 | 0.862 | 39.32 | 0.667 |
| | 35. Been uncomfortable with questions | 0.4 | 74.1 | 4.61 | 0.755 | -2.06 | 0.858 | 39.0 | 0.759 |
| **Total** | | | | 43.61 | 6.52 | -1.44 | 0.878 | | |
| **Total[d]** | | 0 | 56.8 | 4.45 | 0.71 | -1.04 | | | |

[a] Cronbach's alpha if item deleted; [b] Scale mean if item deleted; [c] Corrected item – total correlation; [d] back to Likert scale; [e] positive formulated items, answer scale reversed Skewness SE = 0.156,

**Table S2b.** Floor and ceiling effects, descriptive statistics of the items of the COHIP-38, descriptive statistics of OHRQoL scores and subscale scores, and reliability analysis

| (Sub)scale | Item | Responses | | Descriptives | | | (Sub)scale reliability | | |
|---|---|---|---|---|---|---|---|---|---|
| | | Always (%) | Never () | Mean | SD | Skew | α[a] | Mean[b] | r[c] |
| **Functional** | 11. Had trouble biting off or chewing foods | 1.6 | 64.6 | 4.43 | 0.912 | -1.72 | 0.504 | 30.7 | 0.358 |
| | 15. Difficulty saying certain words | 0.8 | 75.7 | 4.56 | 0.866 | -1.98 | 0.494 | 30.56 | 0.395 |
| | 16. Had difficulty eating food | 0 | 83.1 | 4.78 | 0.510 | -2.36 | 0.508 | 30.34 | 0.466 |
| | *18. No difficulties with cleaning*[e] | 11.1 | 35.4 | 3.75 | 1.29 | -0.93 | 0.541 | 31.37 | 0.272 |
| | 22. Had trouble with sleeping | 0 | 91.4 | 4.90 | 0.338 | -3.62 | 0.556 | 30.23 | 0.232 |
| | *29. Able to eat what he/ she wanted*[e] | 13.2 | 58.8 | 4.13 | 1.35 | -1.54 | 0.589 | 31.0 | 0.175 |
| | 30. Had difficulties to be understandable | 1.6 | 78.6 | 4.64 | 0.80 | -2.63 | 0.505 | 30.49 | 0.369 |
| | 32.Had difficulty keeping the teeth clean | 0.8 | 38.3 | 3.92 | 1.02 | -0.43 | 0.554 | 31.21 | 0.208 |
| **Total** | | | | 35.13 | 3.76 | -0.78 | 0.565 | | |
| **Total[d]** | | 0 | 52.3 | 4.49 | 0.57 | -0.55 | | | |
| **Peer** | 23. Found him or herself ugly | 0.8 | 69.1 | 4.50 | 0.850 | -1.64 | 0.406 | 22.93 | 0.546 |
| | 28. Felt left out by peers | 1.2 | 83.1 | 4.90 | 0.377 | -4.36 | 0.527 | 22.53 | 0.42 |
| | 24. Got angry about the teeth | 0 | 91.8 | 4.74 | 0.682 | -3.32 | 0.435 | 22.69 | 0.547 |
| | *31. Felt that she was attractive*[e] | 21.0 | 37.0 | 3.36 | 1.570 | -0.36 | 0.725 | 24.07 | 0.272 |
| | 37. Did not want to know new people | 0.8 | 96.7 | 4.96 | 0.237 | -6.36 | 0.542 | 22.47 | 0.488 |
| | 38. Had a fight because of the teeth | 0 | 97.9 | 4.98 | 0.180 | -8.14 | 0.564 | 22.46 | 0.388 |
| **Total** | | | | 27.43 | 2.72 | -1.53 | 0.571 | | |
| **Total[d]** | | 0 | 66.7 | 4.6 | 0.55 | -1.16 | | | |
| **Overall** | | | | | | | | | |
| **Total** | | | | 166.25 | 14.06 | -1.17 | 0.87 | | |
| **Total[d]** | | 0 | 49.0 | 4.46 | 0.55 | -0.36 | | | |

[a] Cronbach's alpha if item deleted; [b] Scale mean if item deleted; [c] Corrected item – total correlation; [d] back to Likert scale; [e] positive formulated items, answer scale reversed Skewness SE = 0.156,

**Table S3.** Concurrent criterion validity of COHIP assessed with Spearman correlation between COHIP-ortho and COHIP-38 scores

| | COHIP-ortho items | | |
|---|---|---|---|
| | **df** | **ρ** | **p-value** |
| Oral Health | 241 | 0.74 | 0.00 |
| School | 241 | 0.38 | 0.00 |
| Social Emotional Well-being | 241 | 0.67 | 0.00 |
| Functional Well-being | 241 | 0.38 | 0.00 |
| Peer Interaction | 241 | 0.63 | 0.00 |
| COHIP-38 | 241 | 0.87 | 0.00 |

**Table S4.** Kappa correlations between COHIP-38 and COHIP-ortho scores reverse transformed to a 5-point Likert scale

| | COHIP-38 | | | | | | |
|---|---|---|---|---|---|---|---|
| **COHIP-ortho** | Always | Very often | Sometimes | Almost never | Never | **Total** | |
| Always | 0 | 0 | 0 | 0 | 0 | 0 | |
| Very often | 0 | 0 | 1 | 0 | 0 | 1 | |
| Sometimes | 0 | 0 | 5 | 8 | 0 | 13 | |
| Almost never | 0 | 0 | 1 | 102 | 35 | 138 | |
| Never | 0 | 0 | 0 | 7 | 84 | 91 | |
| Total | 0 | 0 | 7 | 117 | 119 | 243 | Kappa= 0.605, p-value <0.00 |

# Chapter 2.2

## Three-dimensional photographs for determining the Index of Orthodontic Treatment Need in scientific studies

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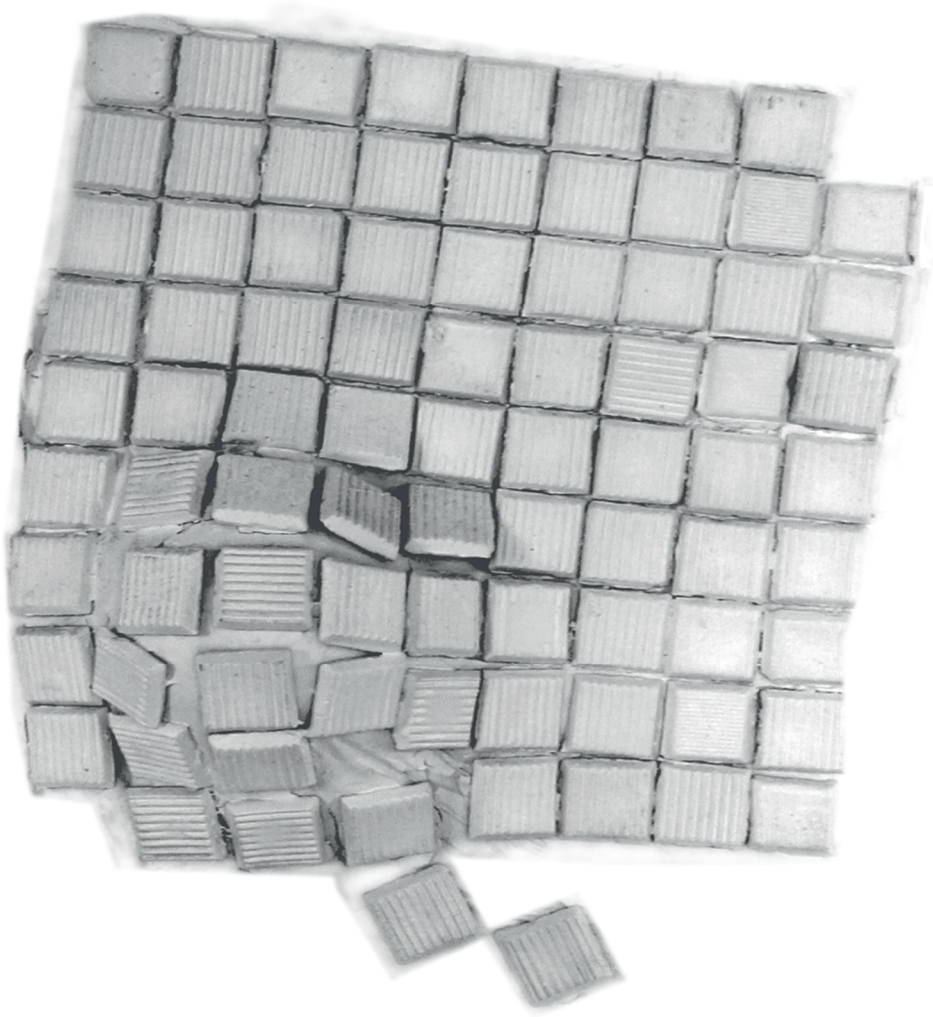

Abstract

*Introduction* Plaster casts as medium for data collection in orthodontic studies pose disadvantages. This study aims to assess the validity and reliability of using 3D photos instead of plaster casts to determine the Index of Orthodontic Treatment Need (IOTN).

*Methods* Data were collected retrospectively from clinical records of 91 subjects. The IOTN grades were independently determined first from plaster casts, second from 2D and 3D photos only, and third from 2D and 3D photos combined with radiographs. IOTN grade agreement was assessed using kappa statistics and percentage agreement.

*Results* The percentage agreement between both photographic sets and the plaster casts varied among different occlusal traits from 63.7% to 93.4%. Agreement between IOTN grades obtained from 2D and 3D photos only and IOTN grades obtained from plaster casts was fair (K = 0.35). The reliability of using 2D and 3D photos instead of plaster casts was improved when those were combined with radiographs.

*Conclusion* In general terms, orthodontic treatment need can be assessed from 2D and 3D pictures, however the individual occlusal traits are only sufficiently assessed when these pictures are combined with radiographs. Plaster casts remain currently the preferred method compared to 3D pictures for assessment of the IOTN.

Keywords: Orthodontic treatment need, epidemiologic studies, (stereo-) photography, validity

## Introduction

Malocclusions have varying prevalence among different countries but are one of the most common oral health problems worldwide (1-3). Despite the extensive application of orthodontics, large-scale population-based epidemiologic studies, such as prospective cohort studies, that investigate the possible beneficial effects of orthodontic treatment are scarce (4). Whether orthodontic treatment can reduce susceptibility to dental caries, periodontal disease, temporomandibular disorder, traumatic dental injury or psychosocial problems is still inconclusive (5-8). In general, the matter of treatment may eventually be better described as correcting a deviation from the norm than in healing an acute disease and the impact on oral health may occur after many years in which also oral habits, parafunction and other influences on the occlusion might develop (9). This nature of the orthodontic specialty challenges epidemiologic research like long term evaluation of treatment, but it also highlights its importance.

One of the most important parts of epidemiologic studies is a systematic, accurate and credible data collection. Occlusal indices have been proposed as a means of acquiring descriptive orthodontic data, like the Index of Orthodontic Treatment Need (IOTN). The IOTN is very valuable as a simple and quick measure for scientific studies, because it only needs to measure the worst feature of the malocclusion (10-12). In addition, intra- and inter-examiner variability is reduced when calibrated examiners use the IOTN to assess orthodontic treatment need (13).

The IOTN grade is usually obtained during clinical examination both in combination with intra-oral and extra-oral photos, radiographs and plaster casts (14). The agreement of the IOTN applied to plaster casts and the IOTN applied during oral examinations is reported to be high (12). However, clinical examination and taking plaster casts provide both practical constraint to the collection of orthodontic data for example in clinical trials or large-scale observational cohort studies. Although direct intra-oral examination is the gold standard to apply the IOTN in clinical practice, this cannot meet the prerequisite of gathering scientific data in terms of repetition and validation. The dental impression process in turn is time-consuming and unpleasant for study participants.

Application of the IOTN solely based on photographic records, that burden study participants less, would simplify the conduction of large-scale studies. In dental practice, 2D photos are already an important part of orthodontic planning, but are only valid for occlusion judgement when used in combination with plaster casts and radiographs. The advent of 3D imaging techniques may provide opportunities for orthodontic large scale studies without requiring plaster casts. Recently, intraoral scanners were introduced to dental practice to face the disadvantages of traditional impression processes, like patient burden and information errors (15). However, for large population-based cohort studies, with an enormous amount of data collection outside the orthodontic practice and from diverse clinical specialities, like observational cohort studies, these scanners are too time-consuming (16). A cheaper, faster and even more comfortable alternative for epidemiological studies might be given by extra-oral highly accurate surface imaging; however, this possibility of 3D photos has not yet been evaluated in the literature.

The present study aims to investigate the validity and reliability of determining orthodontic treatment need by the IOTN based on 2D and 3D extra-oral photos of the dentition. For this purpose, we compare the grades of the IOTN -dental health component

(DHC) applied to 2D and 3D photos, once combined with radiographs and once without radiographs, with the grades of the IOTN-DHC applied to plaster casts from clinical records.

## Material and Methods

This validation study was reviewed and approved by the METC of the Erasmus MC Rotterdam, the Netherlands (METC-2013-098). Informed consent was given by the participants' parents before the photos were taken. The study is carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans.

### *Participants*

The study population was a convenience sample recruited from one orthodontic practice in the Netherlands. Eligible study participants were all children at the time that their orthodontic treatment was initiated. Children with clefts or other dentofacial deformities were excluded. We conducted a power calculation based on the proposed method by Cicchetti and obtained a minimal required sample size of n = 50 (17). In total 91 children with a mean age of 11.77 ± 1.39 years were included in the present study. All of them were in the mixed dentition stage.

### *Data Collection*

For this validation study, data were retrospectively collected by means of plaster casts and photos. No data were acquired from direct clinical examination. Except for the 3D photo, all materials used in this study were obtained as part of the standard clinical procedure. All assistants were trained and calibrated in taking the photos and dental impressions.

The impression to make plaster casts were taken by the orthodontic practice staff prior to the start of orthodontic treatment. An orthopantomogram (OPG), a cephalogram, and three intra-oral 2D photos were also taken at the start of treatment. Using a Panasonic lumix DMC-TZ7, intra-oral photos were taken from three perspectives: frontal view, left buccal view, and right buccal view. The children were asked to show their teeth with the help of cheek retractors. We also obtained a 3D photo using the 3dMD imaging system and 3dMDvultus Viewer software (3DMD Imaging Equipment, Atlanta, GA). The photo was taken of the face while the child made the teeth visible with cheek retractors. Figure 1 represents a complete radiographic set of a case without the lateral 2D photos.

### *Measure*

The IOTN was used to assess orthodontic treatment need and malocclusions (11, 18). The IOTN recognizes five grades of orthodontic treatment need, ranging from no need (1) to very great need (5). There are two IOTN components: the Aesthetic Component (AC) and the Dental Health Component (DHC). This study focused on the DHC, which categorizes the detrimental effects of various deviant occlusal traits (11). When using the DHC of the IOTN, only the most severe grade and specification of the deviant occlusal trait is typically recorded (final IOTN-DHC grade). However, we documented addi-

tionally every applicable trait specification in order to identify the particular benefits of 3D photos as well as the possible sources of disagreement in retrospect.
The IOTN-DHC employs a systematic and reliable scoring technique using a hierarchical scale based on the following order of severity of deviant occlusal traits: missing teeth, overjet, crossbite, displacement of contact points (DOCP; also referred to as crowding), and overbite (including deep and openbite) (19).
For each participating child, the IOTN grade was assessed three times using the IOTN-DHC scoring based on different materials. First, each patient's plaster cast was assessed by one calibrated examiner in combination with the patient file. Second, the IOTN-DHC grade was determined on intraoral 2D photos in combination with the 3D photo (2D3D-set) by two other calibrated examiners. Third, the IOTN-DHC grade was assessed on the 2D3D-set combined with the radiographs by the same two examiners (radiographic set). Randomly selected subgroups were reassessed by the same and the other examiner as well as the same and the other photographic set.

### *Statistics*

Statistical analyses were performed with SAS 9.3 (SAS Institute, Cary, NC).
We used linear weighted kappa statistics for intra-rater reliability (between same photographic sets), test-retest reliability (between same photographic sets) and inter-method reliability (between different photographic sets) among the 5 grades of the IOTN. Linear weighted kappa was also used for the comparison between grades from plaster cast with the 2D3D-set or radiographic-set, respectively. Linear weights were applied because, with regard to the accuracy of the method, the difference between the second and third IOTN grade is of the same importance as the difference between the third and fourth or between the fourth and fifth category. The IOTN developers recommend analysis of kappa agreement with linear weights for the DHC (20).
The ability to assess different occlusal traits using both sets of photos was evaluated based on the unweighted Cohen's kappa coefficient, because the specifications are nominal data. The occlusal traits per group of the hierarchical scale of the IOTN-DHC that were identified from the two photographic sets were each compared to the occlusal traits determined using the plaster casts. The agreement for all kappa correlations was evaluated using the guidelines suggested by *Landis and Koch (21).*
After we have categorized the different IOTN grades into treatment need (grade > 3) and no treatment need (grade < 3), we calculated sensitivity, specificity, positive predictive value, negative predictive value, and positive and negative likelihood ratio for both photographic sets with the plaster casts as the reference (22). Treatment need was determined using a cut-off value of grade >3 as suggested by *Roberts and Richmond (20).*

## Results

Table I shows the frequencies of the highest IOTN grades when using each of the three different assessment methods. With the IOTN grades obtained from plaster casts, we found an orthodontic treatment need prevalence of 71.4%.
Table II presents the comparison between the final IOTN grade obtained from both sets of photos and the final IOTN grade obtained using the plaster casts. Table III presents

the comparison of deviant occlusal traits identified from both sets of photos and with those from plaster casts.

### *2D3D-set vs. plaster casts*

The percentage of agreement between the 2D3D-set and plaster casts varied among the different occlusal traits from 68.1% to 93.4%.

We found a fair agreement between the IOTN grades obtained from the 2D3D-set and those obtained from plaster casts, with an overall K-value of 0.35 (95% CI: 0.21–0.50, n=91).

**Table 1.** Frequencies of obtained highest IOTN- DHC grades as well as trait specifications based on plaster casts, the 2D3D-set and the radiographic set

| | Plaster casts | 2D3D-set | Radiographic set |
|---|---|---|---|
| **Grades** | | | |
| IOTN grade 2 | 2 | 8 | 4 |
| IOTN grade 3 | 24 | 24 | 23 |
| IOTN grade 4 | 50 | 47 | 50 |
| IOTN grade 5 | 15 | 12 | 14 |
| **Specifications[1]** | | | |
| Missing teeth | 12 | 7 | 8 |
| Overjets | 64 | 58 | 61 |
| Crossbites | 17 | 14 | 13 |
| Displacement of contact points | 90 | 84 | 84 |
| Open bite | 69 | 85 | 82 |
| Other | 15 | 6 | 17 |

[1]number of specifications add up to > N=91 because multiple traits per person possible

**Table 2.** Agreement between the IOTN grades based on photographic sets and the IOTN grades based on plaster casts presented as linear weighted kappa (K [95% CI]) and percentage agreement (%).

| | 2D3D-set vs plaster casts | | | Radiographic set vs plaster casts | | |
|---|---|---|---|---|---|---|
| | **N** | **K** | **%** | **N** | **K** | **%** |
| **Total** | 91 | 0.35 [0.21-0.50] | 53.7 | 91 | 0.44 [0.29-0.59] | 62.6 |
| **Examiner 1** | 49 | 0.34 [0.12-0.56] | 61.2 | 51 | 0.50 [0.29-0.71] | 70.6 |
| **Examiner 2** | 42 | 0.37 [0.19-0.54] | 47.6 | 40 | 0.42 [0.25-0.60] | 52.5 |

Strength of agreement for the Kappa coefficient: ≤0=poor, 0.01-0.20=slight, 0.21-0.40=fair, 0.41-0.60=moderate, 0.61-0.80=substantial, 0.81-1=almost perfect

**Table 3.** Agreement between occlusal trait specifications assessed on plaster casts and the two sets of photos presented as kappa values ( K [95% CI]) and percentage agreement (%).

| | | 2D3D-set | | Radiographic set | |
|---|---|---|---|---|---|
| | **N($n_1$)** | K | **%** | K | **%** |
| **M**issing teeth | 91(15) | 0.28 [0.06-0.56] | 85.7 | 0.59 [0.36-0.82] | 90.1 |
| **O**verjet | 91(64) | 0.67 [0.51-0.82] | 84.6 | 0.69 [0.53-0.84] | 85.7 |
| **C**rossbite | 91(17) | 0.65 [0.44-0.86] | 90.1 | 0.68 [0.48-0.89] | 91.2 |
| **D**isplacement of contact points | 91(90) | 0.24 [0.0-0.62] | 93.4 | 0.24 [0.0-0.62] | 93.4 |
| **O**verbite | 91(69) | 0.30 [0.14-0.46] | 68.1 | 0.22 [0.07-0.38] | 63.7 |
| Other | 91(12) | 0.41 [0.11-0.70] | 89.0 | 0.53 [0.31-0.75] | 86.8 |

[1]number of cases that do have this particular occlusal trait determined on plaster casts

Strength of agreement for the Kappa coefficient: ≤0=poor, 0.01-0.20=slight, 0.21-0.40=fair, 0.41-0.60=moderate, 0.61-0.80=substantial, 0.81-1=almost perfect

The agreement between the 2D3D-set and the plaster casts on the different occlusal traits varied from fair (K = 0.24, 95% CI: 0.0–0.62) for the displacement of contact points to substantial (K = 0.65, 95% CI: 0.0–0.62) for the assessments of crossbites.
With IOTN grades based on the 2D3D-set, the sensitivity of scoring treatment need was 0.75 and the specificity was 0.62, the positive predictive value was 0.83, and the negative predictive values were 0.50. The positive likelihood ratio was 1.96 for the 2D3D-set and the respective negative likelihood ratio was 0.40 (Table IV).

### *Radiographic set vs plaster casts*

The percentage of agreement between the radiographic set and the plaster casts varied among the different occlusal traits from 63.7% to 93.4%.
Agreement between the grades obtained from the radiographic set and the grades obtained from plaster casts was moderate with K = 0.44 (95% CI: 0.29-0.59, n=91).
The agreement between the radiographic sets and the plaster casts varied among occlusal traits and ranged from fair (K = 0.22, 95% CI: 0.07–0.38) for overbites to substantial (K = 0.69, 95% CI: 0.53–0.84) for overjets. The categories 'missing teeth' and 'other' were clearly better assessed by the radiographic set than by the 2D3D-set. Figure 1 clearly shows, how the overjet can be seen on the 3D photo and cephalogram. Displacement of contact points is only visible on the 3D photo and the extent of the open bite can also be seen more accurately on the 3D photo.
With IOTN grades based on the radiographic set, the sensitivity of scoring treatment need was 0.80 and the specificity was 0.54, the positive predictive values was 0.82, and the negative predictive value was 0.52. The positive likelihood ratio was 1.72 for the radiographic set, and the respective negative likelihood ratio was 0.37 (Table IV).

### *Reliability of IOTN applied to photographic sets*

We found substantial intra-rater reliability for Examiner 1 (K = 0.68, 95% CI: 0.43–0.94, *n* = 29) and Examiner 2 (K = 0.66, 95% CI: 0.47–0.99, *n* = 29), indicating good reproducibility of applying the IOTN to both photographic sets. The test-retest reliability was almost perfect for the 2D3D-set (K = 0.80, 95% CI: 0.66–0.95, *n* = 29) and moderate for the radiographic set (K = 0.57, 95% CI: 0.35–0.80, *n* = 29). Inter-method reliability adjusted for the examiners was substantial (K = 0.70 (95% CI: 0.47-0.93, n=45)).
The crosstabs of all validity and reliability analyses are given in the supplemental file (SI to SXI).

**Table 4.** Validity of assessing orthodontic treatment need (IOTN-DHC > 3) by applying the IOTN-DHC to the photographic set compared to applying the IOTN-DHC to plaster casts

| | Sensitivity [95%] | Specificity [95%] | PPV [95%] | NPV [95%] | LR+ [95%] | LR- [95%] |
|---|---|---|---|---|---|---|
| 2D/3D-set | 0.75 [0.64-0.84] | 0.62 [0.43-0.78] | 0.83 [0.74-0.93] | 0.50 [0.33-0.67] | 1.96 [1.18-3.25] | 0.40 [0.24-0.68] |
| Radiographic set | 0.80 [0.69-0.88] | 0.54 [0.36-0.71] | 0.81 [0.72-0.91] | 0.52 [0.33-0.71] | 1.73 [1.13-2.67] | 0.37 [0.20-0.68] |

PPV = positive predictive value; NPV = negative predictive value; LR+ = positive likelihood ratio; LR- = negative likelihood ratio

## Discussion

In the present sample, almost all of the occlusal traits specified by the IOTN were identified on plaster casts, as well as all IOTN grades from 2 to 5. The overall agreement between the plaster cast assessment and the 2D3D-set was fair, while that between the plaster cast assessment and the radiographic set was moderate for the highest IOTN grade, according to the guidelines of *Landis and Koch* (21). Sensitivity and specificity of assessing need for orthodontic treatment (IOTN grade > 3) was satisfactory for both photographic sets with plaster casts assessment as reference.

**Figure 1.** A radiographic set of a participant consisting of the 3D photo presented from different angles, the extra-oral 2D, the orthopantomogram and the lateral cephalogram. A censored complete facial photo is added for completeness.

Because we collected the information of all present occlusal traits in the dentition, we were able to see which occlusal traits lead to the disagreement on the highest IOTN grade. The 2D3D-set performed only in the categories 'missing teeth' and 'other' differently than the radiographic set, which was expected. Since in general the orthodontic problems identified on radiographs around the age of 12 years are often those that

would typically be seen as missing teeth, supernumerary or impacted teeth. In the other different occlusal trait subgroups, the two photographic sets had around the same agreement with the plaster casts. This is in agreement with another study that examined the use of radiographs for identifying orthodontic treatment need, and found that radiographs were only of additional importance in selected cases (27).

Several authors have previously described the value of 2D photos in the identification and evaluation of malocclusion, hard and soft tissue health, and orthodontic treatment need, but have emphasized the limitations regarding accurately measuring space requirements and overjets (24, 25). However in our study, the agreement of both photographic sets with the plaster casts was high for the overjet subgroup, and the agreement of both photographic sets with the plaster casts was fair for DOCP. This showed that the assessment of overjets was better assessed with 3D photos than with 2D photos. 3D photos can be viewed from any desired side, enabling a good inspection of both distances between teeth and overlapping of teeth. Therefore, our fair agreement between both photographic sets and the plaster casts with regard to crowding was most likely based on the use of 3D photos. Altogether, our results and above considerations support an eminent addition of 3D photos for the assessment of IOTN grades in epidemiologic studies. Still, radiographs improve the applicability for the IOTN categories "missing teeth" and "other".

We used plaster casts instead of clinical examination as a gold standard in our study, which somewhat limits our result. However, calibrated examiners ensure high reliability of IOTN grades from both oral examination as well as plaster casts (11, 14, 26, 27) Appropriately, our examiners showed high intra-examiner reliability. However, we did not explicitly assess inter-examiner variability here. Indeed, certain occlusal traits or borderline cases can reduce reliability of using photographic records as likely as when using plaster casts or in oral examination (28). Yet the strength of photographic records is that they can easily be re-evaluated.

One of the main challenges of taking photos for the assessment of orthodontic treatment need is to achieve the correct patient positioning (29). A standardized natural head position is needed to evaluate the right skeletal pattern, i.e. Class 1 to Class 3. The subjects have to bite together correctly to ensure valid open- and deep-bite evaluation. Preventing unwanted light reflections or underexposure requires accurate light settings. Sufficient removal of saliva from the teeth, for example with matting powder, would further improve a study with 3D stereophotogrammetry. Demonstration of occlusion on a photo is more valid when all of these factors are considered (29). With improved external circumstances the use of 3D stereophotogrammetry has the potential to even replace the use of 2D photography.

Despite these concerns, the present study definitely creates opportunities for epidemiological research in the orthodontic area. It is not the first study that responds to the demand of facilitating the assessment of orthodontic treatment need in epidemiologic studies, but previous studies have used different approaches. For example, Ovsenik *et al.* evaluated the use of plaster casts for the general assessment of malocclusions; Burden *et al.* suggested the use of a modified IOTN that is easier to use and would overcome the need to calibrate examiners; and Mok *et al.* took 2D photos of plaster casts (26, 27, 30). However, the present study is the first to assess the possibility of evaluat-

ing orthodontic treatment need solely based on photographic records to promote epidemiologic orthodontic research.

There are several advantages of using only photographic records to assess orthodontic treatment need in epidemiologic large-scale studies. Unlike assessment by clinical examination, photos are repeatedly accessible at any time and place. The use of only photos solves the issue of the great storage space needed for plaster casts. Digital photos are also much more easily portable compared to plaster casts, making them eminently suited for data transmission, which is beneficial for multicentre research. Taking a 3D photo of study participants is less invasive than taking plaster casts and especially for children it might be even fun. Finally, taking photos is a relatively easy, fast, and cost-saving procedure. In the future, it might even be possible to evaluate orthodontic problems with or without the IOTN by using automatic landmarking software.

## Conclusion

The reliability of 2D and 3D photos versus plaster casts is only fair to determine the individual IOTN grades, but suitable to determine a need for orthodontic treatment in general terms. Whereas in the past the evaluation of overjets and crowding was impossible from 2D photos, we show that 3D photos enable an inspection of distances. Finally, the combination of 2D and 3D photos with consideration of radiographs enables better assessment of the full range of deviant occlusal traits.

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## Supplemental Material

IOTN grades assessed on plaster casts and 2D/3D photographic set or radiographic photo set are presented in simple cross tables. The following cross tables are about the validity of using photographic sets for the assessment of the IOTN.

**Table S1**. Cross table of given IOTN grades based on plaster casts and photographic set of both examiners combined

| Both Examiners | | Plaster cast IOTN | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **2D/3D** | **2** | 0 | 2 | 0 | 0 |
| | **3** | 5 | 9 | 10 | 0 |
| | **4** | 2 | 11 | 33 | 4 |
| | **5** | 1 | 2 | 4 | 8 |

Table S1 shows the number of observed agreements: 50 (54.9% of the observations); Kappa= 0.351; SE of kappa = 0.075; 95% confidence interval: 0.205-0.497. The strength of agreement is considered to be 'fair'.

**Table S2.** Cross table of given IOTN grades based on plaster casts and radiographic set of both examiners combined

| Both Examiners | | Plaster cast IOTN | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **2D/3D/x-ray** | **2** | 0 | 1 | 1 | 0 |
| | **3** | 2 | 11 | 11 | 0 |
| | **4** | 2 | 10 | 35 | 3 |
| | **5** | 0 | 1 | 3 | 11 |

Table S2 shows the number of observed agreements: 57 (62.6% of the observations); Kappa= 0.440; SE of kappa = 0.079; 95% confidence interval: 0.285-0.594. The strength of agreement is considered to be 'moderate'.

**Table S3**. Cross table of given IOTN grades based on plaster casts and photographic set of examiner 1

| Examiner 1 | | Plaster cast IOTN | | |
|---|---|---|---|---|
| | | **3** | **4** | **5** |
| **2D/3D** | **3** | 6 | 9 | 0 |
| | **4** | 2 | 22 | 4 |
| | **5** | 0 | 4 | 2 |

Table S3 shows the number of observed agreements: 30 (61.2% of the observations); Kappa= 0.336; SE of kappa = 0.113; 95% confidence interval: 0.115-0.558. The strength of agreement is considered to be 'fair'.

**Table S4.** Cross table of given IOTN grades based on plaster casts and radiographic set of examiner 1

| Examiner 1 | | Plaster cast IOTN | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **2D/3D/x-ray** | **2** | 0 | 0 | 1 | 0 |
| | **3** | 0 | 8 | 11 | 0 |
| | **4** | 0 | 1 | 24 | 1 |
| | **5** | 0 | 0 | 1 | 4 |

Table S4 shows the number of observed agreements: 36 (70.6% of the observations); Kappa= 0.499; SE of kappa = 0.107; 95% confidence interval: 0.290-0.709. The strength of agreement is considered to be 'moderate'.

**Table S5**. Cross table of given IOTN grades based on plaster casts and photographic set of examiner 2

| Examiner 2 | | Plaster cast IOTN | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **2D/3D** | **2** | 0 | 2 | 0 | 0 |
| | **3** | 5 | 3 | 1 | 0 |
| | **4** | 2 | 9 | 11 | 0 |
| | **5** | 1 | 2 | 0 | 6 |

Table S5 shows the number of observed agreements: 20 (47.6% of the observations); Kappa= 0.365; SE of kappa = 0.091; 95% confidence interval: 0.186-0.544. The strength of agreement is considered to be 'fair'.

**Table S6**. Cross table of given IOTN grades based on plaster casts and radiographic set of examiner 2

| Examiner 2 | | Plaster cast IOTN | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **2D/3D/x-ray** | **2** | 0 | 1 | 0 | 0 |
| | **3** | 2 | 3 | 0 | 0 |
| | **4** | 2 | 9 | 11 | 2 |
| | **5** | 0 | 1 | 2 | 7 |

Table S6 shows the number of observed agreements: 21 (52.5% of the observations); Kappa= 0.420; SE of kappa = 0.089; 95% confidence interval: 0.246-0.593. The strength of agreement is considered to be 'moderate'.

IOTN grades assessed on 2D/3D-set or radiographic set are presented in simple cross tables. The following cross tables are about the reliability of using photographic sets for the assessment of the IOTN.

**Table S7.** Cross table of IOTN grades given by examiner 1 for both photographic sets (intra-rater reliability)

| Examiner 1 | | Repeated assessment | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **First assessment** | **2** | 0 | 0 | 0 | 0 |
| | **3** | 1 | 1 | 1 | 0 |
| | **4** | 0 | 1 | 21 | 0 |
| | **5** | 0 | 0 | 1 | 3 |

Table S7 shows the intra-rater reliability for Examiner 1 combined for both photographic sets: 19 (65.5% of the observations); Kappa= 0.68; SE of kappa = 0.129; 95% confidence interval: 0.427-0.936. The strength of agreement is considered to be 'substantial'.

**Table S8**. Cross table of IOTN grades given by examiner 2 for both photographic sets (intra-rater reliability)

| Examiner 2 | | Repeated assessment | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **First assessment** | **2** | 4 | 1 | 1 | 0 |
| | **3** | 4 | 6 | 2 | 0 |
| | **4** | 0 | 0 | 6 | 0 |
| | **5** | 0 | 0 | 2 | 3 |

Table S8 shows the intra-rater reliability for Examiner 2 combined for both photographic sets: 19 (86.2% of the observations); Kappa= 0.66; SE of kappa = 0.097; 95% confidence interval: 0.467-0.994. The strength of agreement is considered to be 'substantial'.

**Table S9.** Cross table of IOTN grades based on 2D3D-set for both examiners (intra-method reliability)

| Both Examiner | | Repeated assessment | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **First assessment** | **2** | 3 | 0 | 0 | 0 |
| | **3** | 2 | 5 | 2 | 0 |
| | **4** | 0 | 0 | 10 | 0 |
| | **5** | 0 | 0 | 2 | 5 |

Table S9 shows the inter-method reliability between both photographic sets: 23 (79.3 % of the observations); Kappa= 0.80; SE of kappa = 0.07; 95% confidence interval: 0.657-0.946. The strength of agreement is considered to be 'substantial'.

**Table S10.** Cross table of IOTN grades based on radiographic set for both examiners (intra-method reliability)

| Both Examiner | | Repeated assessment | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **First assessment** | **2** | 1 | 1 | 1 | 0 |
| | **3** | 3 | 2 | 1 | 0 |
| | **4** | 0 | 1 | 17 | 0 |
| | **5** | 0 | 0 | 1 | 1 |

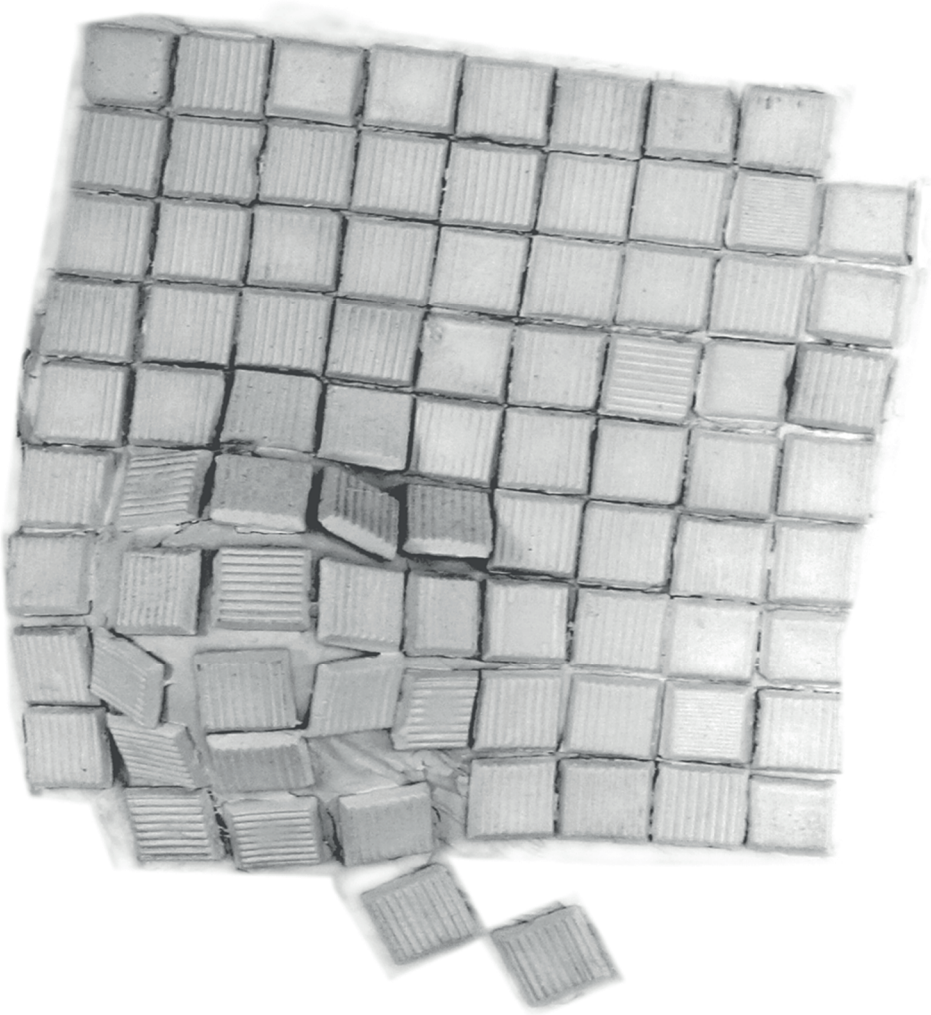
Table S10 shows the inter-method reliability between both photographic sets: 21 (72.4 % of the observations); Kappa= 0.57; SE of kappa = 0.12; 95% confidence interval: 0.345-0.800. The strength of agreement is considered to be 'moderate'.

**Table S11.** Cross table of IOTN grades based on 2D3D-set and radiographic set for both examiners (inter-method reliability)

| Both Examiner | | Repeated assessment | | | |
|---|---|---|---|---|---|
| | | **2** | **3** | **4** | **5** |
| **First assessment** | **2** | 1 | 0 | 0 | 0 |
| | **3** | 0 | 9 | 1 | 0 |
| | **4** | 0 | 2 | 24 | 0 |
| | **5** | 1 | 2 | 0 | 5 |

Table S11 shows the inter-method reliability between both photographic sets: 39 (86.7 % of the observations); Kappa= 0.70; SE of kappa = 0.12; 95% confidence interval: 0.465-0.925 The strength of agreement is considered to be 'substantial'.

# Chapter 3
# Biological and physiological factors influencing OHRQoL



# Chapter 3.1

## The impact of malocclusions on oral health-related quality of life in children - A systematic review and meta-analysis

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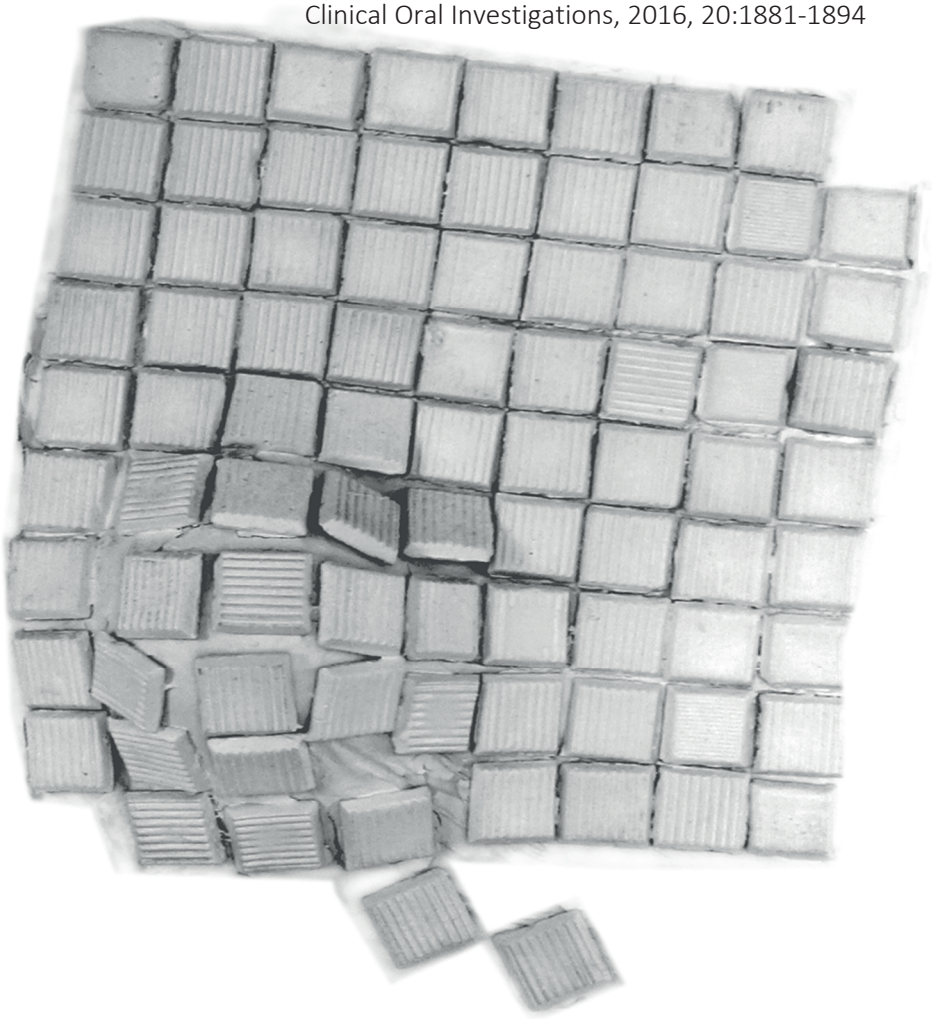
Clinical Oral Investigations, 2016, 20:1881-1894



Abstract

*Introduction* A limited amount of systematic literature reviews on the association between malocclusions and oral health related quality of life (OHRQoL) summarize inconclusive results. Therefore we conduct a systematic review and meta-analysis on the association of malocclusions with OHRQoL in children.

*Methods* Relevant studies were identified in Pubmed, Embase, Cochrane, Google Scholar and other databases. All studies with data on malocclusions or orthodontic treatment need and OHRQoL in children were included. Methodological quality of the studies was assessed with the Newcastle-Ottawa Scale (NOS). Random effects models were used to estimate summary effect measures for the association between malocclusion and OHRQoL in a continuous and a categorical data analysis. Tests for heterogeneity, publication bias and sensitivity of results were performed.

*Results* In total, 40 cross-sectional studies were included in the meta-analyses. Summary measures of the continuous data show that OHRQoL was significantly lowered in children with malocclusions (standardized mean difference (95%CI) = 0.29 (0.19-0.38)). The summary odds ratio for having an impact on OHRQoL was 1.74 times higher in children with malocclusion than in children without malocclusions. Heterogeneity among studies was partly explained by malocclusion assessment, age of the children and country of study conduction.

*Conclusion* Our results provide evidence for a clear inverse association of malocclusion with OHRQoL. We also showed that the strength of the association differed depending on the age of the children and their cultural environment.

*Clinical relevance* Dentists benefit from understanding the patient differences regarding the impact of malocclusions.

*Registration number* CRD42015019522

Keywords: meta- analysis, quality of life, malocclusions, children

## Introduction

Malocclusion is one of the most common oral disorders in the Netherlands. In 2005, half of the Dutch adolescents have had orthodontic treatment, and in 2011 this proportion increased to 60 % (1). A variety of deviant occlusal traits exist that in itself can vary in severity.

The concept of oral health related quality of life (OHRQoL) arose in the orthodontic literature to explain the variability in professionally determined (objectively) and patient-determined (subjectively) need for orthodontic treatment (2, 3). OHRQoL is a patient reported outcome assessed by questionnaires to measure the psychological impact of the dentition. More precisely, OHRQoL is the interplay of oral health variables such as biological and physiological functional status, as well as personal attributes like role functioning, social functioning and psychological functioning, that represents the multidimensional and individual perception of oral health (4). In this way, it describes the standard of the oral and related tissues which enables an individual to eat, speak and socialize without active disease, discomfort or embarrassment and which contributes to general well-being (5). In the last 15 years, the literature on the association of malocclusions and OHRQoL has greatly expanded (6).

Most studies in the orthodontic literature on OHRQoL use small convenience samples, which limits their evidence. In 2006, Zhang et al emphasized the impact of heterogeneous population groups and measurement tools on the conflicting evidence in orthodontic OHRQoL research (3). Indeed, Liu et al reviewed the literature in 2009, but found only a modest association between malocclusion and quality of life among mixed ages (2). A recent meta-analysis on malocclusions, orthodontic treatment and OHRQoL in adults found a moderate increase of OHRQoL after treatment (standardized mean difference (SMD)(95%CI): 1.29 (0.67-1.92)), but the difference in OHRQoL between people with and people without malocclusion was small (SMD(95% CI): 0.84 (0.25-1.43)) (7). Both reviews suffered from the considerable differences in study design.

The impact of malocclusions and OHRQoL might be different in children than in adults as they deal differently with disease, but also with psychological, social and emotional factors (8). In addition, children and adult OHRQoL measures are different, thus they should not be investigated simultaneously. When the focus lies on OHRQoL in children, a variety of instruments exist without one universally accepted. One of the first instruments used in adolescents is the Oral Health Impact Profile-14 (OHIP-14) (9). In 2002, the Child Perception Questionnaire (CPQ) was developed and further developed into the Child Oral Health Impact Profile (COHIP) (8, 10). And in 2004, the Child Oral Impact of Daily Performances (OIDP) was derived from its adult form (11). Finally in 2007, an instrument for very young children was developed, the Early Childhood Oral Health Impact Scale (ECOHIS) (12). These various instruments have much in common, but there are also differences as some focus on the severity whereas other focus on the frequency of oral impacts on OHRQoL, or some instruments make use of parents forms whereas other address the questions directly to the children. A systematic review and meta-analysis with a sufficient amount of studies could explore and explain the influence of differences among studies on the association between malocclusion and OHRQoL. Because the majority of orthodontic patients are children and adolescents, this review

focusses on the relationship of malocclusions or orthodontic treatment need and OHRQoL in subjects up to 18 years old.

## Aim of the study

The primary objective of this study is to give a complete overview on the influence of malocclusion, assessed as occlusal trait or orthodontic treatment need, on OHRQoL measured with validated questionnaires in children and adolescents.
The secondary objective of this study is to explain the differences in the association between malocclusion and OHRQoL in children by investigating sources of heterogeneity among the included studies.

## Material & Methods

The present study was performed according to the guidelines of the PRISMA statement for conducting a systematic review (13). The review protocol can be accessed via the webpage:
http://www.crd.york.ac.uk/PROSPERO/ (Registration number: CRD42015019522).

### *Literature search*

Relevant articles about the impact of malocclusion and orthodontic treatment need on OHRQoL measured by a questionnaire were retrieved by searching Medline OvidSP, Embase, Web-of-sciences, Cochrane central, PsycINFO, OvidSP, Scopus, PsycINFO, Cinahl and finally Google Scholar. The search strategy was built with text words and medical subject headings (MeSH). The main terms were orthodontics, (different) malocclusions, treatment need, quality of life and self-perception. The term self-perception was added to the search strategy to ensure that all articles were found with outcome on OHRQoL. The full search strategy was built with the support of the librarian of the Erasmus Medical Centre and is available in the supplemental material (S1). The search was performed by two reviewers (LK, BD) independently. At first the titles of all articles were screened for their relevance. Here upon the abstracts of relevant articles were retrieved and read. After the abstract selection, full-text copies of the selected papers were retrieved and the final selection for inclusion was made. After both reviewers performed the complete selection procedure, the results of the searches were compared and discussed in case of disagreement.

### *Study selection*

For this systemic review all original and peer-reviewed human studies on the relationship of orthodontic treatment need or malocclusion with OHRQoL in children were searched. The first search was conducted to include all articles until June 2013, a second search was performed to update the relevant articles in September 2014. Finally, the search in PUBMED was repeated in September 2015 to check whether new relevant articles were available. For the selection of studies predefined criteria were used.
All English written studies providing quantitative information about the association of malocclusions with OHRQoL assessed by a questionnaire validated for the use in children were included.

Letters to the editors, conference proceedings, unpublished studies, case reports, and series as well as reviews were excluded from the study selection. When multiple papers were identified on the same population, the study with more information on the data was included in the present review. Studies with participants requiring orthognathic surgery or with syndromic patients were excluded. Also studies using general (health related) quality of life measures were excluded. Studies that only measured the impact of orthodontic treatment, or had a before-after design were excluded when they had no appropriate information on control groups before treatment started. Also studies with children that already had orthodontic treatment or studies that did not use a healthy comparison group (no or less malocclusion resp. orthodontic treatment need) were excluded from this review. Finally, only studies with subjects having a mean age under 18 years were included in this review.
Studies that did not provide sufficient information on number of participants and number of patients with impacts on OHRQoL or means with standard deviation of OHRQoL per subgroup, either directly or to be calculated were excluded from the meta-analysis, but summarized in a narrative way. Studies that assessed orthodontic treatment need only with the Aesthetic Component of the Index of orthodontic treatment need (IOTN-AC) were also excluded from the meta-analysis, but included in the narrative review, because it is not clear whether the IOTN-AC is assessed by the professional or the patient. In figure 1 the flowchart of the study selection is presented. The narrative review is available in the supplement (supplement S2).

*Data extraction*

From the final set of relevant studies, the following data were extracted: study characteristics (first author, publication year, country where the study was conducted, study design, study size, number of cases and controls), description and assessment of the exposure (malocclusion or orthodontic treatment need), description of the outcome assessment (name, length and administration of the questionnaire). For studies that used a continuous OHRQoL measure, the mean and standard deviations of OHRQoL as well as the number of subjects per subgroups were extracted. For studies that used a dichotomous OHRQoL measure, the number of patients with lowered OHRQoL per subgroup as well as the total number of subjects per subgroup were extracted. If a study reported on more than one occlusal index or OHRQoL measure, results from all were extracted to be used for subgroup analysis. For the overall summary measure the results based on the Dental Aesthetic Index (DAI) were used (14-17). One study reported results with two OHRQoL measure, in which the only difference was the specific age group, i.e. CPQ8-10 and CPQ11-14 (18). For this study both results are included in the meta-analysis. In three studies a generic and a condition-specific OHRQoL measure was used, but for the analyses the condition-specific measure was taken only (14, 19, 20).

*Data synthesis*

When data were presented separately for girls and boys these were combined to one group. Mean and standard deviations were re-calculated following the Cochrane Handbook (21). One study did not present results on the overall OHRQoL but presented the result per questions (22). In this case, the OR (95%CI) was calculated per question. Af

terwards, all OR (95%CI) were pooled with a fixed effects meta-analysis and the number of events per subgroup were re-calculated proportional to the sample size of the study. All analysis were performed with a dichotomous independent variable malocclusion (malocclusion vs no malocclusion). Therefore, for the studies that presented their results in more defined subgroups, e.g. a borderline need category, the subgroups were re-grouped following the guidelines of the Cochrane Handbook (21). The following cut-offs for orthodontic treatment need indices were used to indicate no malocclusion: For the Dental Aesthetic Index (DAI) the value of 'minor/none', grade one or a score ≤ 25 was used (14, 16-19, 23-32), for the IOTN the grade ≤ 3 or borderline need was used (14, 17, 20, 33) and for the Index of complexity outcome and need (ICON) a score ≤ 31 or a cut-of value of ≤ 43 was used (17, 34).
OHRQoL was assessed with various questionnaires among the different studies. In general all measures indicated better OHRQoL with a lower score. Only the COHIP indicated better OHRQoL with a higher score (35, 36). In the meta-analysis, the absolute mean differences were used for the results based on the COHIP to make them comparable to the results of other studies in the meta-analysis.

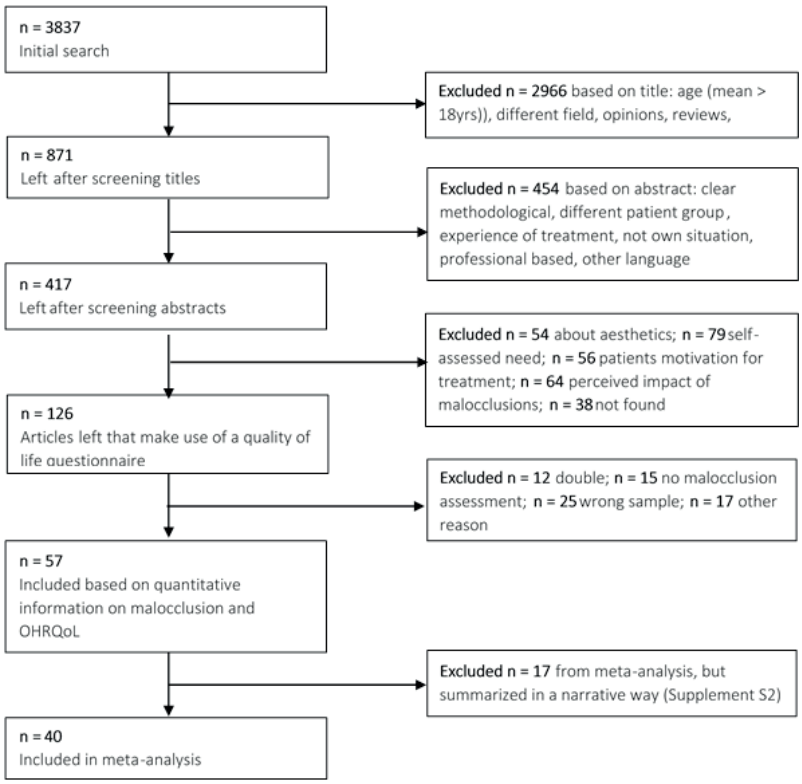



**Figure 1.** Flowchart of study selection

*Quality assessment*

We assessed the methodological quality of the individual studies with the Newcastle-Ottawa Scale (NOS) adapted for cross-sectional studies (37, 38). This scale rates the quality of the included studies on three topics: selection of the study population, comparability of the groups under study and the outcome assessment. The maximum score of this scale is 10, and we assigned high methodological quality to a study if a score > 5 was given.

*Statistical analysis*

Statistical analysis were performed in Review Manager 5.3 from the Cochrane Collaboration.

The studies were analyzed in two ways. On the one hand studies that used a continuous OHRQoL scale (mean ± SD) were grouped in one meta-analysis. On the other hand, studies that used a categorical OHRQoL outcome (no impact vs impact) were grouped into another meta-analysis. This grouping was not mutually exclusive and when possible we included the studies in both meta-analyses.

Random effect models were used for the meta-analyses to calculate summary SMD with 95% CIs for the continuous analyses and summary OR (SOR) with 95% CIs for the categorical analysis.

Heterogeneity was assessed with the $I^{2}$-statistic. The $I^{2}$-statistic quantifies the relative inconsistency between studies. $I^{2}$ values above 50% were considered to indicate substantial inconsistency due to heterogeneity (39). First, studies were grouped based on their outcome measure, i.e. the OHRQoL questionnaire. After that, we stratified the analysis where possible by the following predefined variables to explain heterogeneity and inconsistency in results: Malocclusion assessment, mean age of the study population, country of study conduction and whether the sample was recruited from schoolchildren or from prospective orthodontic patients. Studies using the CPQ as OHRQoL measure were stratified on age specific measurements instead of mean age. We tested for subgroup differences with the $Chi^{2}$-test.

Small study bias, respectively publication bias, was inspected in funnel plots (40). An asymmetric funnel shape was used to inspect a biased relationship between study size and effect size. We performed sensitivity analyses to test the robustness of the summary estimate by omitting one study at a time from the random effects model. We also tested for differences in summary estimates between high and low quality studies for both meta-analysis.

## Results

*Malocclusion assessment and OHRQoL measures of studies included in meta-analysis*

The most commonly (n = 18) used OHRQoL questionnaires were the two CPQs, i.e. for the age group 8-10 years (18, 23, 30, 41) and the age group 11-14 years (17, 18, 24-29, 31, 33, 42-46). Also the OIDP was often (n=9) used in children and adolescents of 10-19 years old (14, 15, 19, 20, 47-51). Five stud ies used the ECOHIS in 1-5 year old children (52-56). Two studies used the COHIP (35, 36) in children from 9-18 years and five studies used the OHIP-14 in children aged 11-17 years (14, 22, 34, 57, 58).

Finally, two studies used the Psychosocial Impact of Dental Aesthetics Questionnaire (PIDAQ) to measure OHRQoL in 12-20 year old children/adolescents (16, 32). One study used additionally the Oral Aesthetic Subjective Impact Scale (OASIS), however this questionnaire is not further considered in this review (44).
Studies used several methods to assess malocclusions or orthodontic treatment need in their study population. Most of the time (n= 19) the DAI was used (14-19, 23-32, 46, 51, 57). The IOTN-DHC was used in 11 studies (14, 16, 17, 20, 22, 33, 35, 36, 44, 48, 58). The ICON was used in two studies (17, 34) and Angles classification system was also used in two studies (43, 47). Two studies assessed the relationship of tooth agenesis and OHRQoL (45, 49). Finally, 9 studies assessed presence of any malocclusion trait or anterior malocclusion trait (15, 41, 42, 50, 52-56).

*Meta-analysis*
In summary, 40 studies, reporting on 41 different samples, were eligible for a quantitative analysis. This resulted in two different meta-analyses, one giving a summary SMD of OHRQoL between children with and children without malocclusions based on 26 studies (figure 2) and the other giving a SOR on the impacts of malocclusions on OHRQoL based on 20 studies (figure 3). The methodological quality of the individual studies ranged from 3 to 8 points (Supplement S3).

*Malocclusions and OHRQoL continuously analyzed*
The summary results show a small but significant SMD in OHRQoL scores between children with malocclusions (n= 7772) and without malocclusion (n= 6549) (SMD: 0.29, 95%CI: 0.19-0.39). We observed high heterogeneity ($I^2$ = 85%) among the studies that were combined for the summary measure on malocclusions and OHRQoL scores, which only partly could be explained by the different OHRQoL measures. Although there were significant differences in summary estimates among the different OHRQoL measures ($X^2$ = 23.07, $p < 0.001$), all indicated a small significant SMD difference in OHRQoL between children with and without malocclusions. Only when OHRQoL was measured with the ECOHIS there was no difference in OHRQoL between children with and without malocclusions (SMD: 0.00, 95%CI = -0.15-0.16).

*Malocclusions and OHRQoL dichotomously analyzed*
The summary result shows that children with malocclusion (n=9293) are 1.74 times more likely to have an impact on OHRQoL than children without malocclusions (n = 10717) (SOR = 1.74, 95% CI = 1.46-2.08). Again we observed high heterogeneity ($I^2$ = 81%) among the studies that were combined for the summary measure on impacts of malocclusions and OHRQoL, which only partly, but more than in the continuous meta-analysis, could be explained by the different OHRQoL measures. The difference in SOR between the different OHRQoL measures was significant ($X^2$ = 33.00, $p < 0.001$) and again when OHRQoL was measured with the ECOHIS, no association was found between malocclusions and OHRQoL.

**Table 1a.** Study characteristics of included studies included in the narrative review and meta-analysis (N=40)

| | Author | Year | Country | Study type | Participants | Age (Mean) | N | OHRQoL | Malocclusion | Ref |
|---|---|---|---|---|---|---|---|---|---|---|
| 1 | Abanto, J.et al. | 2011 | Brasil | cross-sectional | Preschool children seeking dental care | 3.8 | 260 | ECOHIS | Anterior trait | (52) |
| 2 | Aguilar-Diaz, F.C. et al. | 2011 | Mexico | cross-sectional | Children living in San Luis Potosi | 8.98 | 212 | CPQ8-10 | DAI | (23) |
| 3 | Anosike, A.N. et al. | 2010 | Nigeria | cross-sectional | Children randomly selected | 14.5 | 805 | OHIP-14 | DAI | (57) |
| 4 | Asgari, I. et al. | 2013 | Iran | cross-sectional | High-school children | 14.9 | 597 | COHIP | IOTN | (35) |
| 5 | Barbosa, T.S. et al. | 2009 | Brasil | cross-sectional | School children | 9.0/12.5 | 90 /120 | CPQ8-10/11-14 | DAI | (18) |
| 6 | Bhayat, A. et al. | 2014 | Saudi - Arabia | cross-sectional | Male school children | - | 268 | CPQ11-14 | Angles classification | (43) |
| 7 | Bekes, K. et al. | 2012 | Germany | cross-sectional | Recruited from annual screening | 12.3 | 1061 | CPQ11-14 | Presence/Absence | (42) |
| 8 | Bernabe, E. et al. | 2008 | United Kingdom | cross-sectional | School children | 16.42 | 200 | CS-OIDP/OHIP14 | DAI/ IOTN | (14) |
| 9 | Bernabe, E. et al. (1) | 2008 | Brasil | cross-sectional | Students from secondary schools | - | 220 | CS-OIDP | Angles classification | (47) |
| 10 | Bernabe, E. et al. (1) | 2009 | Brasil | cross-sectional | Students from secondary schools | - | 1060 | OIDP/ CS-OIDP | DAI | (19) |
| 11 | Bernabe, E. et al. | 2009 | Thailand | cross-sectional | School children | - | 1034 | OIDP/CS-OIDP | IOTN | (20) |
| 12 | Brown A. et al. | 2006 | Saudi - Arabia | cross-sectional | Dental patients | - | 174 | CPQ11-14 | None/ slight/ moderat/ severe (IOTN) | (33) |
| 13 | Carvalho, A.C. et al. | 2013 | Brasil | cross-sectional | Preschool children | 5.4 | 1069 | ECOHIS | Presence/Absence | (53) |
| 14 | Dawoodbhoy, I. et al. | 2013 | Saudi-Arabia | cross-sectional | Hospital volunteers | 12.6 | 278 | CPQ11-14 | DAI | (24) |
| 15 | Feu D. et al. | 2010 | Brasil | cross-sectional | Prospective patients & school children | 13.6 | 194 | OHIP-14 | IOTN-DHC/ AC-IOTN | (58) |
| 16 | Foster Page, L.A. et al. | 2005 | New Zealand | cross-sectional | School children | 12.7 | 430 | CPQ11-14 | DAI | (25) |
| **17** | Foster Page, L.A. et al. | 2013 | New Zealand | cross-sectional | Dunedin Adolescents | - | 353 | CPQ11-14 | DAI | (26) |
| **18** | Ghijselings, I. et al. | 2014 | Belgium | cross-sectional | Prospective orthodontic patients | 13.8 | 386 | CPQ11-14/ OASIS | IOTN-DHC/ IOTN- AC | (44) |
| **19** | Gomes, M.C. et al. | 2014 | Brasil | cross-sectional | Pre-school children | 3.95 | 843 | ECOHIS | Presence/Absence | (54) |
| **20** | Herkrath, F.J. et al. | 2012 | Brasil | cross-sectional | School children | 12 | 201 | C-OIDP | IOTN-DHC, DAI | (48) |
| **21** | Hvaring, C.L. et al. | 2014 | Norway | cross-sectional | Patients | 13.6 | 163 | OIDP | Hypodontia | (49) |
| **22** | Kolawole, et al. | 2011 | Nigeria | cross-sectional | School children | 12.54 | 248 | CPQ11-14 | DAI | (27) |
| **23** | Kragt, et al. | 2015 | Netherlands | cross-sectional | Referred to orthodontic clinic | 11.85 | 243 | COHIP | IOTN | (36) |
| **24** | Kramer, P.F. et al. | 2013 | Brasil | cross-sectional | School children | 3.5 | 1036 | ECOHIS | Presence/Absence | (55) |
| **25** | Laing, E. et al. | 2010 | United Kingdom | cross-sectional | Prospective orthodontic patients | 13.6 | 123 | CPQ11-14 | Hypodontia | (45) |

**Table 1b.** Study characteristics of included studies included in the narrative review and meta-analysis (N=40)

| | Author | Year | Country | Study type | Participants | Age (Mean) | N | OHRQoL | Malocclusion | Ref |
|---|---|---|---|---|---|---|---|---|---|---|
| **26** | Locker, D. et al. | 2007 | Canada | cross-sectional | Prospective orthodontic patients | 12.5 | 141 | CPQ11-14 | DAI/ PAR | (28) |
| **27** | Manijth, C.M. et al. | 2012 | India | cross-sectional | Adolescents seeking orthodontic treatment | 13.0 | 200 | OHIP-14 | IOTN-DHC | (22) |
| **28** | Marques, L.S. et al. | 2006 | Brasil | cross-sectional | School children | 13.2 | 333 | OIDP | DAI / any malocclusion | (15) |
| **29** | Mbawalla, H. S. et al. | 2011 | Tanzania | cross-sectional | School children from two sides | 13.0 | 2678 | CS-OIDP | Presence/Absence | (50) |
| **30** | Montiel-Company, J.M. et al. | 2013 | Spain | cross-sectional | School children | 13.25 | 627 | PIDAQ | IOTN/ DAI | (16) |
| **31** | Onyeaso, et al. | 2009 | Nigeria | cross-sectional | School children | - | 274 | OHIP-14 | ICON | (34) |
| **32** | Paula, J.S. et al. | 2012 | Brasil | cross-sectional | School children | - | 515 | CPQ11-14 | DAI | (46) |
| **33** | Paula Jr, et al. | 2011 | Brasil | cross-sectional | Adolescents from public high school | 16.1 | 301 | PIDAQ | DAI | (32) |
| **34** | Peres, K.G. et al. | 2013 | Brasil | cross-sectional | Brasilian Oral Health Survey (SBBrasil 2010) | 16.84 | 5445 | OIDP | DAI | (51) |
| **35** | Sardenberg F. et al. | 2013 | Brasil | cross-sectional | School children | - | 1204 | CPQ8-10 | Presence/Absence | (41) |
| **36** | Scapini, A. et al. | 2013 | Brasil | cross-sectional | School children | - | 632 | CPQ11-14 /ISF:20 | DAI | (29) |
| **37** | Scarpelli, A.C. et al. | 2013 | Brasil | cross-sectional | Preschool children | - | 1632 | ECOHIS | Presence/Absence | (56) |
| **38** | Schuch, H.S. et al. | 2014 | Brasil | cross-sectional | School children | 9.15 | 750 | CPQ8-10 | DAI | (30) |
| **39** | Ukra, A et al. | 2013 | New Zealand | cross-sectional | School children | - | 783 | CPQ11-14 | DAI | (31) |
| **40** | Zang , M. et al. | 2009 | China | cross-sectional | sample seeking orthodon-tic treatment | 13.2 | 121 | CPQ11-14 | DAI/ICON/IOTN | (17) |

*Subgroup analyses*

We performed several subgroup analyses in both meta-analyses to understand the heterogeneity among the studies assessing the association between malocclusion and OHRQoL. Subgroup analysis based on method of malocclusion assessment reduced only slightly heterogeneity in summary esti mates, but we found significant differences in summary estimates between the subgroups (continuously: $X^2 = 12.92$, df = 3, p = 0.005; dichotomous: $X^2 = 18.07$, df = 4, p = 0.001). In the continuous analysis the association between malocclusion and OHRQoL scores was lost, when malocclusions assessment was based on hypodontia or simply presence/absence of any malocclusion trait. In contrast, the dichotomous analysis shows that children with malocclusion based on hypodontia are most likely to have any impact on OHRQoL compared to children with malocclusions based on other assessments.

Subgroup analysis based on age of the participants reduced heterogeneity to a bigger extend and we found significant differences in summary estimates between the subgroups (continuously: $X^2 = 25.98$, df = 3, $p < 0.001$; dichotomous: $X^2 = 27.58$, df = 3, $p < 0.001$). In the continuous as well as in the dichotomous analysis we could not see a significant association of malocclusions and OHRQoL in children of age < 8 years. Children between 11-14 years old were the most likely to have an impact of malocclusions on OHRQoL (SOR= 2.28, 95%CI= 1.61-3.24), whereas the biggest difference in OHRQoL scores was seen in children older than 14 years old (SMD=0.59, 95%CI= 0.40-0.78).

After stratification based on country of study conduction we did not found significant differences between subgroups in the continuous meta-analysis, but we did between the subgroups in the dichotomous analysis (continuously: $X^2 = 11.50$, df = 6, p = 0.07; dichotomous: $X^2 = 13.57$, df = 4, p = 0.009).

In general, children with malocclusion were significantly more likely to have lower OHRQoL than children without malocclusions among all countries, except for the studies conducted in Nigeria/Tanzania, where the association based on the continuous analysis goes in the other direction (SMD= -0.06 95%CI = -0.30-0.17). Stratification based on sample recruitment neither reduced heterogeneity nor showed differences between the subgroups.

*Publication bias*

We investigated publication bias visually with funnel plots for both overall meta-analyses. No indication for bias was given. The funnel plots for the continuous and categorical meta-analyses are presented in the supplement (supplement S4).

*Sensitivity analyses*

No or only little differences appeared in the summary estimates, when one of the studies was omitted. None of the changes in the summary estimates were significant (Supplement S5).There were no significant differences in summary estimates between studies of high and low methodological quality (NOS score < 5) in both meta-analyses (table 2).

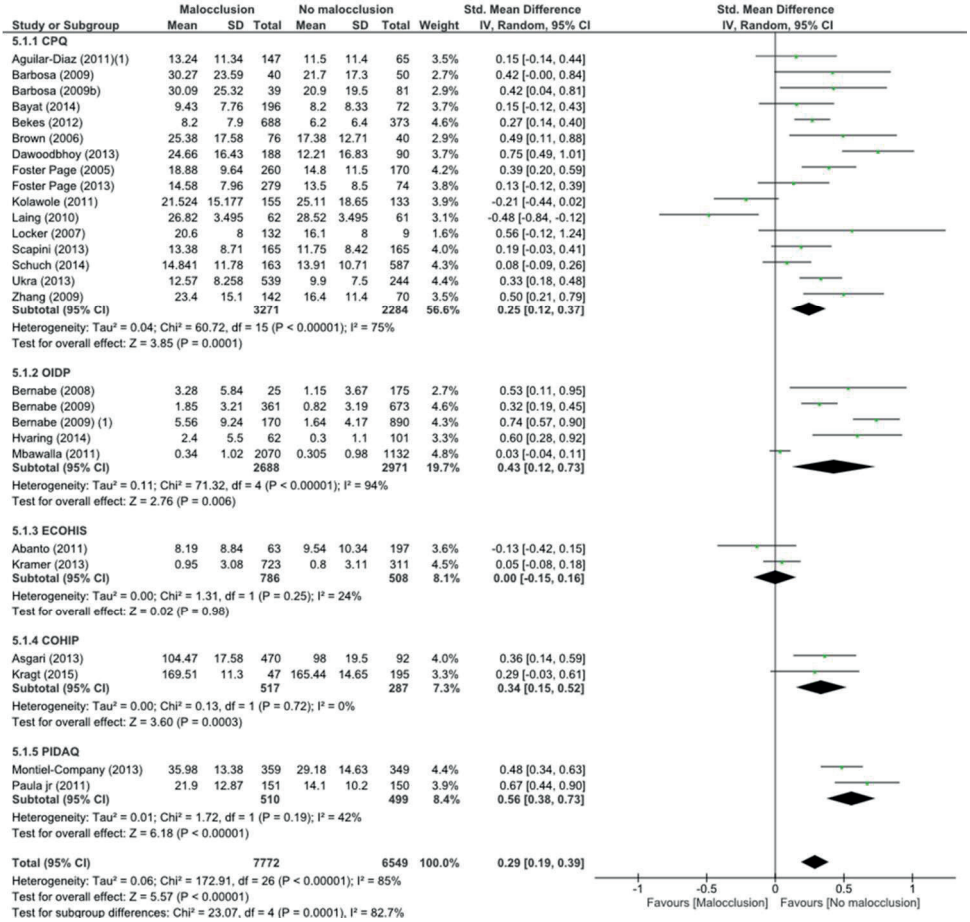



**Figure 2.** Forest plot and summary measure of the association between malocclusions/orthodontic treatment need and OHRQoL measured with different questionnaires (continuous).

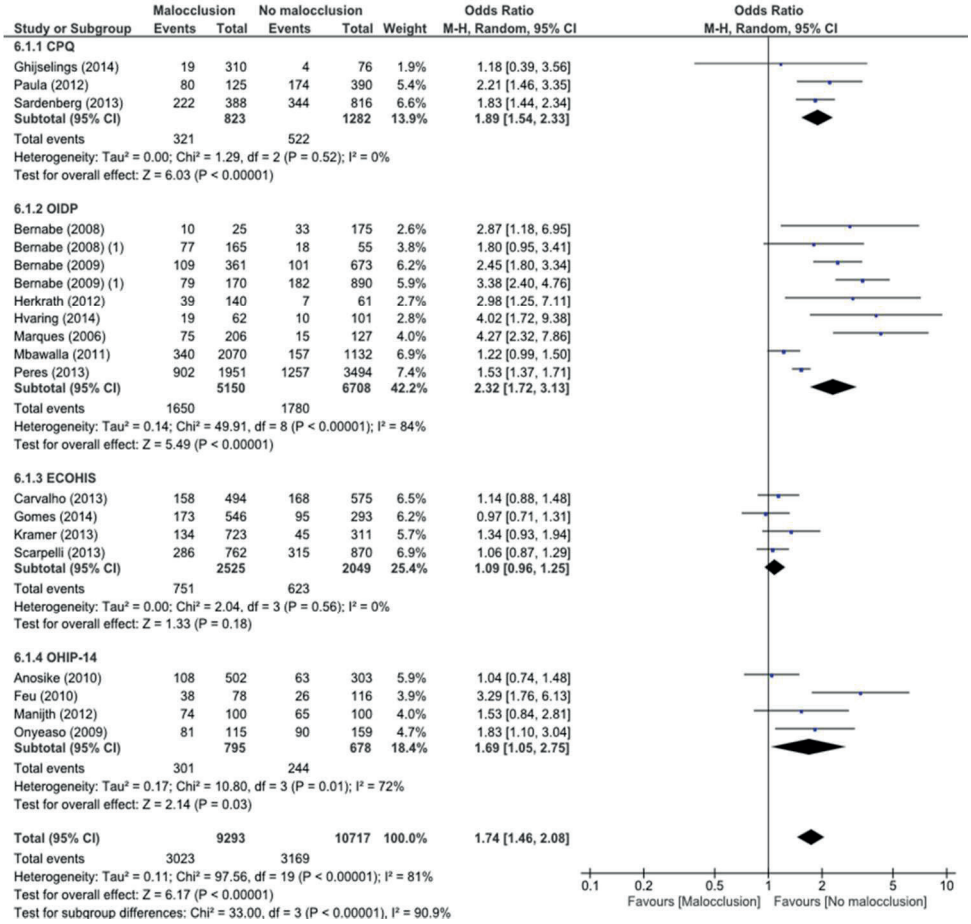



**Figure 3.** Forest plot and summary measure of the association between malocclusions/orthodontic treatment need and OHRQoL measured with different questionnaires (dichotomous)

## Discussion

In this meta-analysis we show that malocclusions in children and adolescents between the age of 8-18 years are associated with lowered OHRQoL. We clearly see an impact of malocclusions on OHRQoL, albeit this impact seems small. There was high heterogeneity among the studies included in the present meta-analysis, which was partly explained by different factors.

OHRQoL "reflects people's comfort when eating, sleeping and engaging in social interaction; their self-esteem and their satisfaction with respect to their oral health" (4). Thus, it encompasses the physical, social and psychological aspects of oral health. Consequently, OHRQoL is suggested to be a multidimensional concept, influenced by individual factors and not stable, but dynamic, over time. This idea is supported by our subgroup analyses, as we show significant differences in the association of malocclusions and OHRQoL among several subgroups.

Firstly, we have shown that the age of the children had a major influence on the association between malocclusions and OHRQoL. Children between the age of 11-14, the age when they undergo major life changes, were most likely to have any impact of malocclusions on OHRQoL, but children older than 14 years showed the biggest impact of malocclusions on OHRQoL. In contrast, we did not see any association of malocclusions with OHRQoL in the younger age groups. Correspondingly we could not see a relationship between malocclusions and the OHRQoL measure designed for and commonly used in toddlers, the ECOHIS (12). Thus, based on our results it seems that the older the children get, the more their malocclusion affects their OHRQoL and this relation gets at first evident around the children's age of eight years old. Longitudinal cohort studies that follow children from the age of 8 years into adulthood would contribute to a better understanding of the dynamics within the relationship of malocclusion with OHRQoL.

Secondly, we also showed differences in the association of malocclusion and OHRQoL between the countries of study conduction, which reflects possible cultural differences. We think that cultural differences may be expressed in both the perception of malocclusions, as well as in the interpretation of OHRQoL. This is in agreement with the World Health Organization Quality of Life group describing quality of life as an 'individuals perception of his/her position in life in the context of culture and value systems in which they live (…)' (59). Also other authors suggested that the perception of oral health, in this case malocclusion, and its influence on OHRQoL might be influenced by the local health care system, which adds to the explanation of the differences in the association of malocclusion with OHRQoL between countries (60). Finally, the effect of malocclusions on OHRQoL might depend on how prevalent other oral diseases are and how important dental aesthetics are seen in certain sociocultural structures, which could explain the big difference in the association of malocclusions with OHRQoL between Brazil and African countries. In general, children and their parents may have problems to relate malocclusion to oral health as most orthodontic conditions are asymptomatic (6, 61). This would explain why we see a clear but relatively small differences in OHRQoL scores between children with malocclusions and children without malocclusions.

The size of the overall summary SMD obtained in the present meta-analysis likely reflects changes in one or two questions of OHRQoL measures.

The association of malocclusion and OHRQoL is based on several ideas. Patients with severe or long-term untreated malocclusions, might suffer from pain due to temporomandibular disorders or dental trauma (3, 62, 63). Malocclusion might also cause functional problems, like problems with speaking, mastication and subsequent restricted food choice (3, 64). Most often however, researchers write about the impact of malocclusions on the social-emotional domain of OHRQoL. This domain reflects appearance of the dentition and related bullying, reduced self-esteem related to oral health, and being ashamed of laughing or in interaction with peers (3, 65-67). We could not investigate these subdomains of OHRQoL individually in this meta-analysis. However, we showed that the associations between malocclusions and OHRQoL varied among the different subgroups of malocclusion assessments. Those assessment methods focus on different aspects of the occlusion, and therefore the associations within this subgroups could be translated to a certain domain of OHRQoL. In our meta-analysis we saw the biggest difference in OHRQoL scores between children with and children without malocclusions, when the latter was assessed with the DAI. The DAI is an orthodontic treatment need index based on socially defined aesthetic standards (68). This supports, that malocclusions largely impact the social emotional domain of OHRQoL. In addition, we have seen in our narrative review that some evidence about the association of the IOTN-AC with OHRQoL points in the same direction. Also, our narrative review points to a missing association between the IOTN-DHC or ICON and OHRQoL (Supplement S2). The IOTN-DHC and ICON do measure malocclusion traits that might not be related to the domains of OHRQoL, like crossbites or impacted teeth in an early stage.

This is the first meta-analysis on the association of malocclusions or orthodontic treatment need and OHRQoL in children and adolescents. An important factor in meta-analysis is the quality of the included studies. We did not exclude studies based on their methodological quality, because our main aim was to give a complete comprehensive overview of the topic. All studies in this meta-analysis were cross-sectional, which is considered to be the study design of lowest quality because of its susceptibility to reverse causation. However reverse causation is not matter of concern to the association of malocclusion with OHRQoL. In addition we evaluated the methodological quality of the individual studies and we did not find significant differences between studies of high and low quality. Generally, we extracted the descriptive data from the selected articles.

Therefore our data are all crudely analyzed, without adjustments for confounders like gender, social economic status (SES) or other oral diseases. However, in this way we were able to include a maximum of studies. We also did not adjust the results for whether the OHRQoL instruments address the questions directly to the children (OIDP, CPQ, OHIP) or make use of parent forms (ECOHIS, COHIP). Accordance between parental and child reports on OHRQoL is widely described in the literature. Especially in orthodontics, parents are seen as valid proxies for the assessment of their children's OHRQoL (69-72). If discrepancies between parents and children assessments exist, children tend to report their own OHRQoL higher than their caregivers do, which means that the associations between children’s malocclusion and OHRQoL assessed by parents are rather underestimated than overestimated. Though, we conducted the meta-analysis on 40 studies including 28496 children and therefore we think that the benefits of the quantitative analysis outweigh the limitations of this meta-analysis.

**Table 2.** Subgroup analysis of Malocclusions/orthodontic treatment need and OHRQoL

| | | Continuous (overall hetergoneity 85 %) | | | | | Dichotomous (overall hetergoneity 81 %) | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| **Stratum** | **Subgroup** | **N[a]** | **SMD[b] (95%CI)** | **I²-statistic[c]** | **X²-statistic[d]** | **p-value[e]** | **N[a]** | **OR (95%CI)** | **I²-statistic[c]** | **X²-statistic[d]** | **p-value[e]** |
| **Malocclusion** | DAI | 16 | 0.37 (0.24, 0.51) | 81% | | | 7 | 2.24 (1.54, 3.26) | 85% | | |
| | IOTN | 4 | 0.34 (0.24, 0.44) | 0% | | | 4 | 2.20 (1.55, 3.13) | 34% | | |
| | ICON | - | - | - | | | 1 | 1.83 (1.10, 3.04) | 64% | | |
| | Hypodontia | 2 | 0.06 (-1.00, 1.12) | 95% | | | 1 | 4.02 (1.72, 9.38) | - | | |
| | Presence/Absence | 5 | 0.09 (-0.03, 0.21) | 69% | 12.92 | 0.005* | 7 | 1.26 (1.06, 1.50) | 64% | 18.07 | 0.001* |
| **Age** | < 8 years | 3 | 0.04 (-0.08, 0.15) | 2% | | | 4 | 1.09 (0.96, 1.25) | 0.0% | | |
| | 8-10 years | 2 | 0.19 (-0.11, 0.50) | 51% | | | 1 | 1.83 (1.44, 2.34) | - | | |
| | 11-14 years | 18 | 0.28 (0.16, 0.40) | 83% | | | 9 | 2.28 (1.61, 3.24) | 77.0% | | |
| | > 14 years | 4 | 0.59 (0.40, 0.78) | 59% | 25.98 | < 0.001* | 6 | 1.85 (1.30, 2.63) | 81.0% | 27.58 | < 0.001* |
| **Sample** | School children | 19 | 0.27 (0.16, 0.38) | 86% | | | 15 | 1.71 (1.37, 2.14) | 84% | | |
| | Prospective patients | 5 | 0.28 (-0.12, 0.68) | 83% | | | 4 | 2.30 (1.34, 3.95) | 51% | | |
| | Other | 3 | 0.46 (0.06, 0.86) | 82% | 0.77 | 0.44 | 1 | 1.53 (1.37, 1.71) | - | 2.63 | 0.27 |
| **Country** | Brazil/Mexico | 10 | 0.30 (0.10, 0.51) | 87% | | | 12 | 1.77 (1.41, 2.23) | 85% | | |
| | New Zeeland | 3 | 0.31 (0.18, 0.44) | 22% | | | - | - | - | | |
| | Canada | 1 | 0.56 (-0.12, 1.24) | - | | | - | - | - | | |
| | European Countries | 5 | 0.25 (0.00, 0.51) | 85% | | | 3 | 2.58 (1.32, 5.02) | 34% | | |
| | Saudi-Arabia/Iran | 4 | 0.44 (0.18, 0.69) | 71% | | | | | | | |
| | China/Thailand | 2 | 0.36 (0.22, 0.50) | 16% | | | 1 | 2.45 (1.80, 3.34) | - | | |
| | India | - | - | - | | | 1 | 1.53 (0.84, 2.81) | - | | |
| | Nigeria/Tanzania | 2 | -0.06 (-0.30, 0.17) | 75% | 11.50 | 0.07 | 3 | 1.25 (0.98, 1.58) | 37% | 13.57 | 0.009* |
| **Methodological quality** | Low | 8 | 0.21 (0.06, 0.36) | 70% | | | 4 | 1.42 (1.02, 1.97) | 28 % | | |
| | High | 18 | 0.31 (0.19, 0.44) | 85% | 1.05 | 0.31 | 16 | 1.80 (1.47, 2.21) | 83 % | 1.48 | 0.22 |

[a] N=number of studies included in subgroups; [b]SMD=standardized mean difference; [c]$I^2$- statistic indicated heterogeneity among studies within subgroup; [d+ e]$X^2$ -statistic and p-value indicates differences between subgroups ; * = indicates significance

Several systematic reviews about malocclusions and OHRQoL have been conducted, however to our knowledge only two have restricted their studies to children and adolescents (2, 3, 6, 7, 73). Barbosa and Gaviaõ wrote about contradictory results among 6 studies on the association of malocclusions and OHRQoL (6). Dimberg et al have recently published a systematic review on the association of malocclusions and OHRQoL and tried to limit variability by restricting their review to high quality studies (n = 6) (73). In both reviews, the researchers suggest that the effect of malocclusions is mainly on the social emotional wellbeing domain, but they can only speculate on other sources of inconsistency (6, 73). The strength of our study is, that we are able to explain some sources of this variability in the association between malocclusion and OHRQoL. Another strength of our study is, that we analyzed both dichotomous and continuous data on the association of malocclusions and OHRQoL. In this way we maximized the amount of included studies. In addition, we could not only write about whether there is an impact of malocclusions on OHRQoL, but we make conclusion about the size of the impact. Finally, we have also shown that the results of our summary measures are robust and have not been affected by publication bias.

Unfortunately in this systematic review, we were not able to focus on more personal factors influencing the association of malocclusions with OHRQoL, like SES, gender or self-esteem. Only one study, included in the narrative review, noticed that the association between malocclusion and OHRQoL is attenuated in children with low self-esteem (74). This might be one reason why we could not explain all heterogeneity among the studies. However, we noticed increasing research interest in the modifying role of personal factors in the association of malocclusions with OHRQoL and based on this meta-analysis we highly recommend to continue this research strand.

## Conclusion

The association of malocclusion and OHRQoL has mainly been assessed in cross-sectional studies. From these studies it can be concluded that children perceive a small impact of malocclusions on OHRQoL. The effect of malocclusions on OHRQoL is modified by the age of the children and their cultural environment. Further research should investigate whether remaining heterogeneity in the association of malocclusions with OHRQoL can be explained by other individual factors of the children.

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## Supplemental Material

### 1. Search Strategy

*Embase*
('quality of life'/exp OR 'self concept'/exp OR 'psychological well being'/de OR ((qualit* NEAR/3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR 'facial profile' OR psycholog*) NEAR/3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self NEXT/1 (concept* OR esteem* OR percept* OR perceiv*)) OR 'personal appearance' OR 'well being' OR wellbeing OR (social NEAR/3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) NEAR/6 (treatment* OR intervention*))):ab,ti) AND (orthodontics/de OR malocclusion/de OR (orthodont* OR (occlus* NEAR/3 adjust*) OR malocclusion* OR (palat* NEAR/3 expan*) OR (bite NEAR/3 correction*) OR (serial NEAR/3 extraction*) OR overbite OR underbite OR (occlus* NEAR/3 disorder*)):ab,ti)

*Medline OvidSP*
("quality of life"/ OR exp "self concept"/ OR "Personal Satisfaction"/ OR ((qualit* ADJ3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR "facial profile" OR psycholog*) ADJ3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self ADJ (concept* OR esteem* OR percept* OR perceiv*)) OR "personal appearance" OR "well being" OR wellbeing OR (social ADJ3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) ADJ6 (treatment* OR intervention*))).ab,ti.) AND (orthodontics/ OR exp "Orthodontics, Corrective"/ OR exp malocclusion/ OR (orthodont* OR (occlus* ADJ3 adjust*) OR malocclusion* OR (palat* ADJ3 expan*) OR (bite ADJ3 correction*) OR (serial ADJ3 extraction*) OR overbite OR underbite OR (occlus* ADJ3 disorder*)).ab,ti.)

*Cochrane central*
(((qualit* NEAR/3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR 'facial profile' OR psycholog*) NEAR/3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self NEXT/1 (concept* OR esteem* OR percept* OR perceiv*)) OR 'personal appearance' OR 'well being' OR wellbeing OR (social NEAR/3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) NEAR/6 (treatment* OR intervention*))):ab,ti) AND ((orthodont* OR (occlus* NEAR/3 adjust*) OR malocclusion* OR (palat* NEAR/3 expan*) OR (bite NEAR/3 correction*) OR (serial NEAR/3 extraction*) OR overbite OR underbite OR (occlus* NEAR/3 disorder*)):ab,ti)

*Web-of-science*
TS=((((qualit* NEAR/3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet*

OR esthet* OR appear* OR attractive* OR "facial profile" OR psycholog*) NEAR/3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self NEAR/1 (concept* OR esteem* OR percept* OR perceiv*)) OR "personal appearance" OR "well being" OR wellbeing OR (social NEAR/3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) NEAR/6 (treatment* OR intervention*)))) AND ((orthodont* OR (occlus* NEAR/3 adjust*) OR malocclusion* OR (palat* NEAR/3 expan*) OR (bite NEAR/3 correction*) OR (serial NEAR/3 extraction*) OR overbite OR underbite OR (occlus* NEAR/3 disorder*)))) AND DT=(Article)

*Scopus*

TITLE-ABS-KEY((((qualit* W/3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR "facial profile" OR psycholog*) W/3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self PRE/1 (concept* OR esteem* OR percept* OR perceiv*)) OR "personal appearance" OR "well being" OR wellbeing OR (social W/3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) W/6 (treatment* OR intervention*)))) AND ((orthodont* OR (occlus* W/3 adjust*) OR malocclusion* OR (palat* W/3 expan*) OR (bite W/3 correction*) OR (serial W/3 extraction*) OR overbite OR underbite OR (occlus* W/3 disorder*)))) AND DOCTYPE(ar)

*PsycINFO OvidSP*

("quality of life"/ OR "Life Satisfaction"/ OR "Well Being"/ OR exp "self concept"/ OR "Self Perception"/ OR exp "Satisfaction"/ OR Dissatisfaction/ OR ((qualit* ADJ3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR "facial profile" OR psycholog*) ADJ3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self ADJ (concept* OR esteem* OR percept* OR perceiv*)) OR "personal appearance" OR "well being" OR wellbeing OR (social ADJ3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) ADJ6 (treatment* OR intervention*))).ab,ti.) AND ((orthodont* OR (occlus* ADJ3 adjust*) OR malocclusion* OR (palat* ADJ3 expan*) OR (bite ADJ3 correction*) OR (serial ADJ3 extraction*) OR overbite OR underbite OR (occlus* ADJ3 disorder*)).ab,ti.) NOT book.pt.

*PubMed publisher*

((quality of life*(tiab) OR QoL(tiab) OR HRQL(tiab) OR HRQoL(tiab) OR OHRQL(tiab) OR OHRQoL(tiab) OR ((malocclus*(tiab) OR occlus*(tiab) OR overbite(tiab) OR underbite(tiab) OR emotion*(tiab) OR aesthet*(tiab) OR esthet*(tiab) OR appear*(tiab) OR attractive*(tiab) OR facial profile OR psycholog*(tiab)) AND (impact(tiab) OR percept*(tiab) OR perciev*(tiab) OR aware*(tiab) OR satisf*(tiab) OR dissatisf*(tiab))) OR self concept*(tiab) OR self esteem*(tiab) OR self percept*(tiab) OR self perceiv*(tiab) OR personal appearan*(tiab) OR well being*(tiab) OR wellbeing(tiab) OR social function*(tiab) OR ((desire*(tiab) OR demand*(tiab) OR need(tiab) OR needs(tiab) OR motivat*(tiab)) AND (treatment*(tiab) OR intervention*(tiab))))) AND ((orthodont*(tiab) OR (occlus*(tiab) AND adjust*(tiab)) OR malocclusion*(tiab) OR (palat*(tiab) AND ex-

pan*(tiab)) OR (bite AND correction*(tiab)) OR serial extraction*(tiab)) OR overbite(tiab) OR underbite(tiab) OR (occlus*(tiab) AND disorder*(tiab)))) AND publisher(sb)

*Cinahl*

(MH "quality of life+" OR MH "self concept+" OR MH "Personal Satisfaction+" OR ((qualit* N3 life) OR QoL OR HRQL OR HRQoL OR OHRQL OR OHRQoL OR ((health OR malocclus* OR occlus* OR overbite OR underbite OR emotion* OR aesthet* OR esthet* OR appear* OR attractive* OR "facial profile" OR psycholog*) N3 (impact OR percept* OR perciev* OR aware* OR satisf* OR dissatisf*)) OR (self N1 (concept* OR esteem* OR percept* OR perceiv*)) OR "personal appearance" OR "well being" OR wellbeing OR (social N3 function*) OR ((desire* OR demand* OR need OR needs OR motivat*) N6 (treatment* OR intervention*)))) AND (MH orthodontics+ OR MH "Orthodontics, Corrective+" OR MH malocclusion+ OR (orthodont* OR (occlus* N3 Nust*) OR malocclusion* OR (palat* N3 expan*) OR (bite N3 correction*) OR (serial N3 extraction*) OR overbite OR underbite OR (occlus* N3 disorder*)))

*Google Scholar*

"quality of life"|"self concept"|psychological|"well being"|wellbeing|impact|
awareness|
satisfaction|dissatisfaction|appearance orthodontics|"occlusive|occlusion adjustment|
disorder"|malocclusion|"bite correction"|"serial extraction"|overbite|underbite

**Table S1.** Study characteristics of studies included in the narrative review but excluded from the meta-analysis (N=17)

| Author | Year | Country | Study type | Participants | Age (Mean) | N | OHRQoL | Malocclusion | Ref |
|---|---|---|---|---|---|---|---|---|---|
| Abanto, J.et al. | 2014 | Brasil | cross-sectional | Pre-school children | 2.53 | 879 | ECOHIS | Presence/Absence | (1) |
| Agou, S. et al | 2008 | Canada | cross-sectional | Children seeking orthodontic treatment | 12.6 | 191 | CPQ11-14 | DAI | (2) |
| Aguilar-Diaz, F.C. et al. | 2011 | Mexico | cross-sectional | Schoolchildren | 8.8 | 315 | CPQ8-10 | DAI | (3) |
| Ahn,Y.S. et al. | 2012 | Korea | cross-sectional | Korean Oral Health Survey 2010 | 11.8 | 2236 | COHIP | IOTN | (4) |
| Bianco, A. et al. | 2010 | Italy | cross-sectional | Students from secondary schools, Catanzaro | - | 530 | child OIDP | IOTN-AC | (5) |
| Castro, R.D.A.L. et al. | 2011 | Brasil | cross-sectional | Students from public schools, Rio de Janeiro | 12.0 | 571 | child OIDP | Presence/Absence | (6) |
| De Baets, E. et al. | 2012 | Belgium | cross-sectional | Healthy child registered for a first consultation at the Orthodontic Department | 13.22 | 223 | CPQ11-14 | IOTN-DHC/ -AC | (7) |
| de Oliveira C.M. et al. | 2008 | United Kingdom | cross-sectional | Referred to orthodontic clinical | 12.21 | 187 | OIDP | IOTN-DHC | (8) |
| de Paula J.S. et al. | 2013 | Brasil | cross-sectional | School children | - | 286 | CPQ11-14 | DAI | (9) |
| Do, et al. | 2008 | Australia | cross-sectional | nested on cohort | - | 842 | CPQ8-10/CPQ11-14 | DAI | (10) |
| Heravi, et al. | 2011 | Iran | cross-sectional | High school students | - | 120 | CPQ | ICON | (11) |
| Johal, A. et al. | 2007 | United Kingdom | cross-sectional | - | - | 90 | CPQ11-14 | overjet/ spacing | (12) |
| Kotecha, et al | 2013 | United Kingdom | cross-sectional | clinical sample + comparison | 12.5 | 116 | CPQ11-14 | Hypodontia | (13) |
| Marshman, et al. | 2005 | United Kingdom | cross-sectional | Consecutive sample | 12.4 | 89 | CPQ11-14 | IOTN | (14) |
| Motamedi, M.R.K. et al. | 2014 | Iran | cross-sectional | Schoolchildren | 13.0 | 336 | CPQ11-14 | IOTN | (15) |
| Sousa, R. V. et al. | 2014 | Brasil | cross-sectional | Preschool children | 3.9 | 732 | ECOHIS | Overbite, -jet, crossbite | (16) |
| Thelen, D.S. et al. | 2011 | Norway | cross-sectional | Adolescents in schools | 17.2 | 493 | OIDP | IOTN-AC | (17) |

## 2. Narrative review of studies not included in meta-analysis

Of all 57 studies, 17 studies could not be included in the meta-analysis, because the mean OHRQoL score was not reported (1, 2, 7-9, 12-15), the number of cases per subgroup were missing (3-6, 10, 11, 16), or orthodontic treatment need was only assessed with the IOTN-AC (5, 7, 15, 17).

### *Malocclusion assessment and OHRQoL measures of studies included in meta-analysis*

The most commonly (n = 10) used OHRQoL questionnaires were the two Child Perception Questionnaires (CPQ), i.e. for the age group 8-10 years (3, 10) and the age group 11-14 years (2, 7, 9-15). Also the Oral Impacts of Daily Performances (OIDP) was often used (n = 4 ) (5, 6, 8, 17). Two studies used the Early Childhood Oral Health Scale (ECOHIS) (1, 16).One study used the Child Oral health impact profile (COHIP) (4).

Studies used several methods to assess malocclusions or orthodontic treatment need in their study population. Most of the time (n= 5) the IOTN-DHC was used (4, 7, 8, 14, 15). The DAI was used in four studies (2, 3, 14, 15). The ICON was used in one study (11). Four studies only assessed presence of any malocclusion trait or anterior malocclusion trait. One study assessed hypodontia (13).

Nine studies used the Aesthetic Component of the Index of Orthodontic treatment need (IOTN-AC), either exclusive or in combination with the IOTN-DHC (5, 7, 15, 17-22).

### *Narrative review*

All studies that investigated the association between malocclusions measured with the DAI and OHRQoL measured by the CPQ found significant lower OHRQoL in children with malocclusions (2, 3, 9, 10). However, these association were mostly weak and one study showed that this association between malocclusion based on DAI scores and CPQ scores was lost in children with low self-esteem (2). All studies that investigated the association between malocclusions measured with the IOTN-DHC and OHRQoL measured by the CPQ found no relationship between OHRQoL and orthodontic treatment need (IOTN-DHC >3 ) (7, 8, 14, 15). Only one study investigated additionally the correlation between IOTN scores and CPQ scores, which was significant but weak (7). One study investigated the relationship of malocclusions measured with the ICON and showed only non-significant higher OHRQoL scores (CPQ) in the severe and moderate group compared to the acceptable group (11). Johal et al found highly statistical significant differences between groups with either an increased overjet or spaced dentition compared to a control group (12). Finally, one study investigated the relationship between tooth agenesis with OHRQoL measured by the CPQ. Tooth agenesis was associated with lower OHRQoL, however there was no correlation with the number of missing teeth (13).

Two studies that were not included in the meta-analysis used the ECOHIS to assess OHRQoL in children up to 5 years old (1, 16). Both studies investigated the relationship between the presence of malocclusion and ECOHIS scores as well as kind of malocclusion with ECOHIS scores, but no significant association between any malocclusion trait and children's OHRQoL was found (16, 23).

One study used the COHIP to assess OHRQoL in children. Ahn et al. related the IOTN-DHC scores with OHRQoL measured by the COHIP and found significant but little worse

OHRQoL in children with orthodontic treatment need (IOTN-DHC > 3) (4). Castro et al used the OIDP to assess OHRQoL in children and found a significant association between inadequate position of the teeth with lower OHRQoL (OIDP > 0) (6).
In total, 4 articles of the studies that were not included in the meta-analyses investigated the relationship between the IOTN-AC and OHRQoL in children (5, 7, 15, 17). Of these, three article did not find a relationship between IOTN-AC scores and OHRQoL measured with the CPQ (15) or the OIDP (5, 17). One article found a significant but weak association between the IOTN-AC and OHRQoL measured with the CPQ (7). In contrast, the studies that were included into the meta-analyses and assessed the IOTN-AC found significant relations between the IOTN-AC scores and OHRQoL (19, 22), except Kragt et al. who did only find a borderline significant relation (20).

**Table S2b.** Quality assessment of studies included in the meta-analysisbased on the Newcastle-Ottawa Scale adapted for cross-sectional studies

| | | Selection: | | | | Comparability | Outcome | | Score |
|---|---|---|---|---|---|---|---|---|---|
| *Artikel* | *Ref* | *Sample representativeness* | *SS* | *Non respondents* | *Exposure assessment* | *Control for differences* | *Outcome assessment* | *Statistics* | |
| Abanto, J.et al. | (23) | 0 | 0 | 1 | 1 | 1 | 1 | 1 | 5/10 |
| Aguilar-Diaz, F.C. et al. | (24) | 0 | 0 | 1 | 2 | 1 | 1 | 1 | 6/10 |
| Anosike, A.N. et al. | (25) | 1 | 0 | 1 | 2 | 0 | 1 | 1 | 6/10 |
| Asgari, I. et al. | (26) | 1 | 1 | 1 | 2 | 0 | 1 | 1 | 7/10 |
| Barbosa, T.S. et al. | (27) | 1 | 0 | 0 | 2 | 0 | 1 | 1 | 5/10 |
| Bhayat, A. et al. | (28) | 1 | 0 | 1 | 1 | 0 | 1 | 1 | 5/10 |
| Bekes, K. et al. | (29) | 1 | 0 | 0 | 1 | 0 | 1 | 0 | 3/10 |
| Bernabe, E. et al. | (30) | 1 | 1 | 1 | 2 | 1 | 1 | 1 | 8/10 |
| Bernabe, E. et al. (1) | (31) | 1 | 1 | 0 | 1 | 1 | 1 | 1 | 6/10 |
| Bernabe, E. et al. (1) | (32) | 1 | 1 | 0 | 2 | 1 | 1 | 1 | 7/10 |
| Bernabe, E. et al. | (33) | 0 | 1 | 1 | 2 | 1 | 1 | 1 | 7/10 |
| Brown A. et al. | (34) | 1 | 0 | 1 | 1 | 0 | 1 | 1 | 5/10 |
| Carvalho, A.C. et al. | (35) | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 7/10 |
| Dawoodbhoy, I. et al. | (36) | 1 | 1 | 0 | 2 | 0 | 1 | 1 | 6/10 |
| Feu D. et al. | (18) | 1 | 1 | 1 | 2 | 1 | 1 | 1 | 8/10 |
| Foster Page, L.A. et al. | (37) | 1 | 1 | 1 | 2 | 0 | 1 | 1 | 7/10 |
| Foster Page, L.A. et al. | (38) | 1 | 0 | 0 | 2 | 1 | 1 | 1 | 6/10 |
| Ghijselings, I. et al. | (19) | 1 | 1 | 0 | 2 | 0 | 1 | 0 | 5/10 |
| Gomes, M.C. et al. | (39) | 1 | 1 | 1 | 1 | 1 | 1 | 1 | 7/10 |
| Herkrath, F.J. et al. | (40) | 1 | 1 | 0 | 1 | 0 | 1 | 1 | 5/10 |
| Hvaring, C.L. et al. | (41) | 0 | 1 | 1 | 2 | 1 | 1 | 1 | 7/10 |
| Kolawole, et al. | (42) | 1 | 1 | 1 | 2 | 0 | 1 | 1 | 7/10 |
| Kragt, et al. | (20) | 1 | 0 | 1 | 2 | 0 | 1 | 1 | 7/10 |
| Kramer, P.F. et al. | (43) | 1 | 0 | 1 | 1 | 1 | 1 | 1 | 7/10 |
| Laing, E. et al. | (44) | 0 | 1 | 0 | 2 | 1 | 1 | 1 | 6/10 |
| Locker, D. et al. | (45) | 0 | 0 | 0 | 1 | 0 | 1 | 1 | 3/10 |
| Manijth, C.M. et al. | (46) | 0 | 0 | 0 | 2 | 1 | 1 | 0 | 4/10 |
| Marques, L.S. et al. | (47) | 1 | 1 | 1 | 2 | 0 | 1 | 1 | 7/10 |
| Mbawalla, H.S. et al. | (48) | 0 | 1 | 0 | 1 | 1 | 1 | 1 | 5/10 |
| Montiel-Company, J.M. et al. | (22) | 1 | 0 | 0 | 2 | 1 | 1 | 1 | 6/10 |
| Onyeaso, et al. | (49) | 1 | 1 | 0 | 2 | 1 | 1 | 1 | 7/10 |
| Paula, J.S. et al. | (50) | 1 | 1 | 0 | 2 | 1 | 1 | 1 | 7/10 |
| Paula Jr, et al. | (51) | 1 | 0 | 0 | 2 | 1 | 1 | 1 | 6/10 |
| Peres, K.G. et al. | (52) | 1 | 0 | 1 | 2 | 1 | 1 | 1 | 7/10 |

Table S2b. Quality assessment of studies included in the meta-analysisbased on the Newcastle-Ottawa Scale adapted for cross-sectional studies

| | | Selection: | | | | Comparability | Outcome | | Score |
|---|---|---|---|---|---|---|---|---|---|
| *Artikel* | *Ref* | *Sample representativeness* | *SS* | *Non respondents* | *Exposure assessment* | *Control for differences* | *Outcome assessment* | *Statistics* | |
| Sardenberg F. et al. | (53) | 1 | 1 | 0 | 2 | 1 | 1 | 1 | 7/10 |
| Scapini, A. et al. | (54) | 1 | 1 | 1 | 2 | 1 | 1 | 1 | 8/10 |
| Scarpelli, A.C. et al. | (55) | 1 | 1 | 0 | 1 | 1 | 1 | 1 | 6/10 |
| Schuch, H.S. et al. | (56) | 1 | 1 | 0 | 2 | 0 | 1 | 1 | 6/10 |
| Ukra, A. et al. | (57) | 1 | 0 | 0 | 2 | 1 | 1 | 1 | 6/10 |
| Zhang, M. et al. | (21) | 1 | 0 | 1 | 2 | 1 | 0 | 1 | 6/10 |

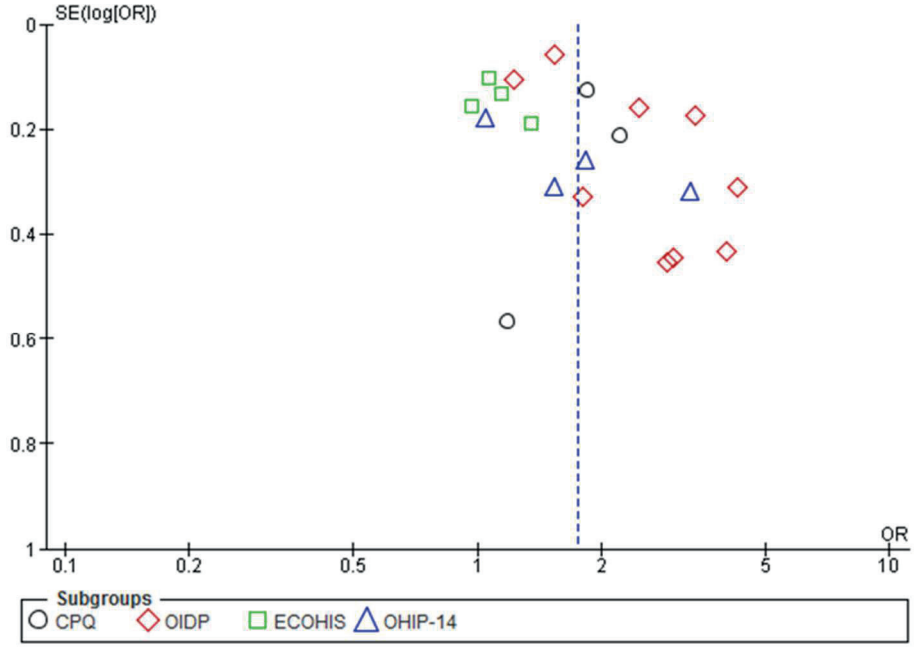


**Figure S1.** Funnel plot for the evaluation of publication bias for studies evaluating the association of malocclusion and OHRQoL categorically

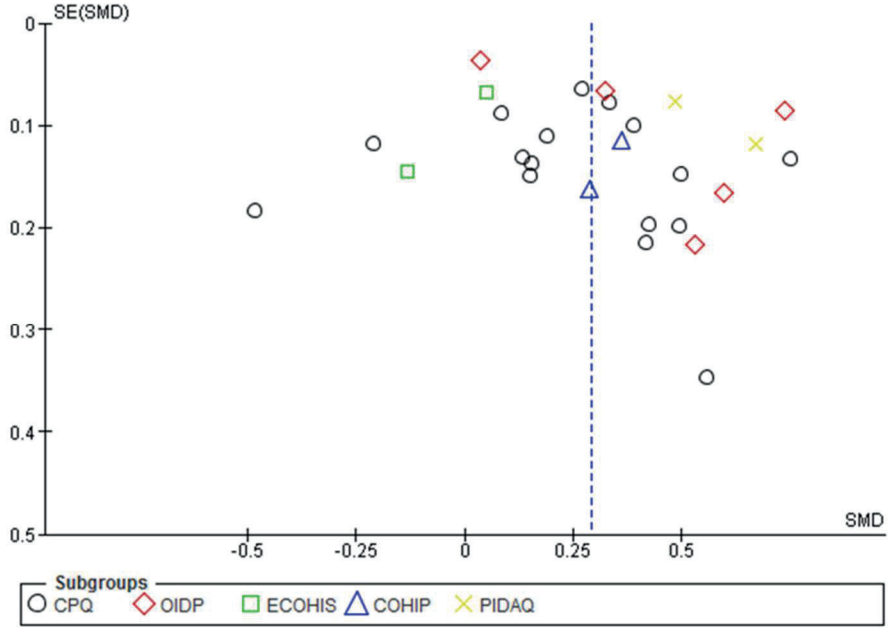


**Figure S2.** Funnel plot for the evaluation of publication bias for studies evaluating the association of malocclusion and OHRQoL dichotomously

**Table S3.** Sensitivity analysis for the different meta-analyses about OHRQoL and malocclusions

| | OHRQoL | Study Omitted | Ref | SMD / OR (95% CI) | P-value for difference |
|---|---|---|---|---|---|
| continuous | CPQ[1] | Aguilar-Diaz (2011) (1) | (24) | 0.30 (0.19, 0.40) | 0.892 |
| | | Barbosa (2009) | (27) | 0.29 (0.18, 0.39) | 1.000 |
| | | Barbosa (2009b) | (27) | 0.29 (0.18, 0.39) | 1.000 |
| | | Bhayat (2014) | (28) | 0.30 (0.19, 0.40) | 0.892 |
| | | Bekes (2012) | (29) | 0.29 (0.18, 0.40) | 1.000 |
| | | Brown (2006) | (34) | 0.28 (0.18, 0.39) | 0.892 |
| | | Dawoodbhoy (2013) | (36) | 0.27 (0.17, 0.37) | 0.782 |
| | | Foster Page (2005) | (37) | 0.29 (0.18, 0.39) | 1.000 |
| | | Foster Page (2013) | (38) | 0.30 (0.19, 0.40) | 0.892 |
| | | Kolawole (2011) | (42) | 0.31 (0.21, 0.41) | 0.782 |
| | | Laing (2010) | (44) | 0.31 (0.21, 0.41) | 0.782 |
| | | Locker (2007) | (45) | 0.29 (0.18, 0.39) | 1.000 |
| | | Scapini (2013) | (54) | 0.30 (0.19, 0.40) | 0.892 |
| | | Schuch (2014) | (56) | 0.30 (0.19, 0.41) | 0.895 |
| | | Ukra (2013) | (57) | 0.29 (0.18, 0.40) | 1.000 |
| | | Zhang (2009) | (21) | 0.28 (0.18, 0.39) | 0.892 |
| | OIDP[2] | Bernabe (2008) | (30) | 0.28 (0.18, 0.39) | 0.892 |
| | | Bernabe (2009) | (33) | 0.29 (0.18, 0.40) | 1.000 |
| | | Bernabe (2009) (1) | (32) | 0.27 (0.17, 0.37) | 0.782 |
| | | Hvaring (2014) | (41) | 0.28 (0.18, 0.38) | 0.890 |
| | | Mbawalla (2011) | (48) | 0.30 (0.20, 0.41) | 0.892 |
| | ECOHIS[3] | Abanto (2011) | (23) | 0.31 (0.20, 0.41) | 0.787 |
| | | Kramer (2013) | (43) | 0.30 (0.20, 0.41) | 0.892 |
| | COHIP[4] | Asgari (2013) | (26) | 0.29 (0.18, 0.39) | 1.000 |
| | | Kragt (2015) | (20) | 0.29 (0.19, 0.40) | 1.000 |
| | PIDAQ[5] | Montiel-Company (2009) | (22) | 0.28 (0.18, 0.39) | 0.892 |
| | | Paula jr (2011) | (51) | 0.27 (0.17, 0.38) | 0.787 |
| dichotomous | CPQ | Ghijselings (2014) | (19) | 1.76 (1.47, 2.10) | 0.929 |
| | | Paula (2012) | (50) | 1.72 (1.43, 2.06) | 0.929 |
| | | Sardenberg (2013) | (53) | 1.74 (1.44, 2.10) | 1.000 |
| | OIDP | Bernabe (2008) | (30) | 1.72 (1.44, 2.06) | 0.929 |
| | | Bernabe (2008) (1) | (31) | 1.74 (1.45, 2.09) | 1.000 |
| | | Bernabe (2009) | (33) | 1.70 (1.42, 2.03) | 0.857 |
| | | Bernabe (2009) | (33) | 1.65 (1.40, 1.95) | 0.859 |
| | | Herkrath (2012) | (40) | 1.72 (1.44, 2.05) | 0.856 |
| | | Hvaring (2014) | (41) | 1.70 (1.42, 2.03) | 0.857 |
| | | Marques (2006) | (47) | 1.67 (1.41, 1.98) | 0.852 |
| | | Mbwalla (2011) | (48) | 1.80 (1.49, 2.17) | 0.798 |
| | | Peres (2013) | (52) | 1.79 (1.45, 2.21) | 0.810 |
| | ECOHIS | Carvalho (2013) | (35) | 1.80 (1.49, 2.16) | 0.797 |
| | | Gomes (2014) | (39) | 1.81 (1.51, 2.16) | 0.760 |
| | | Kramer (2013) | (43) | 1.77 (1.47, 2.14) | 0.897 |
| | | Scarpelli (2013) | (55) | 1.81 (1.51, 2.17) | 0.762 |
| | OHIP-14 | Anosike (2010) | (25) | 1.87 (1.56, 2.26) | 0.614 |
| | | Feu (2010) | (18) | 1.76 (1.46, 2.11) | 0.930 |
| | | Manjith (2012) | (46) | 1.82 (1.51, 2.20) | 0.735 |
| | | Onyeaso (2009) | (49) | 1.81 (1.50, 2.18) | 0.766 |

## 3. Supplemental References


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# Chapter 3.2

## The association of subjective orthodontic treatment need with oral health-related quality of life

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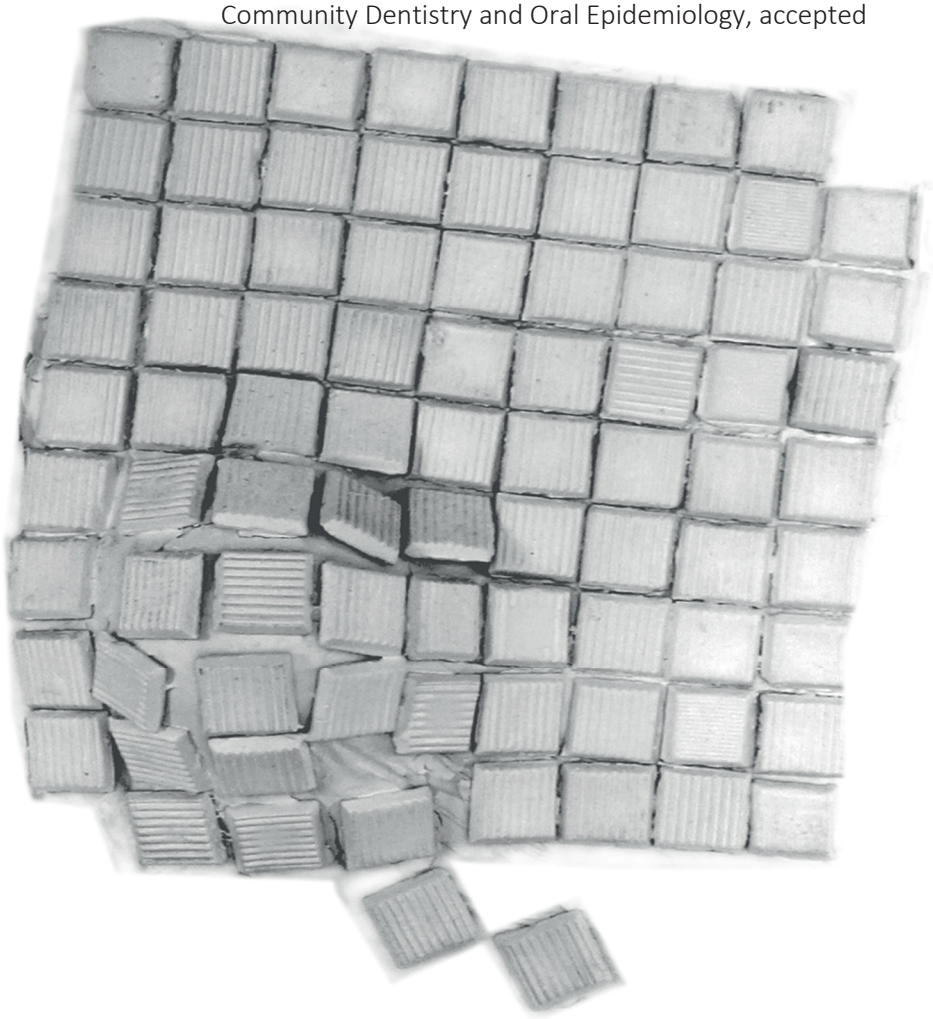

## Abstract

***Objectives*** The existing body of evidence reports an inconsistent association between subjective and objective orthodontic treatment need. The concept of oral health-related quality of life (OHRQoL) might help to explain the differences in subjective and objective orthodontic treatment need. Our aim was to investigate the association of subjective orthodontic treatment with OHRQoL in children.

***Methods*** This cross-sectional study was embedded in the Generation R Study, a population-based prospective cohort study. OHRQoL and subjective orthodontic treatment need were assessed by parental questionnaires. Questionnaire items were individually compared among children with no, borderline and definite subjective orthodontic need. The association between subjective orthodontic treatment need and OHRQoL was investigated in multivariate regression analysis with weighted least squares. Differences by sex and levels of objective orthodontic treatment need were evaluated.

***Results*** In total, 3774 children were included in the analysis. Children with borderline subjective orthodontic treatment need and those with definite subjective orthodontic treatment need had significantly poorer OHRQoL based on the fully adjusted model (adjusted regression coefficient [aβ]= -0.49, 95%CI: -0.75, -0.30; [aβ]= -1.58, 95%CI: -1.81, -1.58, respectively). The association between subjective orthodontic treatment need and OHRQoL was stronger in girls than in boys and stronger in children with objective orthodontic treatment need than in those with none.

***Conclusions*** OHRQoL is poorer in children with subjective orthodontic treatment need. This has not been investigated before in such a large-population based study and clearly offers an explanation for the lack of concurrence between objective and subjective orthodontic treatment need.

## Introduction

In 2013, a Dutch oral health report stated that 60% of young adults have had orthodontic treatment (1). Reasons for providing orthodontic treatment are based on prevention of oral diseases and improvement of aesthetics (2). The need for orthodontic treatment comes either subjectively from the patient or objectively from the care provider. The existing body of evidence shows a highly inconsistent association between subjective and objective orthodontic treatment need (3).

The concept of oral health-related quality of life (OHRQoL) was introduced in the orthodontic literature to help understand differences in subjective and objective orthodontic treatment need (4, 5). Quality of life measures assess the impact of health on social, emotional and functional aspects of life (6). OHRQoL measures the particular impact of oral conditions in terms of oral symptoms, functional limitations, emotional and social wellbeing on daily life (7). Thus, OHRQoL measures aim to capture subjective oral health in a more standardized way, so that they can augment traditional measures of oral health (8). Naturally, various oral disorders influence OHRQoL. Whereas many studies have focused on the association between objective orthodontic treatment need and OHRQoL, the association between OHRQoL and subjective orthodontic treatment need has rarely been investigated. However, this is of particular importance as treatment decisions are often for a big part influenced by what patients and their parents want.

In the literature, objective orthodontic treatment need is assessed using clinical oral health features, such as with the dental health component (DHC) of the index of orthodontic treatment need (IOTN), or based on aesthetic impairments, such as with the IOTN aesthetic component (AC). Studies on the association between objective orthodontic treatment need and OHRQoL have shown weak and inconclusive associations between objective orthodontic treatment need and OHRQoL (5, 9-12). Subjective orthodontic treatment need has been inconsistently assessed in a small number of existing studies. Some studies used OHRQoL as a surrogate for subjective orthodontic treatment need (13, 14). However, OHRQoL can be distinguished from subjective need, since OHRQoL is a dynamic concept that results from the interaction between health, social and contextual factors (7). Also, different studies have equated aesthetic impairment and subjective orthodontic treatment need (13-15). However, there is little evidence for this assumption and it might be wrong, because, for example, having a worse IOTN-AC score does not implicitly mean having more perceived treatment need. In addition, dental attractiveness, which can be one of the reason for subjective orthodontic treatment need, is not necessarily associated with OHRQoL (13, 14, 16, 17). Though never evaluated, still the children with more aesthetic impairment might show a stronger association between subjective orthodontic treatment need and OHRQoL than children with less aesthetic impairment. In summary, little is known about the association between subjective orthodontic treatment need and OHRQoL.

Accordingly, the aim of this study was to quantify the association between subjective orthodontic treatment need, not assessed by an objective index but a simple question, and OHRQoL. In particular, we were interested in whether subjective orthodontic treatment need in children is associated with poorer OHRQoL independent of their objective orthodontic treatment need. The secondary aim of this study was to see

whether the association between subjective orthodontic treatment-need and OHRQoL varied by sex or different degrees of objective orthodontic treatment need.

## Methods

This cross-sectional study was embedded in the Generation R Study, a population-based prospective cohort study that previously has been described in detail (18). The study protocol and its conduct were in accordance with the guidelines of the Declaration of Helsinki and approved by the Medical Ethical Committee of the Erasmus MC, University Medical Centre Rotterdam (MEC-2012-165). Participating parents have given written informed consent before the data collection in children had started (n=7393). Information on children's OHRQoL was given by the parents of 3796 children (51.3%), of whom 3774 (51.0%) also provided subjective orthodontic treatment need.

Subjective orthodontic treatment need was assessed in parental questionnaires with the question: "Do you think your child needs braces?". The response to the question was given by the mothers on a five-point Likert scale from strongly disagree to strongly agree. For the analysis subjective orthodontic treatment need was categorized into: 'No subjective orthodontic treatment need' for children whose mothers strongly or somewhat disagreed with the statement. 'Borderline subjective orthodontic treatment need' for children whose mothers did not agree but also did not disagree with the statement and 'Definite subjective orthodontic treatment need' for children whose mothers somewhat or strongly agreed with the statement.

OHRQoL was measured with the COHIP-ortho (19). The COHIP-ortho is a questionnaire addressed to parents measuring OHRQoL of the child with 11 questions, covering the different domains of oral health, including social-emotional wellbeing, functional well-being and school and peer interaction (Appendix Table S1). These questions were answered on a five-point Likert scale (never, almost never, sometimes, fairly often, almost all the time). The responses scored from 1 to 5, and were finally summed for each individual. The total overall score of the COHIP-ortho ranges from 0 to 55 and higher scores correspond to higher OHRQoL. Missing values in the responses to the OHRQoL questionnaire (COHIP-ortho) were replaced by the personal mean score of the remaining answers to the questions, as proposed by researchers who used the original version of the COHIP (20). If more than 30% of the answers were missing, the participant was excluded from the analysis.

The association between children's subjective orthodontic treatment need and OHRQoL is most likely influenced by other factors, and so the following parental characteristics were considered as covariates: maternal educational level (low, high), household income (<2000€, 2000-3200€, >3200€) and ethnicity (Dutch, other Western, non-Western) as indicators for social economic status (SES); and the following children's characteristics were considered as covariates: sex, age and objective orthodontic treatment need. Objective orthodontic treatment need was assessed with the dental health component (DHC) and aesthetic component (AC) of the IOTN. The IOTN was assessed from photographic and radiographic records of the children (median (90% range) age 9.78 (9.49 - 10.45)). Assessment of the IOTN on a combination of photographic and radiographic records has been validated previously (21). After 6 months,

10% of the photographs were reassessed to calculate the intra-rater reliability (linear weighted K = 0.84).

Statistical analyses used Statistical Package for Social Sciences (IBM SPSS statistics) version 21, SPSS Inc, Chicago, IL, USA. Characteristics of the participants were summarized and stratified by sex. Differences between males and females were investigated with chi-square tests and Mann-Whitney-U-tests. Mean scores for the individual items of the COHIP-ortho in the group of unsure and definite orthodontic treatment need were separately compared with the mean scores for the individual questions of the no subjective orthodontic treatment need group. To evaluate the differences in the mean item scores between these groups, Cohen's effect sizes were calculated. Following Cohen's suggestions, effect sizes of 0.2 were considered small, 0.5 were considered medium and 0.8 were considered large (22). Differences between the groups were evaluated with the Mann-Whitney-U test ($p < 0.05$). Furthermore, weighted least square (WLS) linear regression models were calculated with subjective orthodontic treatment need as the determinant and the summary score for OHRQoL as the outcome. We used WLS regression models, because of the heteroscedasticity in the OHRQOL data. In multivariate WLS regression analysis with (potential) confounders (child's age and sex (crude model), child's ethnicity and other indicators of socio-economic status (Model 1) and finally orthodontic characteristics (Model 2) were added. The selection of covariates into the model was based on the current orthodontic literature and significant associations between covariates with both subjective orthodontic treatment need and OHRQoL. We also performed a test for trend analysis by treating the categorized variable (subjective orthodontic treatment need) as a continuous term. We tested for differences in the association of subjective orthodontic treatment need and OHRQoL between girls and boys and children with and without objective orthodontic treatment need based on either the IOTN-DHC or the IOTN-AC by including interactions terms in the model. For all variables, significant interactions were present (Table 3). Significant differences in the associations between the strata were evaluated with a test for heterogeneity. For all analyses, a p value $< 0.05$ was considered to be statistically significant.

Missing values for covariates were handled with multiple imputation by using the Markov Chain Monte Carlo method. Objective orthodontic treatment need had the largest amount of missing data (IOTN-AC (22.9 %), IOTND-DHC (20.3%), Table 4). We generated 5 independent datasets with a fully conditional specified model and we present the pooled effect estimates (β (95% Confidence intervals (CI)). Rubin's rules were applied for pooling of the effect estimates (23). We generated 5 independent datasets because the pooled effect estimates did not change with more imputations and because based on Rubin's rules the relative efficiency of 5 imputed datasets appeared sufficient, namely higher than 95.6% in case of 22.9% missing data (23). Imputations were based on the associations between all variables used in this study, but the main determinant (subjective orthodontic treatment need) and outcome (OHRQOL) were not imputed (22). Finally, we also conducted a sensitivity analysis in the original dataset. The obtained effect estimates (β(95%)) of the sensitivity analysis were comparable with the pooled effect estimates on the relation between subjective and objective orthodontic treatment need (Appendix, Table S2 ).

To evaluate potential selection bias, children with missing data on OHRQOL and subjective orthodontic treatment need (n=3619) were compared to those without missing data on OHRQOL and subjective orthodontic treatment need (n=3774). Data on OHRQOL and subjective orthodontic treatment need were more often missing in children from parents with lower socio-economic status (for all socio-economic indicators p value < 0.001, Table 4).

## Results

In Table 1, the characteristics of the study sample are presented. In total, 3774 children were included in the final analysis, of whom 1767 (46.8%) had definite subjective orthodontic treatment need, 958 (25.4%) were unsure about their orthodontic treatment need and 1049 (27.8%) did not perceive any subjective orthodontic treatment need. Boys had slightly higher OHRQoL and perceived less orthodontic treatment need than girls. These differences between boys and girls were significant (p <0.001).

**Table 1.** Characteristics of the study sample by sex (n=3774)

| *Characteristics* | | Boys n= 1873 | Girls n=1901 | p value* |
|---|---|---|---|---|
| **Age in years** | | | | |
| | Median (range) | 9.8 (9.5-10.4) | 9.9 (9.5-10.5) | 0.643 |
| **Ethnicity [a] (%)** | | | | |
| | Dutch | 1278 (68.2) | 1295 (68.1) | |
| | Other western | 147 (7.8) | 180 (9.4) | |
| | Non-western | 438 (23.4) | 413 (21.7) | 0.135 |
| **Maternal education level [a]** | | | | |
| | Low | 619 (33.0) | 615 (32.3) | |
| | High | 1143 (61.0) | 1158 (60.9) | 0.782 |
| **Household income[a] (%)** | | | | |
| | < 2000 | 321 (17.1) | 298 (15.7) | |
| | 2000-3200 | 552 (29.5) | 535 (28.1) | |
| | > 3200 | 877 (46.8) | 936 (49.2) | 0.230 |
| **IOTN-DHC[a] (%)** | | | | |
| | 2 | 578 (30.6) | 541 (28.5) | |
| | 3 | 364 (19.4) | 392 (20.6) | |
| | 4 | 428 (22.9) | 443 (23.3) | |
| | 5 | 128 (6.8) | 135 (7.1) | 0.449 |
| **IOTN-ac[a] (%)** | | | | |
| | ≤ 5 | 1045 (55.8) | 1018 (53.5) | |
| | > 5 | 394 (21.0) | 451 (23.7) | 0.049 |
| ***OHRQoL*** | | | | |
| | Median (range) | 50.0 (43.00-53.00) | 49.0 (42.00-53.00) | < 0.001 |
| **Subjective treatment need (%)** | | | | |
| | No | 565 (30.2) | 484 (25.5) | |
| | Borderline | 498 (26.6) | 460 (24.2) | |
| | Yes | 810 (43.2) | 957 (50.3) | <0.001 |

*Based on chi square test for categorical variables and t-test or Mann –Whitney U for continuous variables

[a] May not add up to 3774, because of missing values: Maternal education: 6.3 %; Ethnicity : 0.6%; IOTN: 21.9 %; Household income: 6.7%, IOTN-DHC: 20.2%; IOTN-AC: 22.9%

Table 2 shows the mean COHIP-ortho item scores of the children with no perceived orthodontic treatment need, borderline perceived orthodontic treatment need and definite perceived orthodontic treatment need. Children with borderline perceived orthodontic treatment need had lower scores than children with no perceived need for the items about 'crooked teeth', 'discolored teeth' and 'bleeding gums'. Children with definite orthodontic treatment need showed lower scores than children without per-

ceived orthodontic treatment need on all items except 'pain', 'bad breath' and 'attractiveness'. Most of the effect sizes were small except for the item 'crooked teeth' in the borderline perceived and definite orthodontic treatment need groups (d=0.36, p ≤0.001; d=0.98, p ≤0.001) as well as the item 'anxious' in the definite perceived orthodontic treatment need group (d=0.34, p ≤0.001).

**Table 2.** COHIP-ortho scores by question for children with unsure or definite subjective orthodontic treatment need versus no subjective orthodontic treatment need (n=3774)

| | COHIP-ortho mean scores per question (mean ± standard deviation) | | | | |
|---|---|---|---|---|---|
| Questions | No subj need | Unsure subj need | Effect size[a] | Definite subj need | Effect size [a] |
| Pain | 4.8 (0.5) | 4.8 (0.5) | 0.02 | 4.8 (0.5) | 0.02 |
| Crooked teeth | 4.8 (0.6) | 4.5 (0.8) | 0.36** | 3.8 (1.2) | 0.98** |
| Discolored teeth | 4.7 (0.7) | 4.6 (0.8) | 0.13** | 4.5 (0.9) | 0.22** |
| Bad breath | 4.5 (0.8) | 4.5 (0.8) | 0.05 | 4.8 (0.9) | 0.07 |
| Bleeding gums | 4.7 (0.6) | 4.7 (0.6) | 0.08* | 4.6 (0.8) | 0.22** |
| Eating foods | 4.9 (0.4) | 4.9 (0.4) | 0.00 | 4.8 (0.5) | 0.15* |
| Anxious | 4.8 (0.6) | 4.7 (0.6) | 0.05 | 4.5 (0.9) | 0.34** |
| Speaking | 5.0 (0.1) | 5.0 (0.2) | 0.00 | 5.0 (0.3) | 0.14** |
| Bullied | 5.0 (0.3) | 5.0 (0.2) | 0.04 | 4.9 (0.4) | 0.15** |
| Attractiveness | 1.8 (1.2) | 1.8 (1.2) | 0.02 | 1.8 (1.1) | 0.00 |
| Pronunciation | 5.0 (0.3) | 5.0 (0.3) | 0.04 | 4.9 (0.5) | 0.16** |

[a] Cohens effect size (d) for differences between either No subjective need and borderline subjective need or No subjective need and definite subjective need. p values are based on Mann Whitney U test for differences in mean scores * ≤ 0.05, **≤ 0.001

In Table 3 the findings of the regression model for subjective orthodontic treatment need and total COHIP scores are shown. In contrast to children without subjective orthodontic treatment need, children with borderline orthodontic treatment need as well as children with definite subjective orthodontic treatment need had significant lower total COHIP scores after adjustments for SES and objective orthodontic treatment need (adjusted regression coefficient [aβ]= -0.49, 95%CI:-0.75, -0.30; [aβ]= -1.58, 95%CI:-1.81, -1.58, respectively). The trend estimates for the association between subjective orthodontic treatment need and total COHIP scores were significant (p <0.001).In the group without subjective orthodontic treatment need, girls had generally lower total COHIP scores than boys (Appendix Table S4/S5). In addition, the effect of definite subjective orthodontic treatment need on OHRQoL was significantly stronger in girls than in boys ([aβ]= -1.93, 95%CI: -2.27, -1.60 and [aβ]= -1.27, 95%CI: -1.58, -0.96, respectively, p <0.001).The associations between subjective orthodontic treatment need and OHRQoL stratified by objective orthodontic treatment need are also presented in Table 3. After stratification by objective orthodontic treatment need based on the IOTN-AC, the association between subjective treatment need and total COHIP scores was stronger in children with an IOTN-AC >5 for the borderline and the definite subjective need group than in children with an IOTN-AC ≤5 (p value =0.024). Similarly, after stratification by objective orthodontic treatment need based on the IOTN-DHC, the association between definite subjective treatment need and total COHIP scores was stronger in children with an IOTN-DHC >3 than in children with an IOTN-DHC ≤3 (p =0.039). In contrast, the association between borderline perceived subjective treatment need and total COHIP score was significantly stronger in children with an IOTN-DHC ≤3 ([aβ]= -0.57, 95%CI:-0.85, -0.30) than in children with an IOTN-DHC >3 ([aβ]= -0.42, 95%CI:0.02, -0.85).

**Table 3.** The association between subjective orthodontic treatment need and OHRQoL analyzed overall, stratified by gender and both IOTN components

| | Oral health related quality of life score | | | | | | |
|---|---|---|---|---|---|---|---|
| | **Total** | **Sex** | | **IOTN-DHC** | | **IOTN-AC** | |
| | | **Boys** | **Girls** | **DHC ≤ 3** | **DHC > 3** | **AC ≤ 5** | **AC > 5** |
| | (n= 3774) | (n= 1873) | (n= 1901) | (n=2129) | (n=1645) | (n=2275) | (n=1499) |
| No need | Ref | Ref | Ref | Ref | Ref | Ref | Ref |
| **Borderline need** (β (95% CI)) | | | | | | | |
| Crude [a] | -0.51 (-0.74, - 0.27) | -0.59 (-0.90, - 0.27) | -0.42 (-0.77, - 0.07) | -0.60 (-0.89, 0.31) | -0.30 (-0.77, 0.17) | -0.49 (-0.77, - 0.21) | -0.55 (-1.03, - 0.06) |
| Adjusted [b] | -0.53 (-0.75, - 0.30) | -0.57 (-0.86, - 0.27) | -0.51 (-0.85, - 0.17) | -0.58 (-0.85, 0.31) | -0.42 (-0.87, 0.03) | -0.47 (-0.74, - 0.21) | -0.64 (-1.12, - 0.17) |
| Adjusted [c] | -0.49 (-0.71, - 0.27) | -0.52 (-0.82, - 0.23) | -0.49 (-0.82, - 0.15) | -0.57 (-0.85, - 0.30) | -0.42 (-0.85, 0.02) | -0.45 (-0.72, - 0.19) | -0.64 (-1.11, - 0.17) |
| **Definite need** (β (95% CI)) | | | | | | | |
| Crude [a] | -1.76 (-1.99, - 1.53) | -1.43 (-1.74, - 1.12) | -2.13 (-2.46, - 1.80) | -1.45 (-0.89, - 0.31) | -1.78 (-2.21, - 1.36) | -1.50 (-1.80, - 1.19) | -1.88 (-2.31, - 1.45) |
| Adjusted [b] | -1.76 (-1.98, 1.55) | -1.41 (-1.71, - 1.12) | -2.18 (-2.50, - 1.86) | -1.46 (-1.75, - 1.17) | -1.86 (-2.25, - 1.47) | -1.50 (-1.79, - 1.21) | -1.93 (-2.33, - 1.53) |
| Adjusted [c] | -1.58 (-1.81, - 1.34) | -1.27 (-1.58, - 0.96) | -1.93 (-2.27, - 1.60) | -1.45 (-1.73, - 1.16) | -1.80 (-2.19, - 1.41) | -1.44 (-1.74, - 1.13) | -1.81 (-2.24, - 1.38) |
| ***p for trend*** [d] | <0.001 | <0.001 | <0.001 | <0.001 | <0.001 | <0.001 | <0.001 |
| ***p for interaction*** [e] | | 0.001 | | 0.039 | | 0.024 | |

[a] adjusted only for age and additional for sex in the overall analysis, [a]adjusted for age, ethnicity, household income, maternal education and additional for sex in the overall analysis, [c] adjusted for age, ethnicity, household income, maternal education, additional for sex in the overall analysis as well as the stratification on IOTN-DHC and IOTN-AC; and additional for IOTN-DHC and IOTN-AC in the overall analysis as well as the stratification on sex, [d] p for trend for the fully adjusted model obtained by treating subjective orthodontic treatment need as continuous term, [e] obtained from interaction-term entered into the crude model between subjective orthodontic treatment need and gender, resp. IOTN-DHC and IOTN-AC, ref = reference category

## Discussion

Our study findings suggest that subjective orthodontic treatment need is associated with poor OHRQoL. We showed that more subjective orthodontic treatment need is associated with poorer OHRQoL in children with and without objective orthodontic treatment need and that this association is stronger in girls than in boys. Considering these marked associations, .subjective orthodontic treatment need is not solely related to objective orthodontic treatment need, but also related to OHRQoL. And thus, OHRQoL offers an explanation for the lack of concurrence between objective and subjective orthodontic treatment need.

The main strength of the present study is the large and ethnically diverse study sample obtained from a population-based cohort study, which was designed to be representative for the general population in the Netherlands. However, the study findings should also be seen in the light of several limitations. Non-response analysis showed a higher proportion of children without information on OHRQoL or subjective orthodontic treatment need had parents of lower socio-economic status. This might have caused selection bias if the association between subjective orthodontic treatment need and OHRQoL would be different in included and excluded participants.

**Table 4.** Non response analysis (n = 7393)

| *Characteristics* | Included n= 3774 | Excluded n=3619 | p value* |
|---|---|---|---|
| **Sex** | | | |
| Boys (%[1]) | 1873 (49.6) | 1834 (50.7) | |
| Girls (%[1]) | 1901 (50.4) | 1785 (49.3) | 0.374 |
| *Missing (%[2])* | *0 (0.0)* | *0(0.0)* | |
| **Age in years** | | | |
| Median (range) | 9.78 (9.49 - 10.45) | 9.86 (9.56 - 11.12) | < 0.001 |
| *Missing (%[2])* | *0 (0.0)* | *3252(89.9)* | |
| **Ethnicity (%)** | | | |
| Dutch (%[1]) | 2573 (68.6) | 1619 (47.7) | |
| Other western (%[1]) | 327 (8.7) | 247 (7.3) | |
| Non-western (%[1]) | 851 (22.7) | 1529 (45.0) | < 0.001 |
| *Missing (%[2])* | *23 (0.6)* | *379(10.5)* | |
| **Maternal education level** | | | |
| Low (%[1]) | 1234 (34.9) | 1191 (52.1) | |
| High (%[1]) | 2301 (65.1) | 1093 (47.9) | < 0.001 |
| *Missing (%[2])* | *239 (6.3)* | *1490(41.2)* | |
| **Household income (%)** | | | |
| < 2000 (%[1]) | 619 (17.6) | 358 (31.8) | |
| 2000-3200 (%[1]) | 1087 (30.9) | 353 (31.3) | |
| > 3200 (%[1]) | 1813 (51.5) | 415 (36.9) | < 0.001 |
| *Missing (%[2])* | *255(6.8)* | *2493(68.9)* | |
| **IOTN-DHC (%)** | | | |
| 2 (%[1]) | 1119 (37.2) | 632 (37.7) | |
| 3 (%[1]) | 756 (25.1) | 380 (22.7) | |
| 4 (%[1]) | 871 (28.9) | 496 (29.6) | |
| 5 (%[1]) | 263 (8.7) | 168 (10.0) | 0.185 |
| *Missing (%[2])* | *765(20.3)* | *1943(53.7)* | |
| **IOTN-ac (%)** | | | |
| ≤ 5 (%[1]) | 2063 (70.9) | 1144 (70.8) | |
| > 5 (%[1]) | 845 (29.1) | 471 (29.2) | 0.940 |
| *Missing (%[2])* | *866(22.9)* | *2034(56.2)* | |
| ***OHRQoL*** | | | |
| Median (range) | 50.0 (43.0 - 53.0) | 49.0 (44.3 - 54.9) | |
| *Missing (%[2])* | *0(0.0)* | *3597(99.4)* | *0.959* |
| **Subjective treatment-need (%)** | | | |
| No (%[1]) | 1049 (27.8) | 1 (11.1) | |
| Borderline (%[1]) | 958 (25.4) | 2 (22.2) | |
| Yes (%[1]) | 1767 (46.8) | 6 (66.7) | 0.429 |
| *Missing (%[2])* | *0(0.0)* | *3610* | |

*Based on chi square test for categorical variables and t-test or Mann –Whitney U for continuous variables. [1]percentage of available data within the subgroup; [2] percentage of missing data per subgroup

However, because we have no information on subjective orthodontic treatment need and OHRQoL in the non-responding subsample, this is difficult to ascertain. Another drawback of our study is that in this study OHRQoL and subjective orthodontic treatment need of the children was assessed by asking the parents, thus we assumed that parents are a valid proxy for children's reports. This assumption was based on several studies that found parents to be good proxies for children's OHRQoL (24-26). Still, we cannot exclude an information bias including a social desirability bias. In addition, we also had no information whether children already had started their orthodontic treatment or not which also might have contributed to an information bias in the main determinant. In the Netherlands, parents and dentist start to concern with orthodontic treatment need around the children's age of nine, but it is rather uncommon that children start their orthodontic treatment so early. However, if they have started they were definitely still in orthodontic treatment need, which we assessed and included in the

analysis. Furthermore, a limitation of our study is that the IOTN was assessed from radiographic and photographic records due to logistic reasons in such a large cohort study as the Generation R study. This method is less valid than direct oral examination and might also have introduced some misclassification of participants' orthodontic treatment need. However, this method has been shown to be sufficiently valid for research (21). Objective orthodontic treatment need was assessed with the IOTN. This measure was chosen because it was developed solely based on the opinion of orthodontists (27). In this way, the analysis would be adjusted only for professional based objective orthodontic treatment need. The use of other orthodontic measures such as the Dental Aesthetic Index (DAI), might have been problematic, because this Index not only covers objective orthodontic treatment need, but also social norms. Subjective orthodontic treatment need as well as OHRQoL are both influenced by social norms. Consequently, the use of the DAI to adjust the analysis might have resulted in a weaker association between subjective orthodontic treatment need and OHRQoL. In line with this, a recent meta-analysis showed that the association between objective orthodontic treatment need assessed with the DAI and OHRQoL is highly heterogeneous, whereas this association assessed with the IOTN is not (28). Finally, like in every observational study, our study findings might be affected by residual confounding, although we were able to minimize confounding of the study findings by constructing fully adjusted models including indicators for socio-economic status and objective orthodontic treatment need.In agreement with Kok et al. (2004), we think that aesthetics are limited in their ability to reflect subjective need for orthodontic care (14). For example, subjective orthodontic treatment need can arise when friends wear braces or when the opinion is influenced by the recommendation of the dentist. Furthermore, children with a similar dental aesthetic impairment do not necessarily perceive the same subjective orthodontic treatment need. Nevertheless, based on the stratification analysis, the association between subjective orthodontic treatment need and OHRQoL seemed indeed stronger in children with more dental aesthetic impairment. Next to this, our analysis showed that children with an IOTN-DHC ≤3 do perceive more impact of borderline subjective orthodontic treatment need on OHRQoL than children with an IOTN-DHC >3. Most likely, these are the children who are more aware of their dentition and feel more impairment due to minor malocclusions. In this way, they could be a source for the divergent association between subjective and objective orthodontic treatment need reported by other authors (3). Whether the perceived impairment due to minor malocclusions is related to conditions like Body Dysmorphic Disorder (as suggested by several researchers) might be possible, but is probably not the case, given that it is a rare condition (12, 29).

The sex differences shown in the present study reflect the dynamic, context-specific character of OHRQoL. Based on the literature we expected general poorer OHRQoL in girls (13,16). Surprisingly, the association between definite subjective orthodontic treatment need and OHRQoL was stronger in girls, whereas the association between borderline subjective orthodontic treatment need and OHRQoL was stronger in boys. In line with another study, this suggests that females might be more conscious about their appearance, but boys might be more aware of their malocclusions (16,30). At the age of 9, girls might already be more aware of themselves and how they come across, com-

pare themselves more with their friends and feel more pressure to be like their peers, for example by wanting braces. We saw in the sex specific item analysis that the effect of subjective treatment need on items about bullying and pronunciation was stronger in girls than in boys (Appendix, Table S3/S4), and those items belong to the peer interaction domain of OHRQoL (31). Differences between girls and boys regarding the experience of oral health and its impacts on OHRQoL have been reported in 12 year old children (26). Another study performed in adults showed that women perceive both the negative and positive impacts of oral health on OHRQoL more intensely, and a recently published study found that OHRQoL was worse in girls than in boys after a 3-year follow-up, whether they were orthodontically treated or not (32,33). Thus, although sex differences in oral health research are insufficiently investigated yet, it is generally accepted that girls and boys differ in psychological variables as how they perceive themselves (34). Still, these different studies suggests that the association between subjective orthodontic treatment need with OHRQoL should be investigated at different ages and over time, also with regard to the differences between boys and girls, before valid conclusion can be drawn.

Our study is of clinical relevance in orthodontics, oral epidemiology and community dentistry. The findings contribute to understanding the importance of orthodontic treatment for the young population in terms of quality of life. Our findings give an indication for why boys might be less compliant with treatment, to be specific because they have generally higher OHRQoL. In this way, our findings can support an effective communication between patient and orthodontist. Furthermore, the regression analysis, in combination with the item analysis, showed that subjective orthodontic treatment need is associated with poorer OHRQoL, first independent of objective orthodontic treatment need, and second especially affecting OHRQOL on the social-emotional and functional domain. Thus, whereas the provision of orthodontic treatment nowadays is largely based on oral health factors such as susceptibility to caries or dental trauma or functional problems such as temporomandibular disorders or difficulties with chewing, subjective problems such as avoiding to smile or to speak lie in the social-emotional domain and are those which were in the present study particularly associated with poorer OHRQoL and subjective orthodontic treatment need. Therefore, our findings are also relevant for health education and policy decisions, especially in representing the patient's perspective. Finally, this study helps understanding the importance of OHRQoL as outcome measure in the orthodontic practice as well as Health Service Research.

In summary, we conclude that OHRQoL is poorer in children with subjective orthodontic treatment need. This has not been investigated before in such a large-population based study and clearly offers an explanation for the variability between objective and subjective orthodontic treatment need. Further research should not only focus on the association between subjective orthodontic treatment need and OHRQOL in populations of different ages, but also investigate in more detail the role of personal and environmental factors other than sex, such as socio-economic status, on the association between OHRQoL, subjective orthodontic treatment need and malocclusions.

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## Supplemental Material

**Table S1.** The 11 items of the Cohip-ortho, used in the present study

During the past 3 month, how often has your child:

1. had pain in his/her teeth/toothache
2. had crooked teeth or spaces between his/her teeth
3. had discolored teeth or spots on his/her teeth
4. had bad breath
5. had bleeding gums
6. had difficulty eating foods he/she would like to because of his/her teeth, mouth, or face
7. felt worried or anxious because of his/her teeth mouth or face
8. not wanted to speak/ read out loud in class because of his/her teeth, mouth, or face
9. been teased, bullied or called names by other children because of his/ her teeth, mouth, or face
10. felt that he/she was attractive (good looking) because of his/ her teeth, mouth, or face
11. had difficulty saying certain words because of his/her teeth or mouth

**Table S2.** Sensitivity analysis for the association between self-perceived orthodontic treatment-need and OHRQoL given in mean scores and change in mean score based on original data

| | **Oral health related quality of life score (95% CI)** | | |
|---|---|---|---|
| | **Total** (n= 3774) | **Boys** (n= 1873) | **Girls** (n= 1901) |
| **No need** (Mean (95% CI)) | Ref | Ref | Ref |
| **Borderline need** (β (95% CI)) | | | |
| Crude [a] | -0.51 | -0.58 | -0.42 |
| | (-0.74 - -0.27) | (-0.90 - -0.27) | (-0.77 - -0.07) |
| Adjusted [b] | -0.53 | -0.59 | -0.50 |
| | (-0.75 - -0.31) | (-0.90 - -0.28) | (-0.85 - -0.14) |
| Adjusted [c] | -0.49 | -0.55 | -0.49 |
| | (-0.71 - -0.27) | (-0.91 - -0.19) | (-0.82 - -0.15) |
| **Definite need** (β (95% CI)) | | | |
| Crude [a] | -1.76 | -1.43 | -2.13 |
| | (-1.99 - 1.53) | (-1.74 - -1.12) | (-2.46 - -1.80) |
| Adjusted [b] | -1.78 | -1.45 | -2.14 |
| | (-1.99 - -1.56) | (-1.77 - -1.14) | (-2.47 - -1.80) |
| Adjusted [c] | -1.59 | -1.31 | -1.93 |
| | (-1.83 - -1.36) | (-1.70 - -0.92) | (-2.27 - -1.60) |
| ***p for trend*** | > 0.001 | > 0.001 | > 0.001 |

[a] adjusted only for age and additional for gender in the overall analysis, [a]adjusted for age, ethnicity, household income, maternal education and additional for gender in the overall analysis, [c] adjusted for age, ethnicity, household income, maternal education, IOTN-DHC, IOTN-AC and additional for gender in the overall analysis, *≤ 0.05, **≤ 0.001 significant changes;

**Table S3.** COHIP-ortho scores by question for boys with unsure or definite subjective orthodontic treatment-need compared to no subjective orthodontic treatment-need (n=1873)

| Questions | COHIP-ortho mean scores per question (mean ± standard deviation) | | | | |
|---|---|---|---|---|---|
| | No subj need n = 565 | Doubtful subj need n = 498 | Effect size[a] | Definite subj need n = 810 | Effect size[a] |
| Pain | 4.81 (0.52) | 4.80 (0.53) | 0.02 | 4.81 (0.52) | 0.00 |
| Crooked teeth | 4.83 (0.52) | 4.60 (0.78) | 0.35** | 4.05 (1.15) | 0.87** |
| Discolored teeth | 4.68 (0.76) | 4.64 (4.73) | 0.10 | 4.59 (0.81) | 0.11* |
| Bad breath | 4.57 (0.80) | 4.49 (0.84) | 0.10 | 4.50 (0.85) | 0.08 |
| Bleeding gums | 4.74 (0.61) | 4.67 (0.64) | 0.11* | 4.59 (0.77) | 0.22** |
| Eating foods | 4.89 (0.40) | 4.87 (0.42) | 0.05 | 4.82 (0.52) | 0.15* |
| Anxious | 4.76 (0.64) | 4.73 (0.62) | 0.05 | 4.56 (0.78) | 0.28** |
| Speaking | 4.99 (0.15) | 4.96 (0.25) | 0.15* | 4.96 (0.28) | 0.13* |
| Bullied | 4.96 (0.26) | 4.96 (0.25) | 0.00 | 4.93 (0.35) | 0.10* |
| Attractiveness | 1.68 (1.11) | 1.61 (1.05) | 0.05 | 1.71 (1.08) | 0.03 |
| Pronunciation | 4.96 (0.28) | 4.97 (0.21) | 0.04 | 4.92 (0.42) | 0.11 |

[a]Cohens effect size (d) for differences between either No subjective need and doubtful subjective need or No subjective need and definite subjective need. p values are based on Mann Whitney U test for differences in mean scores * ≤ 0.05, **≤ 0.001

**Table S4.** COHIP-ortho scores by question for girls with unsure or definite subjective orthodontic treatment-need compared to no subjective orthodontic treatment-need (n=1901)

| Questions | COHIP-ortho mean scores per question (mean ± standard deviation) | | | | |
|---|---|---|---|---|---|
| | No subj need n = 484 | Doubtful subj need n = 460 | Effect size[a] | Definite subj need n = 957 | Effect size[a] |
| Pain | 4.81 (0.53) | 4.82 (0.51) | 0.02 | 4.81 (0.52) | 0.00 |
| Crooked teeth | 4.72 (0.65) | 4.45 (0.86) | 0.35** | 3.65 (1.25) | 1.07** |
| Discolored teeth | 4.68 (0.69) | 4.52 (0.82) | 0.21** | 4.43 (0.94) | 0.30** |
| Bad breath | 4.50 (0.85) | 4.50 (0.84) | 0.00 | 4.46 (0.87) | 0.05 |
| Bleeding gums | 4.70 (0.64) | 4.66 (0.64) | 0.06 | 4.56 (0.73) | 0.20** |
| Eating foods | 4.86 (0.41) | 4.88 (0.43) | 0.05 | 4.80 (0.56) | 0.12 |
| Anxious | 4.74 (0.61) | 4.70 (0.63) | 0.06 | 4.44 (0.90) | 0.39** |
| Speaking | 4.98 (0.13) | 5.00 (0.05) | 0.20* | 4.94 (0.29) | 0.18* |
| Bullied | 4.95 (0.29) | 4.98 (0.16) | 0.13 | 4.89 (0.41) | 0.17** |
| Attractiveness | 1.98 (1.25) | 1.99 (1.23) | 0.01 | 1.91 (1.12) | 0.06 |
| Pronunciation | 4.97 (0.21) | 4.97 (0.23) | 0.00 | 4.87 (0.50) | 0.26** |

*Cohens effect size (d) for differences between either No subjective need and doubtful subjective need or No subjective need and definite subjective need. p values are based on Mann Whitney U test for differences in mean scores [a] ≤ 0.05, [b]≤ 0.001

# Chapter 3.3

## Early caries predicts low oral health-related quality of life at a later age

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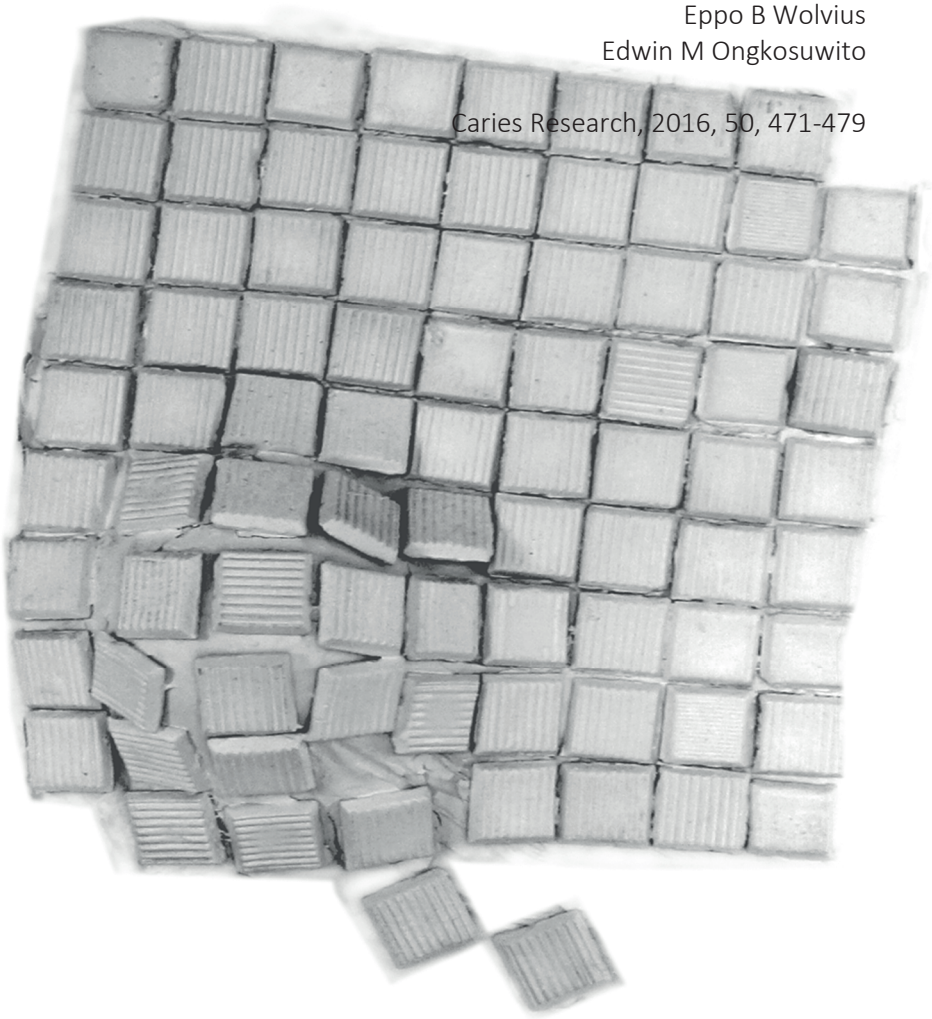

## Abstract

Oral health-related quality of life (OHRQoL) is the perceived impact of one's own oral health on daily life. Oral diseases influence children's OHRQoL directly, but OHRQoL might also be related to oral health experiences from the past. We investigate the relation between dental caries at the age of 6 with OHRQoL assessed at the age of 10. This study was conducted within the Generation R Study, a population-based prospective cohort study. Caries experience was assessed with the decayed, missing and filled teeth index (dmft) at a median (90%range) age of 6.09 (5.73-6.80). OHRQoL was assessed with a short form of the Child Oral Health Impact Profile at the children's age of 9.79 (9.49-10.44). In total, n= 2833 children participated in this study, of which n= 472 (16.6%) had mild caries (dmft 1-3) and n= 228 (8.0%) had severe caries (dmft>3). The higher the dmft-score at the age of 6, the lower the OHRQoL at the age of 10 ($p<0.001$). The children with severe caries at the age of 6 had significant higher odds to be in the lowest OHRQoL quartile at the age of 10 (OR=1.69; 95% CI=1.17-2.45). Our study highlights the importance of oral health during childhood, because those who get a compromised start to oral health are much more likely to follow a trajectory which will lead to poor oral health (-related quality of life) later. OHRQoL is not only related to current oral health experiences, but also to oral health experiences from the past.

Keywords: Caries, childhood, longitudinal study, quality-of-life, prognosis

## Introduction

Oral health-related quality of life (OHRQoL) is the perceived impact of one's own oral health on daily life and has commonly been defined as 'people's comfort when eating, sleeping and engaging in social interaction; their self-esteem; and their satisfaction with respect to their oral health' (1). Whereas OHRQoL had a difficult position in research for a long time, recently its use has enormously increased. Nowadays the scientific community is fully aware of the importance of OHRQoL for evidence-based dentistry. OHRQoL is subjective and multidimensional, encompassing the physical and psychological health state as well as the functioning in social interaction (2). Thus, the concept as such is very suitable to measure the perceived oral health of individuals.

Determinants of OHRQoL have been widely investigated, but are not yet understood in detail. Most research on determinants of OHRQoL is guided by the conceptual model of Wilson and Cleary (3). This model distinguishes between characteristics of the individual (e.g. self-esteem), characteristics of the environment (e.g. socio-economic position (SEP)) and clinical factors (e.g. caries), that influence OHRQoL. Determinants of OHRQoL are mainly investigated in cross-sectional studies. Psychosocial factors like self -esteem or sense of coherence and environmental factors like SEP seem positively correlated with OHRQoL (4, 5). Clinical factors are negatively correlated with OHRQoL (6, 7). The most important clinical factor in oral health research is the diagnosis of caries. Caries experience in childhood has a prevalence of 60 -90 % worldwide and is a strong predictor for later caries experience and other oral diseases (8, 9, 10).

However, all of the different individual, environmental and clinical factors, including caries, do not directly explain much of the variation in children's OHRQoL as shown in cross sectional studies. This raises the question, whether OHRQoL might also be related to oral health experiences from the past. Longitudinal research about different determinants on OHRQoL is scarce. The existing longitudinal literature mainly focusses on the evaluation of dental treatments, but does not investigate the longitudinal influence of disease and its related experiences on OHRQoL (11, 12). However, age emerged consistently as a crucial determinant of OHRQoL (13) and OHRQoL decreases with increasing age (14, 15, 16).

The relationship between childhood caries and OHRQoL at school-age has not yet been investigated in such a large cohort study. Research on the effect of caries treatment has shown, that patients perceive improved OHRQoL after treatment. However, treatment cannot shift the OHRQoL level as high as the OHRQoL level of caries free children (17).

In this study, we relate dental caries at the age of 6 with OHRQoL assessed at the children's age of 10, whether they were treated or not.

## Material and Methods

### Design and cohort

The present study was embedded in the Generation R Study, a population-based prospective cohort study from fetal life onwards, which was initiated to identify early environmental and genetic determinants of growth, development and health. The Generation R Study has been previously described in detail (18, 19). The Generation R Study has been conducted in accordance with the World Medical Association Declaration of

Helsinki and all study phases have been approved by the Medical Ethical Committee of the Erasmus Medical Centre, Rotterdam, The Netherlands (MEC-2012-165). The present study has been performed in compliance with the STROBE guidelines.
All pregnant women living in Rotterdam with an expected delivery date between April 2002 and January 2006 were invited to participate in the study. In the study phase from 5 years onwards n= 6690 visited the research center. Participants with missing information on caries at the age of 6 or OHRQoL at the age of 10 were excluded from this study. A detailed flowchart for the participants' selection is presented in figure 1.

### Caries experience

The decayed, missing and filled teeth index (dmft) was used to assess dental caries in the 6-year old children (20). The dmft-score of each child was obtained from intraoral photographs taken by trained nurses and dental students. Before taking photographs, the children brushed their teeth and the teeth were dried with a cotton roll. The images were taken with one of the two intraoral cameras, the Poscam USB intra-oral (Digital Leader PointNix) and Sopro 717 (Acteon) autofocus camera. Both cameras had a resolution of 640x480 pixels and a minimal scene illumination of f 1.4 and 30 lx. The whole dentition was captured with 10 photographs. The photographs were judged by one pediatric dentist (intra-rater reliability K=0.95) and a second calibrated pediatric dentist judged 10% of the photographs (inter-rater reliability of K =0.62). Scoring dental caries per tooth on intraoral photographs has been described elsewhere with a high sensitivity (85.5%) and specificity (83.6%) compared to ordinary oral examination (21). If one or more primary teeth were not able to be judged on the photographs, no dmft score was given.
Generally, the dmft-score ranges from 0 to 20. For the analysis, the dmft-index was categorized into three groups: caries free children (dmft = 0), children with mild caries experience (dmft-score = 1 -3) and children with severe caries experience (dmft-score > 3). The cut-off for the categorization was based on the mean dmft-score of five-year-old Dutch children (mean±sd: 1.6±2.5) and was also used in other studies (16, 22).

### Oral health-related quality of life

OHRQoL of the children was assessed at children's age of 10 by parental questionnaires with a shortened form of the Child Oral Health Impact Profile (COHIP) (23). This COHIP had 11 items, which covered the following domains of OHRQoL: oral symptoms, functional well-being, emotional well-being, school and peer interaction (appendix Table A.1). The questions referred to the oral health-related experiences in the last 3 month and were answered on a 5-point Likert scale: never (5 points), almost never (4 points), sometimes (3 points), often (2 points), and always (1 point). OHRQoL was scored by the sum of the answers with each question having the same weight. One item was positively formulated and therefore reversed to ensure that the highest score reflected the best possible OHRQoL. The final range of the COHIP score was 11.0 -55.0. Participants that missed answers on more than 3 items were excluded from the analysis. For participants that missed up to three answers, missing values were replaced by the mean answer score based on the remaining items. OHRQoL was not normally distributed and because transformation of the data did not lead to useable results, we decided to categorize

OHRQoL into quartiles (appendix Table A.2). In the descriptive Table 1 we present data on OHRQoL as median with 90% range. The median indicates the OHRQoL value which would separate the study population into two equally sized groups. The 90% range indicates the range of OHRQoL scores in which 90% of the children fall.

### Covariates

The following variables were considered to influence the association between dental caries at the age of 6 years and OHRQoL assessed at the age of 10: child's gender, ethnicity and age at answering the COHIP, socio-economic status (SES) measured as maternal education level (high vs. low), family income (< 2400€ vs. ≥ 2400€) and marital status of the mother (single vs partnership). Information of these potential covariates was collected by questionnaires at the children's age of 6. Furthermore, we considered following oral health (-behavior) variables as potential confounders: tooth-brushing frequency (once a day vs twice or more a day), dental visits in the last year (yes vs. no) and malocclusion at the age of ten. Malocclusion was assessed with the Dental Health Component of the Index of Orthodontic Treatment Need (IOTN-DHC). The IOTN-DHC was assessed from photographic and radiographic records taken at children's age of 10. This method has been described elsewhere (24).

### Data analysis

Statistical analysis were performed using the statistical package for social sciences (SPSS) for Windows, version 21 (IBM SPSS Inc, Chicago, IL, USA). Figures were created in Microsoft Word and Microsoft Excel (Microsoft Office Professional Plus 2010). Characteristics of the study population were explored with frequency tables and cross-tabulations stratified by caries experience at the age of 6 years. Differences in sample characteristics between children with and without dental caries at the age of 6 years were evaluated with Chi-square tests and Mann -Whitney-U tests.

Multinomial logistic regression was used to investigate the association between dental caries at the age of 6 and OHRQoL at the age of 10. We built 3 different models. The first model was the crude model only adjusted for gender and age. The second model was additionally adjusted for SEP and the third model took additionally oral health behavior into account. We only included those socioeconomic factors, that were statistically significant different between children with and without caries experience. We performed a test for trend analysis by treating the categorized variable as a continuous term. We also tested for interaction effects gender, ethnicity as well as orthodontic treatment need with caries, but none of them were significant. For all analyses a p-value < 0.05 was considered to be statistically significant. In order to reduce potential bias associated with missing data, we performed multiple imputation of missing covariates by generating 5 independent datasets using the Markov Chain Monte Carlo method, assuming no monotone missing pattern (25). This method takes into account the uncertainty of the imputed values to generate new datasets based on a fully conditional specified model. Imputations were based on the relationship between all variables considered to be included in the models, determinant and outcome were not imputed. We present effect estimated based on the pooled datasets. However, we also conducted a sensitivity analysis in the original data set (appendix Table A.3).

### Non-response analysis

The children included in this study were compared to the children excluded from the study. Children with missing data on dmft-index or OHRQoL score differed from children without missing these data on all covariates except for gender, tooth brushing frequency, malocclusions presence and OHRQoL (p-values > 0.05, appendix Table A.4).

## Results

### Sample characteristics

In total, n = 2833 children participated in this study. The median (90%range) OHRQoL of the children at the age of 10 was 50.00 (42.00 - 53.00), with a minimum score of 29.0 (prevalence: 0.1%) and a maximum score of 55.0 (prevalence: 0.9%). In Table 1 the maternal and child characteristics of the study population are presented by caries experience. In total, 24.7% (n =700) of the children experienced dental caries at the median (90%range) age of 6.09 (5.73 - 6.80), of which 32.6% (n = 228) had a dmft-score > 3. Mothers of children with dental caries were lower educated (p-value <0.001) had a lower household income (p-value < 0.001) and were more often of non-European origin (p-value <0.001). Child characteristics, as gender, age and oral health behavior did not differ between children with dental caries and children without dental caries. Children with caries at the age of 6 had significantly lower OHRQoL scores at the age of 10 compared to the children that were caries free at the age of 6 (p-value <0.001).

### Associations between dental caries and OHRQoL

In Table 2 the results of the multinomial logistic regression models illustrating the differences on OHRQoL between children without caries and with mild or severe dental caries at the age 6 are presented. Based on the crude model children with a mild dmft-score were 1.33 times and children with a severe dmft-score were 2.05 times more likely to be in the lowest OHRQoL quartile (mean±sd = 43.6±2.7) at the age of 10 compared to children that were caries free at the age of 6. After adjustment for SES, these associations between the children with a mild dmft-score and the lowest OHRQoL quartile lost significance (OR = 1.24 95%CI = 0.94 - 1.64), but for the children with a severe dmft-score the associations remained significant (OR = 1.69 95% CI = 1.17 - 2.45).

Although the association between mild or severe dmft-index and the 2$^{nd}$ and 3$^{rd}$ OHRQoL quartile (mean±sd = 48.2±0.8 resp. 50.0±0.0) were not statistically significant, a clear trend was visible and confirmed with the test for trend analysis. Thus, the higher the dmft-score at the age of 6, the lower the OHRQoL score at the age of 10 in all models (p <0.001). However, in figure 2 it is shown that the actual differences in median OHRQoL scores among the 3 groups (caries free= 50.0 (43.0-53.0), mild = 49.0 (41.0-53.0) and severe caries= 49.0(40.0-53.0)) are rather small.

Oral health related behavior was not associated with dental caries, nor with OHRQoL. Accordingly adjustment for oral health related behavior had no effect on the association between dental caries at the age of 6 and OHRQoL at the age of 10, as can be seen in the small differences in effect estimates between model 2 and model 3.

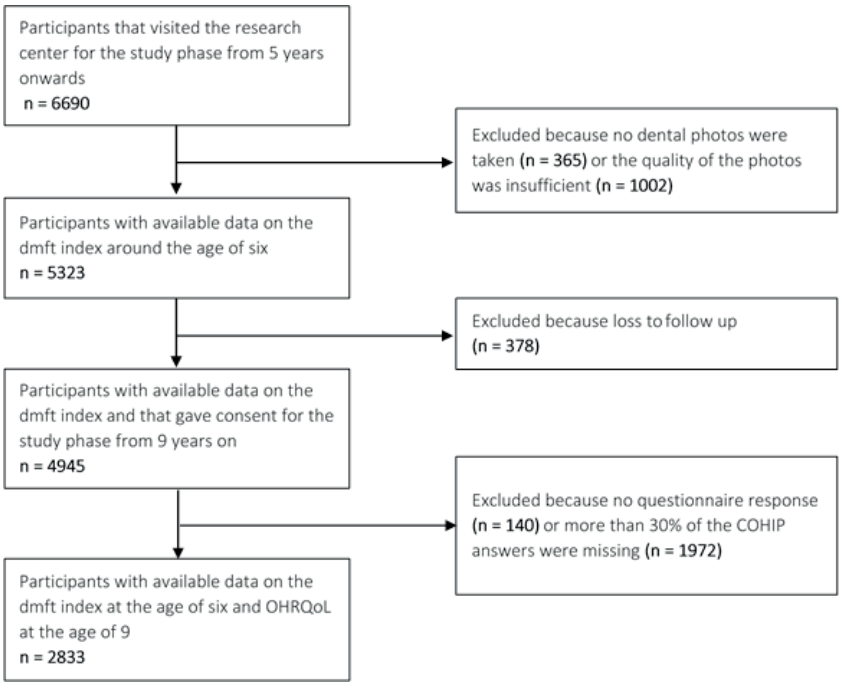


**Figure 1.** Flowchart of study selection. COHIP= Child Oral Health Impact Profile, OHRQoL = Oral health related quality of life.

## Discussion

The results of our study showed that caries experience at the age of 6 and OHRQoL at the age of 10 are inversely related. Especially children with severe caries at the age of 6 are significantly more likely to have lower OHRQoL at the age of 10 compared to children without caries experience at the age of 6. Based on our results, we highly endorse the importance to target oral health prevention strategies at young children.

Several previous studies assessed the influence of caries on OHRQoL. Comparison with our results is difficult, because most of the studies have a cross-sectional design, use a different OHRQoL measure or investigate the effect of treatment instead of disease. Nevertheless, caries had an impact on OHRQoL cross-sectionally assessed (26). Effect estimates of these studies indicated consistently a 3-fold increased risk for lower OHRQoL in children with dental caries compared to children without dental caries (relative risk (RR) = 2.97 (1.61–5.47) (15); RR = 2.74 (2.02–3.72) (27); RR = 3.81 (2.66- 5.46) (28). Because of the large time interval between the caries assessment and OHRQoL assessment in our study, we expected a weaker association compared to the previous cross-sectional studies. Also because one other study showed an RR = 1.4 (1.1-1.7) for the relationship between untreated dental caries at the age of 6 (dmft-score > 3) and OHRQoL at the age of 12 (29).

**Table 1.** Sample characteristics of the study population stratified by caries experience (n=2833)

| Characteristics | | dmft = 0 | dmft > 0 | p -value |
|---|---|---|---|---|
| | | n=2133 | n= 700 | |
| *Maternal characteristics* | | | | |
| **Educational level** | | | | |
| | Low | 124 | 92 | |
| | High | 1925 | 560 | < 0.001 |
| | Missing (%) | 4.0 | 6.9 | |
| **Household income** | | | | |
| | < 2400€ | 396 | 213 | |
| | ≥ 2400€ | 1574 | 414 | < 0.001 |
| | Missing (%) | 7.6 | 10.4 | |
| **Ethnicity** | | | | |
| | European | 1691 | 447 | |
| | Non-European | 434 | 250 | < 0.001 |
| | Missing (%) | 0.4 | 0.4 | |
| **Marital status** | | | | |
| | Single | 167 | 53 | |
| | Partnership | 1851 | 607 | 0.842 |
| | Missing (%) | 5.4 | 5.7 | |
| *Child characteristics* | | | | |
| **Gender** | | | | |
| | Boys | 1065 | 334 | |
| | Girls | 1068 | 366 | 0.309 |
| | Missing (%) | 0.0 | 0.0 | |
| **Age** | | | | |
| | at caries assessment | 5.97 (5.73 - 6.65) | 6.03 (5.76 - 7.19) | < 0.001 |
| | at OHRQoL assessment | 9.78 (9.48 - 10.43) | 9.87 (9.52 - 10.55) | 0.086 |
| | Missing (%) | 0.0 | 0.0 | |
| **Toothbrush frequency** | | | | |
| | Once per day | 408 | 135 | |
| | Twice or more per day | 1595 | 501 | 0.641 |
| | Missing (%) | 6.0 | 9.1 | |
| **Dental visits last year** | | | | |
| | Yes | 1888 | 611 | |
| | No | 122 | 30 | 0.188 |
| | Missing (%) | 5.7 | 8.4 | |
| **Malocclusion at age 9** | | | | |
| | Present | 1138 | 294 | |
| | Absent | 653 | 239 | < 0.001 |
| | Missing (%) | 16.0 | 23.9 | |
| **OHRQoL** | | | | |
| | | 49.00 (40.00 - 53.00) | 48.00 (37.00 - 52.00) | < 0.001 |
| | Missing (%) | 0.0 | 0.0 | |

Numbers are absolute values for categorical variables or median (90% range) for continuous variables, missing values are given in percentages. p values are based on chi-square tests for categorical variables or Mann-Whitney-U tests for continuous variables. dmft = decayed, missing and filled teeth; OHRQoL = oral health related quality of life;

Similarly in our study, children with a dmft-score > 3 at the age of 6 were 1.69 times more likely to be in the lowest OHRQoL quartile at the age of 10 compared to children with a dmft-score = 0. The effect sizes in our study need to be interpreted carefully, because they do not show the actual decrease in OHRQoL but the likelihood to be in the lowest OHRQoL quartile of our particular study population. The absolute decrease in OHRQoL is rather small.Small effect sizes are a common problem in quality of life research, because there is always a tendency towards high scores (30). No consensus exists about the minimal clinical difference of the OHRQoL measure we use, however this might also be less relevant as children with caries experience at the age of 6 have a systematic lower OHRQoL at the age of 10.

**Table 2.** Association between dmft and oral health related quality (n=2833)

| | | Oral health related quality of life | | | | |
|---|---|---|---|---|---|---|
| | | Highest Quartile | Mid highest Quartile | Mid-lowest Quartile | Lowest Quartile | |
| | n | 1100 | 341 | 868 | 524 | |
| | mean±sd | 51.6±1.0 | 50.0±0.0 | 48.2±0.8 | 43.6±2.7 | |
| **Caries measure** | | | OR (95%CI) | OR (95%CI) | OR (95%CI) | $p_{for\ trend}$* |
| **Dmft = 0** (n = 2133) | | | Ref | Ref | Ref | |
| **dmft = 1-3** (n = 472) | Model 1 | | 1.13 (0.82 - 1.58) | 1.17 (0.92 - 1.49) | 1.33 (1.01 - 1.76) | |
| | Model 2 | reference | 1.16 (0.83 - 1.62) | 1.18 (0.93 - 1.51) | 1.24 (0.94 - 1.64) | |
| | Model 3 | | 1.14 (0.82 - 1.60) | 1.16 (0.91 - 1.49) | 1.21 (0.91 - 1.61) | |
| **dmft > 3** (n = 228) | Model 1 | | 0.94 (0.57 - 1.56) | 1.23 (0.87 - 1.73) | 2.05 (1.44 - 2.94) | < 0.001 |
| | Model 2 | reference | 1.02 (0.61 - 1.72) | 1.27 (0.90 - 1.82) | 1.69 (1.17 - 2.45) | 0.002 |
| | Model 3 | | 0.98 (0.60 - 1.65) | 1.19 (0.83 - 1.70) | 1.48 (1.02 - 2.16) | 0.017 |

OR = odds ratios; CI = confidence interval; odds ratios are estimated using multinomial logistic regression models. Model 1 is adjusted for child's gender and age; model 2 is additionally adjusted for socio-economic indicators and model 3 is additionally adjusted for oral health behavior and orthodontic treatment need; dmft = decayed, missing and filled teeth ; *p-value for trend calculated by adding the dmft variable treated continuous term to the model

The result of our study should be seen in the light of several limitations. Mothers of children who were included in the study were on average higher educated, had a higher household income and had more often an European background than those of children who were excluded. This could have led to a selection bias, if the association between caries and OHRQoL differed between those excluded and included, but this seems unlikely. We do not distinguish between the effect of treated and untreated dental caries on later OHRQoL, as was done by Peres et al (29). As we assessed the dmft-index at one time, misclassification of treated and untreated caries would have been likely, because there is no consensus among dentists how to treat dental caries in the primary dentition and treatment could have taken place shortly after the dmft assessment. Furthermore, for our research question dental treatment was less important, because we were interested in the effect of early caries experience on OHRQoL later in life. Finally, the assessment of dental caries from intraoral photographs might be generally limited, since oral examination is the gold standard and intraoral photographs have been validated to perform moderately for the clinical diagnosis of dental caries, but suitable for scientific studies (21, 31) Therefore, the assessment of dmft indices in this study might be affected by a non-differential measurement error leading to misclassification and underestimation of the association between dental caries and OHRQoL. We also did not adjust our analysis for dental caries at the time point of OHRQoL assessment, because these data were not available. At the time point of OHRQoL assessment the children were 10 years old and thus having a transitional dentition, which would have limited the reliability of the dmft-index. Although our analyses were adjusted for SES and oral

health behavior, residual confounding might have occurred. Nevertheless, the present study is the first one worldwide evaluating the longitudinal relationship of caries with OHRQoL in a large population-based prospective cohort study among n = 2833 children. The prospective design of the study is most suitable to evaluate long term effects of caries, as caries is still a highly prevalent disease during childhood (32).

We have two theoretical explanations for the present results. First, the strong relationship between caries experience at the age of 6 and OHRQoL at the age of 10 could be based on the fact that caries experience at young age is a strong predictor for later caries. Different studies have shown that the main risk factor for dental caries is previously experienced caries (9, 10, 33). In this case, the caries assessment at the age of 6 could be seen as a proxy for caries at the age of 10 and our effect estimates are mediated by the cross-sectional relation between dental caries and OHRQoL. Our trend results have supported this hypothesis, because the higher the dmft-score at the age of 6, the lower OHRQoL at the age of 10. Secondly, the strong relationship between caries experience at the age of 6 and OHRQoL at the age of 10 could be rather based on psychological factors. Children with severe caries at a young age remember their bad experiences and therefore still have impaired OHRQoL later in life. Dental caries affects OHRQoL via pain perception (7, 29, 34). Although the effect of negative dental experiences on dental anxiety has been contradictory (35, 36), dental anxiety in turn is strongly related with lower OHRQoL (36, 37). Furthermore, it is likely that OHRQoL is strongly related to personal perceptions of oral health, as it is designed to measure psychological wellbeing related to the oral health status. The personal perception of one owns oral health is formed by experiences (38). Most likely, our results show a mixture of the longitudinal and cross-sectional influences of caries on OHRQoL. Further studies could focus on the individual effects of both, longitudinal and cross-sectional effects of caries experience separately on OHRQoL.

This research gives insight into longitudinal influences of clinical factors, as caries, on OHRQoL and subsequently promotes oral health prevention strategies for young children. Our study highlights the importance to promote good oral health during childhood, because those who get a compromised start to oral health are much more likely to follow a trajectory which will lead to poor oral health (-related QOL) later. The development of risk factors, like unhealthy habits or socio-economic disadvantage, tend to persist and increase through life and need to be tackled as early as possible. Multicomponent interventions targeting at individual level determinants, family level determinants and environmental determinants of poor oral health in childhood should be designed, as it is known from other public health areas that those multi-component interventions are most effective (39).

In conclusion, dental caries at the age of 6 can be seen as a predictor for lower OHRQoL at the age of 10. Whether this association exists because children with former caries experience do also have more caries later in childhood or because the experiences of dental caries at a young age have caused a longitudinal impairment of OHRQoL needs further research.

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## Supplemental Material

**Table S1.** The 11 items of the COHIP, used in the present study

During the past 3 month, how often has your child:

1. had pain in his/her teeth/toothache
2. had crooked teeth or spaces between his/her teeth
3. had discolored teeth or spots on his/her teeth
4. had bad breath
5. had bleeding gums
6. had difficulty eating foods he/she would like to because of his/her teeth, mouth, or face
7. felt worried or anxious because of his/her teeth mouth or face
8. not wanted to speak/ read out loud in class because of his/her teeth, mouth, or face
9. been teased, bullied or called names by other children because of his/ her teeth, mouth, or face
10. felt that he/she was attractive (good looking) because of his/ her teeth, mouth, or face
11. had difficulty saying certain words because of his/her teeth or mouth

**Table S2.** OHRQoL quartiles used in the study

| Group of OHRQoL | N | Mean | SD |
|---|---|---|---|
| **1** | 524 | 43.56 | 2.67 |
| **2** | 868 | 48.20 | 0.83 |
| **3** | 341 | 50.00 | 0.00 |
| **4** | 1100 | 51.60 | 1.03 |

**Table S3.** Association between dmft and oral health related quality - original data

| | | Oral health related quality of life | | | | |
|---|---|---|---|---|---|---|
| | | Highest Quartile n = 1100 | Mid highest Quartile n = 341 | Mid-lowest Quartile n = 868 | Lowest Quartile n = 524 | |
| **Caries measure** | | | OR (95%CI) | OR (95%CI) | OR (95%CI) | $p_{for\ trend}$ |
| **dmft = 1-3** | | | | | | |
| | Model 1 | reference | 1.13 (0.82 - 1.58) | 1.17 (0.92 - 1.49) | 1.32 (1.01 - 1.76) | |
| | Model 2 | | 1.19 (0.84 - 1.69) | 1.20 (0.93 - 1.55) | 1.30 (0.96 - 1.74) | |
| | Model 3 | | 1.01 (0.67 - 1.54) | 1.09 (0.81 - 1.47) | 1.30 (0.92 - 1.83) | |
| **dmft > 3** | | | | | | |
| | Model 1 | reference | 0.94 (0.57 - 1.56) | 1.23 (0.87 - 1.73) | 2.05 (1.44 - 2.94) | < 0.001 |
| | Model 2 | | 1.19 (0.70 - 2.04) | 1.21 (0.82 - 1.79) | 1.93 (1.30 - 2.87) | 0.001 |
| | Model 3 | | 1.08 (0.57 - 2.06) | 1.21 (0.77 - 1.90) | 1.89 (1.18 - 3.02) | 0.007 |

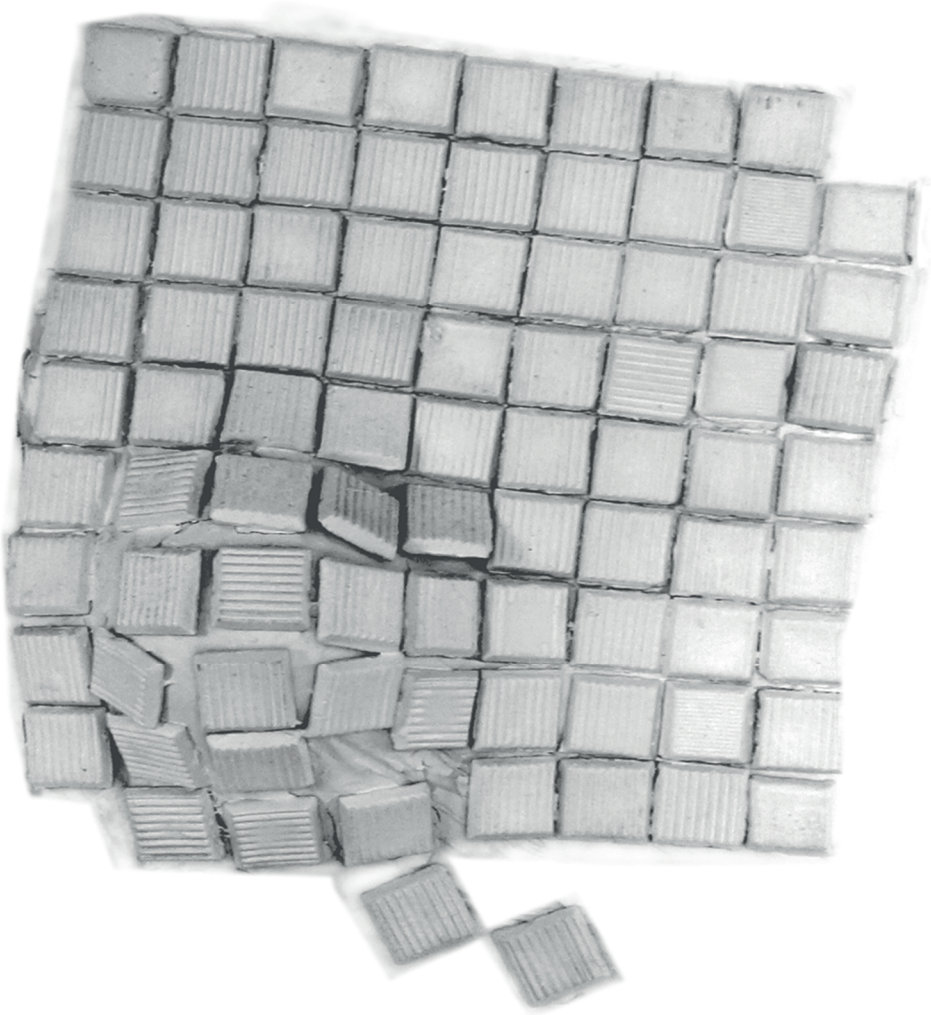
OR = odds ratios; CI = confidence interval; odds ratios are estimated using multinomial logistic regression models. Model 1 is adjusted for childs gender and age; model 2 is additionally adjusted for socio-economic indicators and model 3 is additionally adjusted for oral health behavior.

**Table S4.** Non-response analysis n= 8305

| Characteristics | | Population for analysis | Excluded population | p -value |
|---|---|---|---|---|
| | | n=2833 | n=3857 | |
| *Maternal characteristics* | | | | |
| **Educational level** | | | | |
| | Low | 216 (8.0) | 587 (16.5) | |
| | High | 2485 (92.0) | 2966 (83.5) | < 0.001 |
| **Household income** | | | | |
| | < 2400€ | 609(23.5) | 1196(36.3) | |
| | ≥ 2400€ | 1988(76.5) | 2101(63.7) | < 0.001 |
| **Ethnicity** | | | | |
| | European | 2138(75.8) | 2969 (57.7) | |
| | Non-European | 684(24.2) | 2179(42.3) | < 0.001 |
| **Marital status** | | | | |
| | Single | 220 (8.2) | 751 (15.7) | |
| | Partnership | 2458 (91.8) | 4033 (84.3) | < 0.001 |
| *Child characteristics* | | | | |
| **Gender** | | | | |
| | Boys | 1399 (49.4) | 2789 (51.0) | |
| | Girls | 1434 (50.6) | 2681 (49.0) | 0.166 |
| **Age** | | | | |
| | at caries assessment | 6.09 (5.73 - 6.80) | 6.24 (5.72 - 7.64) | < 0.001 |
| | at OHRQoL assessment | 9.79 (9.49 - 10.44) | 9.78 (9.48 - 10.46) | 0.001 |
| **Toothbrush frequency** | | | | |
| | 1 keer per dag | 543 (20.6) | 726 (21.3) | |
| | ≥ 2 keer per dag | 2096 (79.8) | 2690 (78.7) | 0.521 |
| **Dental visits last year** | | | | |
| | Yes | 2499 (94.1) | 3125 (91.3) | |
| | No | 152 (5.9) | 298 (8.7) | < 0.001 |
| **Malocclusion** | | | | |
| | Present | 892 (38.4) | 889 (38.5) | |
| | Absent | 1432 (61.6) | 1422 (61.5) | 0.952 |
| **OHRQoL** | | | | |
| | | 50.00 (42.00 - 53.00) | 50.00 (43.00 - 53.00) | 0.085 |
| **Dmft-index** | | | | |
| | 0 | 2133 (75.3) | 1515 (60.8) | |
| | 1-3 | 472 (16.7) | 542 (21.8) | |
| | >3 | 228 (8.0) | 433 (17.4) | < 0.001 |

Numbers are absolute values (%) for categorical variables or median (90% range) for continuous variables, missing values are given in percentages. p values are based on chi-square tests for categorical variables or Mann-Whitney-U tests for continuous variables. Dmft = diseased, missing and filled teeth; OHRQoL = oral health related quality of life;

# Chapter 4
# Characteristics of the environment influencing OHRQoL



# Chapter 4.1

## Social inequalities in children's oral health-related quality of life: The Generation R Study

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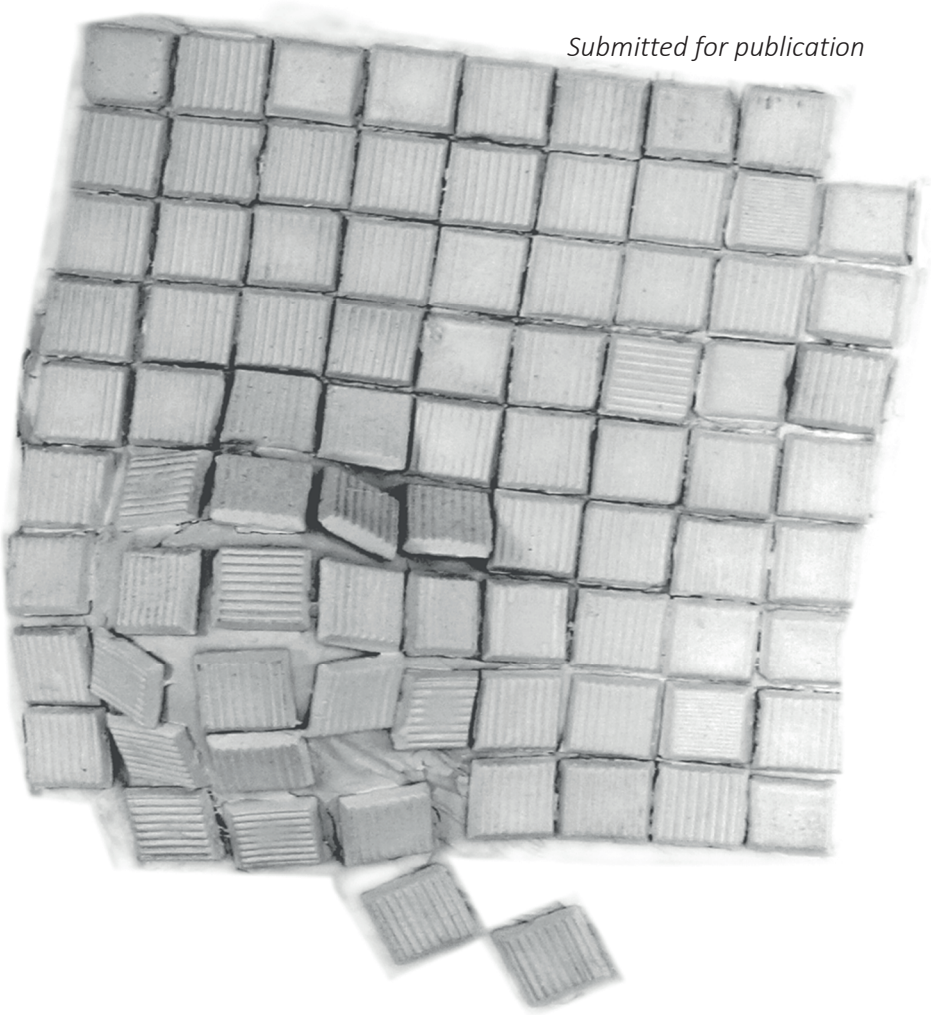
*Submitted for publication*



Abstract

***Purpose*** Oral health-related quality of life (OHRQoL) is the most important patient-reported outcome measure in oral health research. The purpose of the present research was to study the association of family socioeconomic position (SEP) with children's OHRQoL.

***Methods*** This cross-sectional study was embedded in the Generation R Study, a population-based cohort study conducted in Rotterdam, The Netherlands. For the present study, OHRQoL was assessed of 3871 10-year old children. Family SEP was assessed with the following indicators: maternal/paternal education level, maternal/paternal employment status, household income, benefit dependency and family composition. Linear regression analyses were performed to evaluate the (independent) associations of family SEP indicators with OHRQoL.

***Results*** The median(90%range) OHRQoL score of the participating children was relatively high (50.0 (43.0 – 53.0)), however OHRQoL was consistently lower in children with low family SEP. Negative associations were found for all SEP indicators ( p-values < 0.05) except maternal education level and employment status. Benefit dependency, paternal employment and household income were the strongest associated with OHRQoL. No family SEP indicator was significantly associated with OHRQoL independent of the other indicators.

***Conclusions*** Based on the present findings, interventions and policies promoting good oral health and oral wellbeing should target children from low socioeconomic position. More research is needed, however, to understand the pathways of social inequalities in children's OHRQoL especially for the effects of material resources on subjective oral health measures.

***Keywords:*** Quality of life, oral health, social inequalities, children

## Background

Oral health related quality of life (OHRQoL) is a commonly used patient reported outcome measures in dental research. This measure is designed to assess the impact of oral diseases from the patient perspective and is thus subjective and multidimensional. It is particularly suited to assess oral health of individuals, because it encompasses their physical function, psychological state, social interaction and somatic sensation (1). Quality of life measures in medical and dental research become increasingly important because of the patients more active participation in their health treatment, because of the need for new evidence in oral health practices and because more and more diseases cannot be cured by the treatment although they improve the patient's condition (1,2).
Research on OHRQoL is directed by the Wilson and Cleary Model, which shows the possible link between biological and clinical variables, characteristics of the individual and the environment and other non-medical factors on OHRQoL (3,4). Many studies suggest that socioeconomically deprived persons tend to have worse oral health and unmet dental treatment needs (5-9). In line with the Wilson and Cleary model, these inequalities have also been shown in subjective oral health measures among adolescents, adults and elderly (10-13). Also several studies have been conducted on the relation between socioeconomic status and children's OHRQoL, but these make use of various study populations, various socioeconomic indicators, various methods and statistical analyses (13). Yet, the evidence for the relation between family socioeconomic position (SEP) and OHRQoL in preschool-aged children is inconclusive (13, 14).
OHRQoL is important and it is necessary to identify risk groups at an early stage, because poor oral health will track through childhood into adolescence and adulthood (15-17). Therefore, the aim of the present study was to investigate the associations of family SEP with children's OHRQoL. For this research we used the data from The Generation R Study, which is a large multi-ethnic birth cohort in Rotterdam, The Netherlands.

## Material & Methods

### *Study design*

This cross-sectional study is performed within the Generation R Study, which is an ongoing multiethnic population-based prospective cohort study. This study has been described in detail elsewhere (18). The Generation R Study has been conducted following the World Medical Association Declaration of Helsinki and was approved by the Medical Ethics Committee at Erasmus Medical Centre, Rotterdam, The Netherlands (MEC-2012-165). Written consent was obtained from all participants before data collection started.

### *Study population*

Invitations to participate in the study were given to all pregnant women with an expected delivery date between April 2002 and January 2006 living in the study area (Rotterdam, The Netherlands). From the original 9749 live born children included in the Generation R cohort, 7393 children still participated in the follow up period from the children's age of nine years onwards. From these, we selected the children with available data on OHRQoL, which was assessed at the median (90% range) age of 9.79 (9.48 - 10.47). In total, 3871 children were included in this study.

*OHRQoL*

OHRQoL of the children was assessed by parental questionnaires. For this a Dutch 11-item version of the Children's Oral Health Impact Profile (COHIP) was used, which has previously been validated in a comparable population (19). The questions cover the five subdomains of children's oral health: oral symptoms, functional well-being, emotional well-being, school and peer interaction. The questions inquire about the frequency of oral health impacts on daily life and are answered on a 5-point Likert scale: never (5 points), almost never (4 points), sometimes (3 points), often (2 points), and always (1 point). All answers were added up to a final OHRQoL score (range 11-55 points), with the highest score indicating the best quality of life. Missing values in the responses to the questionnaire were replaced by the personal mean score of the remaining answers to the questions, as it is proposed by other researchers that used the original version of the COHIP (20). If there were more than 30% of the answers missing, the participant was excluded from the analysis.

*Family social position*

Following socioeconomic indicators of family SEP were considered in the present study: maternal and paternal education level, maternal and paternal employment status and net household income, which are all traditional family SEP indicators (21). We also used receiving benefits and single parenting as additional family SEP indicators, because these were associated with oral health in previous research (13, 22). Parental education was assessed at the children's age of six by questionnaires and defined as low (no education, primary school, lower or intermediate vocational training, general school or first year of higher vocational training) or high (higher vocational training, university or PhD degree). Also, information on paternal and maternal employment status was assessed by questionnaires at the children's age of six and categorized into no paid job (unemployed, disabled, welfare recipient, housewife or student, other non-paid work) or paid job (paid or self-employed). Information on net household income (≤ 2000€ vs. >2000€), receiving benefits (no vs. social security, unemployment benefits, disability allowances or other) and single parenting were assessed in parental questionnaires around children's age of ten, which were the same as for the assessment of OHRQoL.

*Confounders*

Based on literature and experience in clinical practice child's sex, age and ethnic background were considered confounders in the association between family SEP and OHRQoL. In addition, following oral health variables were considered as potential confounders for the relationship between family SEP and OHRQoL: caries experience, orthodontic treatment need based on either the Dental Health Component (IOTN-DHC) or the aesthetic component (IOTN-AC) of the Index of Orthodontic treatment need and self-perceived orthodontic treatment need (23). Because not all considered confounders were assessed in the same follow-up period as OHRQoL, i.e. around the children's age of 10, we used some measurements from previous time points.

Child's ethnicity was defined following the guidelines for classification by Statistics Netherland (24). Children's ethnic background, assessed at enrollment in the Generation R Study, was based on the country of birth of the parents. Children of parents, that

both were born in the Netherlands were classified as native Dutch. If at least one of the parents was not born in the Netherlands, the child was classified as non-native Dutch.
Caries experience was assessed at the children's age of six with the decayed missing and filled teeth index (dmft) which ranges from 0 to 20. The dmft-score of each child was obtained from intraoral photographs. Before taking photographs, the children brushed their teeth and the teeth were dried with a cotton roll. The images were taken with one of the two intraoral cameras, the Poscam USB intra-oral (Digital Leader PointNix) and Sopro 717 (Acteon) autofocus camera. Both cameras had a resolution of 640x480 pixels and a minimal scene illumination of f 1.4 and 30 lx. The whole dentition was captured with 10 photographs. The photographs were judged by one pediatric dentist (intra-rater reliability K=0.95) and a second calibrated pediatric dentist judged 10% of the photographs (inter-rater reliability of K =0.62). Scoring dental caries per tooth on intraoral photographs has been described elsewhere with a high sensitivity (85.5%) and specificity (83.6%) compared to ordinary oral examination (25). If one or more primary teeth were not able to be judged on the photographs, no dmft score was given. Children were categorized into no caries experience (dmft = 0) versus caries experience (dmft > 0).
The IOTN-DHC and IOTN-AC was assessed from photographic and radiographic records of the children taken around the age of ten. Assessment of the IOTN on a combination of photographic and radiographic records has been validated previously (26). After six months 10% of the photographs were reassessed by first the same examiner (LK) and second by another examiner (EO). With these measurements intra-rater reliability (linear weighted K = 0.84) and inter-rater reliability was calculated (linear weighted K = 0.68).
Self-perceived orthodontic treatment need was measured in the parental questionnaires around children's age of ten with the question: "Do you want your child to get braces?" The question was answered on a five point Likert scale, with answer possibilities ranging from 'strongly disagree' to 'strongly agree'. Answers were categorized into 'self-perceived need' (strongly / somewhat agree), 'borderline self-perceived need' (do not agree or disagree) and 'no need' (strongly / somewhat disagree).

*Statistical analysis*

First, we used descriptive statistics to characterize the study population.
The associations of family SEP indicators with OHRQoL were analyzed with series of weighted least squares linear regression analysis. For all indicators, 3 different models were created. First, we created the crude model adjusted for child's age, gender and ethnic background only. Second, we created model 1 adjusted for confounders. Potential confounders were included in model 1, when they changed the estimate with approx. 10%, when they were significant when entered into the crude model or when the $R^2$ of the model improved. Finally, we created model 2 adjusted for confounding variables and all other family SEP indicators simultaneously. Model 2 was created to evaluate the independent effects of each of the SEP indicators. To assess the explanatory effects of the oral health variables on the association between a particular SEP indicator with OHRQoL, the differences between the crude model and model 1 were compared $((\beta_{\text{crude model}} - \beta_{\text{model 1}})/\beta_{\text{crude model}})*100\%)$. This approach allows also to evaluate the influence of SEP indicators on oral health from the patient perspective. Likewise, the explan-

atory effects of the other family SEP indicators on the association between a particular SEP indicator with OHRQoL were assessed. Significance of the difference was assessed with a test for heterogeneity. Finally, we tested for multicollinearity in model 2 by obtaining the tolerance and VIF values for each determinant and covariate. Tolerance values above 0.10 and VIF values below 10 were considered acceptable to rule out multicollinearity (27, 28).

**Table 1.** Sample characteristics of the study population (n = 3871)

| | | **Total n** (%) | **Missing n** (%) |
|---|---|---|---|
| **Maternal education level** [a, c] | | | |
| | low | 1267 (32.7) | 254 (6.6) |
| | high | 2350 (60.7) | |
| **Paternal education level** [a, c] | | | |
| | low | 1220 (31.5) | 489 (12.6) |
| | high | 2162 (55.9) | |
| **Maternal employment status** [c] | | | |
| | paid job | 2785 (71.9) | 414 (10.7) |
| | no paid job | 672 (17.4) | |
| **Paternal employment status** [c] | | | |
| | paid job | 3164 (81.7) | 558 (14.4) |
| | no paid job | 149 (3.8) | |
| **Household income**[d] | | | |
| | < 2000€ | 643 (16.6) | 273 (7.1) |
| | > 2000€ | 2955 (76.3) | |
| **Receiving benefits** [b, d] | | | |
| | Yes | 394 (10.2) | 117 (3.0) |
| | No | 3360 (86.8) | |
| **Family composition** [d] | | | |
| | One parent | 553 (14.3) | 126 (3.3) |
| | Two parents | 3192 (82.5) | |
| **Ethnicity** | | | |
| | native Dutch | 2626 (67.8) | 61 (1.6) |
| | non- Dutch | 1184 (30.6) | |
| **Childs sex** | | | |
| | male | 1923 (49.7) | 0 (0.0) |
| | female | 1948 (50.3) | |
| **Childs age** | | | |
| | median (90% range) | 9.79 (9.49 - 10.47) | 0 (0.0) |
| **Caries experience** [c] | | | |
| | no | 2167 (56.0) | 991 (25.6) |
| | yes | 713 (18.4) | |
| **Orthodontic need** [d] | | | |
| | no | 1902 (49.1) | 823 (21.3) |
| | yes | 1146 (29.6) | |
| **Aesthetic orthodontic need** [d] | | | |
| | no | 1691 (43.7) | 927 (23.9) |
| | borderline | 1006 (26.0) | |
| | definite | 247 (6.4) | |
| **Self-perceived orthodontic need** [d] | | | |
| | no | 1075 (27.6) | 22 (0.6) |
| | borderline | 980 (25.2) | |
| | yes | 1794 (46.5) | |
| **OHRQOL** [d] | | | |
| | median (90% range) | 50.00 (43.00 - 53.00) | 0 (0.0) |

The Table is based on the non-imputed dataset. Data are presented as absolute numbers with percentages for categorical data and as median with 90% range for continuous data. [a] educational level: low = no education, primary school, vocational training, general secondary school and first year higher vocational training, high = higher vocational training, university or PhD degree; [b] benefits : social security, unemployment benefits, disability allowances and other; [c] assessed at children's age of 6 ; [d] assessed at children's age of 10.

We conducted a non-response analysis by comparing children with data available on OHRQoL with the children that had no data on OHRQoL on all family SEP indicators and confounders using Mann-Whitney-U tests or chi-square tests. Missing data in the determinants and covariates were multiple imputed based on the other determinants, covariates and OHRQoL. Ten imputed datasets were created using a fully specified model of which we present the pooled regression coefficients with 95% confidence intervals (aβ, 95%CI).
All analysis were performed using IBM SPSS Statistics version 21.0 for Windows (SPSS INC., Chicago, IL, USA). A significance level of $p< 0.05$ was used for all analysis.

## Results

### *Sample characteristics*

The non-response analysis showed, that children without information on OHRQoL had more often parents from a lower socio-economic status (supplemental Table S2). In table 1 the characteristics of the study population are presented. Most of the children were native Dutch (67.8%). Approximately one-third of all the children had a mother or father with a low education level (32.7 %, 31.5% resp.) Almost one-fifth of the children lived in a household with an income below 2000€ per month. Prevalence of oral health variables were relatively high, with approximately 18.4% of the children having caries experiences, 29.6% of the children having objective and 46.5% having subjective orthodontic treatment need. The median (90%range) OHRQoL score of the children was 50.00 (43.00 - 53.00).

**Table 2.** Associations of socioeconomic indicators with OHRQOL at children's age of 10

| | | Crude model<br>β [95%CI] | Model 1<br>β [95%CI] | Model 2<br>β [95%CI] |
|---|---|---|---|---|
| **Maternal education level** [a, c] | | | | |
| | low | -0.21<br>[-0.44 - 0.17] | -0.11<br>[-0.33 - 0.10] | 0.03<br>[-0.21 - 0.27] |
| **Paternal education level** [a, c] | | | | |
| | low | -0.33<br>[-0.56 - -0.10] | -0.24<br>[-0.45 - -0.04] | -0.20<br>[-0.44 - 0.03] |
| **Maternal employment status** [c] | | | | |
| | no paid job | -0.09<br>[-0.39 - 0.21] | 0.02<br>[-0.25 - 0.30] | 0.11<br>[-0.18 - 0.40] |
| **Paternal employment status** [c] | | | | |
| | no paid job | -0.59<br>[-1.19 - -0.00] | -0.45<br>[-0.99 - -0.09] | -0.29<br>[-0.84 - 0.26] |
| **Household income**[d] | | | | |
| | < 2000€ | -0.45<br>[-0.77 - -0.12] | -0.40<br>[-0.70 - -0.11] | -0.21<br>[-0.57 - 0.16] |
| **Receiving benefits** [b, d] | | | | |
| | yes | -0.52<br>[-0.91 - -0.14] | -0.51<br>[-0.87 - -0.14] | -0.33<br>[-0.70 - 0.04] |
| **Family composition** [d] | | | | |
| | single parent | -0.39<br>[-0.71 - 0.07] | -0.36<br>[-0.65 - -0.06] | -0.09<br>[-0.42 - 0.24] |

The data are presented as linear regression coefficients (β) with 95%-confidence intervals (95%CI). The crude model is adjusted for gender, age and ethnicity only. Model 1 is additionally adjusted for confounders: caries experiences, orthodontic treatment need, aesthetic treatment need and self-perceived orthodontic treatment need. Model 2 is additionally adjusted for confounders and the other socioeconomic factors. Significant associations are printed bold.

*Association of family SEP indicators with OHRQoL*

The correlation between all family SEP indicators varied between 0.08 and 0.54 (supplemental Table S3). The VIF values for determinants and confounders in the models used to describe the associations between family SEP indicators and OHRQoL were all below 1.5, the tolerance values were all above 0.70 (supplemental Table S4). In table 2 the associations between family SEP and OHRQoL are presented. Children of fathers with low educational level had lower OHRQoL than children of fathers with a high education level (crude model: aβ: -0.33 (95%CI: -0.56 - -0.10)). Similarly, significantly lower OHRQoL was seen in children of unemployed fathers (aβ: -0.59 (95%CI: -1.19 - -0.00)), children with a low household income (aβ: -0.45 (95%CI: -0.77 - -0.35)), children living in a household that receives any kind of benefits (aβ: -0.52 (95%CI:-0.91 - -0.14) or a single-parent family (aβ:-0.39 (95%CI: -0.71 - -0.07). Thus, all family SEP indicators, except maternal educational level, were negatively associated with OHRQoL.

All of these associations, except for paternal employment status, remained significant after adjustment for oral health variables (model 1, table 3). The oral health variables explained between 1.9% and 47.6% of the relationship between the different SEP indicators and OHRQoL. After adjustment for the other family SEP indicators, benefit dependency and paternal employment status were the strongest associated with OHRQoL (aβ: -0.33 (95%CI: -0.70 - -0.04), resp. aβ: -0.29 (95%CI: -0.84 - -0.26)). However, there were no significant independent family SEP associations with OHRQoL found (model 2, table 3). The associations between family SEP indicators and OHRQoL were explained by the other family SEP indicators between 16.7% and 75.0%.

## Discussion & Conclusion

Family SEP was consistently negatively associated with OHRQoL. Moreover, children with lower family SEP perceived lower OHRQoL independent of their objective oral health status.

Our results suggest that not only clinical variables, as caries and malocclusions, are associated with lower OHRQoL, but also different socio-economic and environmental variables interfere significantly in children's conditions of daily life. This is in line with other research that showed how socio-environmental factors are related to lower OHRQoL in 12-year old children (14), as well as with studies that show socioeconomic inequalities in objective oral health (7,29).

Many studies suggest but have not found conclusive evidence, that the association of family SEP with children's OHRQoL may be related to oral health behavior, like tooth brushing frequency, sugar intake and regular dental visits (30-32). However, in different studies, low socioeconomic status was associated with less oral hygiene, higher added sugar intake and less dental service use (33-35). We did not specifically adjust our models for oral health behavior. However, our results stay significant after adjustment for oral health variables like caries experience, which is obviously related to oral health behavior. Thus, we hypothesize that the effect of family SEP on OHRQoL is attributed to several additional factors, so called mediators, rather than simply to oral hygiene and oral health.

One mediator that might contribute to the association between family SEP with OHRQoL is related to aspects of self-esteem and self-perception about oral health and

body image. One study showed that socio-economic disparities in self-perceived oral health might partly be mediated by psychosocial factors like self-esteem (36). Other literature about the influence of self-esteem and (self-perceived) oral health is mainly focused on the orthodontic field and remains inconclusive (37, 38). Unfortunately, studies on the relationship between family SEP and self-esteem are scarce. Whereas self-esteem has been shown to be significantly associated with quality of life, the associations between family SEP indicators and self-esteem appear inconsistent (39, 40). Because we did not include self-esteem in our analysis, we cannot conclude about the role of it in the association between family SEP and OHRQoL. However, considering the Wilson and Cleary model of (oral health-related) quality of life (3), we highly recommend further research to understand the role of self-esteem in relationship with environmental factors and OHRQoL.

The strength of the associations between family SEP indicators and OHRQoL slightly varied. Generally, family SEP indicators are associated with each other (21, 22). This lack of independence among these variables makes it difficult to conclude which factor is most important. Maternal education and employment status were less related to children's OHRQoL. One reason for this might be that the father is still most often the principal earner of the family, which would make maternal variables less appropriate as SEP indicators. Indeed, the correlation between maternal and paternal education and employment status was fairly low (supplemental Table S3). These considerations suggest, that maternal employment status might not be used as favorable socioeconomic indicator in oral health research among populations comparable to the Generation R Cohort. The family SEP indicators directly related to material resources of the household (benefit dependency, paternal employment status and household income) were the strongest related to OHRQoL. Dental treatment and care often involves high costs. As this might indicate that oral health care is less accessible to children with lower family socioeconomic position, this finding should alert oral health care providers and policy makers.

There are several important theoretical reasons why it is important to study family SEP with children's OHRQoL. Family SEP refers to the social and economic factors influencing which position individuals have within the society (21). A low SEP has a negative influence on adult's oral health, and parental oral health in turn is a strong predictor for child oral health (41). In addition, the influences of SEP act over the life course and are therefore important to study as early as possible. Reduced child oral health is a strong predictor for impaired oral health in later life (42). Finally, all the different influences on OHRQoL help to understand why relationships between clinical status and OHRQoL are sometimes weak and inconsistent.

The social inequalities in children's OHRQoL found in the present study indicate that policies and interventions aimed to promote oral health behaviors and prevent oral disease as well as discomfort among socially deprived are highly warranted. Based on our study, these strategies should target at social disadvantage itself, rather than exclusively on potential mediating factors like oral health behavior, cultural differences or self-esteem and could involve education, social benefits for dental treatments or introducing insurance covering various kinds of dental treatments.

Certainly, some limitations of the present study need to be discussed. First, as in every observational study, our results might be affected by residual confounding, although we

have constructed fully adjusted models to assess the independent effects of different family SEP indicators. Yet, family SEP is a complex concept and we did not include all kinds of family SEP indicators, as for example wealth or neighborhood SEP indices (21,22). Second, in this study children's' OHRQoL was assessed by parental questionnaires which might have introduced information bias. We used parental reports because of practical reasons and because several studies found parents to be good proxies for children's OHRQoL (43-45). Third, another potential source of information bias in the present study might be due to the assessment of certain SEP indicators at earlier time points. Certain SEP indicators are dynamic and change over time. However, educational level for example is known to be relatively stable (21). Moreover, we found consistent associations for almost all SEP indicators with OHRQOL, which suggest that the earlier assessed data are good proxies for the current SEP. Fourth, with regard to the original sample size of the Generation R study, this study had a great number of loss to follow-up. This could have resulted in a selection bias, if the association between family SEP and OHRQoL would be different between the excluded and included study population. However, this seems unlikely. Fifth, because the Generation R Study has a generally high SES, the generalizability of our results are potentially limited. Sixth, because we analyzed many different socioeconomic indicators in this study, multiple testing might be seen as a threat to this study. However, because in our opinion testing all these different indicators fit into one single hypothesis, i.e. a consistent relationship between family SEP and OHRQoL, we did not adjust for a multiple testing problem. Last, the various family SEP indicators could be seen as mediators for the association between socioeconomic indicators and OHRQoL. Therefore parts of the effects of these indicators are not confounded but caused by other socioeconomic indicators. This is most easily explained for family composition, as single households for example are linked to lower household income or benefit dependency (21, 22, 46). We saw in our analysis that other socioeconomic indicators explain 75.0% of the association between family composition and OHRQoL. As a consequence relationships between the various socioeconomic indicators with OHRQoL may have been underestimated in model 2. Still, model 2 was not affected by multicollinearity, as tolerance and VIF values for all covariates were within the accepted range.

The advantages of this study consists of the large and ethnically diverse study population and the availability of multiple indicators of family SEP. A post-hoc statistical power calculation indicated that a sample of 910 children would have been sufficient to show the present findings with 80% statistical power (based on number of predictors = 14, $\alpha = 0.05$, lowest $R^2$ tested= 0.02) (47). This study, however, includes 3871 children. To our knowledge, this is the first study that investigates the association between family SEP and OHRQoL in such a large multiethnic cohort.

In conclusion, family SEP was consistently associated with OHRQoL, with children from low family SEP having lower OHRQoL. These association were independent of their clinical oral health status, reinforcing the importance of OHRQoL as outcome measure in oral health research. Given these disparities, interventions and policies promoting good oral health and oral wellbeing should target children from low socioeconomic position. Nevertheless, more research is needed to understand the pathways of social inequalities in children's OHRQoL.

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## Supplemental Material

**Table S1.** The 11 items of the Cohip-ortho, used in the present study

During the past 3 month, how often has your child:

1. had pain in his/her teeth/toothache
2. had crooked teeth or spaces between his/her teeth
3. had discolored teeth or spots on his/her teeth
4. had bad breath
5. had bleeding gums
6. had difficulty eating foods he/she would like to because of his/her teeth, mouth, or face
7. felt worried or anxious because of his/her teeth mouth or face
8. not wanted to speak/ read out loud in class because of his/her teeth, mouth, or face
9. been teased, bullied or called names by other children because of his/ her teeth, mouth, or face
10. felt that he/she was attractive (good looking) because of his/ her teeth, mouth, or face
11. had difficulty saying certain words because of his/her teeth or mouth

**Table S2.** Non response analysis (n = 8548)

| | | **Included** (n = 3871) | **Excluded** (n = 4677) | **p-value** |
|---|---|---|---|---|
| **Maternal education level** [a, c] | | | | |
| | low | 1267 (35.0) | 1369 (54.4) | |
| | high | 2350 (65.0) | 1148 (45.6) | < 0.001 |
| **Paternal education level** [a, c] | | | | |
| | low | 1220 (36.1) | 1140 (51.9) | |
| | high | 2162 (63.9) | 1057 (48.1) | < 0.001 |
| **Maternal employment status** [c] | | | | |
| | paid job | 2785 (80.6) | 1584 (67.6) | |
| | no paid job | 672 (19.4) | 760 (32.4) | < 0.001 |
| **Paternal employment status** [c] | | | | |
| | paid job | 3164 (95.5) | 1964 (92.0) | |
| | no paid job | 149 (4.5) | 171 (8.0) | < 0.001 |
| **Household income**[d] | | | | |
| | < 2000€ | 643 (17.9) | 372 (31.7) | |
| | > 2000€ | 2955 (82.1) | 800 (68.3) | < 0.001 |
| **Receiving benefits** [b, d] | | | | |
| | Yes | 394 (10.5) | 201 (16.3) | |
| | No | 3360 (89.5) | 1032 (83.7) | < 0.001 |
| **Family composition** [d] | | | | |
| | One parent | 553 (14.8) | 282 (23.1) | |
| | Two parents | 3192 (85.2) | 940 (76.9) | < 0.001 |
| **Ethnicity** | | | | |
| | native Dutch | 2626 (68.9) | 1938 (46.3) | |
| | non- Dutch | 1184 (31.1) | 2251 (53.7) | < 0.001 |
| **Childs sex** | | | | |
| | male | 1923 (49.7) | 2416 (51.7) | |
| | female | 1948 (50.3) | 2260 (48.3) | 0.068 |
| **Childs age** | | | | |
| | median (90% range) | 9.79 (9.48-10.47) | 9.98 (9.56-11.18) | < 0.001 |
| **Caries experience** [c] | | | | |
| | no | 2167 (75.2) | 1430 (60.7) | |
| | yes | 713 (24.8) | 925 (39.3) | < 0.001 |
| **Orthodontic need** [d] | | | | |
| | no | 1902 (62.4) | 985 (60.2) | |
| | yes | 1146 (37.6) | 652 (39.8) | 0.139 |
| **Aesthetic need** | | | | |
| | no | 1691 (57.4) | 908 (57.5) | |
| | borderline | 1006 (34.2) | 535 (33.9) | |
| | yes | 247 (8.4) | 136 (8.6) | 0.958 |
| **Self-perceived orthodontic need** [d] | | | | |
| | no | 1075 (27.9) | 1 (9.1) | |
| | borderline | 980 (25.5) | 2 (18.2) | |
| | yes | 1794 (46.6) | 8 (72.7) | 0.199 |

Data are presented as absolute numbers with percentages for categorical data and as median with 90% range for continuous data. P-values are based on chi-square tests and Mann-Whitney-U tests. [a] educational level: low = no education, primary school, vocational training, general secondary school and first year higher vocational training, high = higher vocational training, university or PhD degree; [b] benefits : social security, unemployment benefits, disability allowances and other; [c] assessed at children's age of 6 ; [d] assessed at children's age of 10.

**Table S3.** Correlations between family SEP indicators (n=3796)

| | Maternal education | Paternal education | Maternal employment | Paternal employment | Income | Benefits | Single parenting |
|---|---|---|---|---|---|---|---|
| **Maternal education** | 1** | 0.49** | 0.25** | 0.10** | 0.33** | 0.18** | 0.12** |
| **Paternal education** | - | 1 | 0.12** | 0.14** | 0.34** | 0.17** | 0.18** |
| **Maternal employment** | - | - | 1 | 0.15** | 0.29** | 0.25** | 0.08** |
| **Paternal employment** | - | - | - | 1 | 0.28** | 0.24** | 0.16** |
| **Income** | - | - | - | - | 1 | 0.43** | 0.54** |
| **Benefits** | - | - | - | - | - | 1 | 0.29** |
| **Single parenting** | - | - | - | - | - | - | 1 |

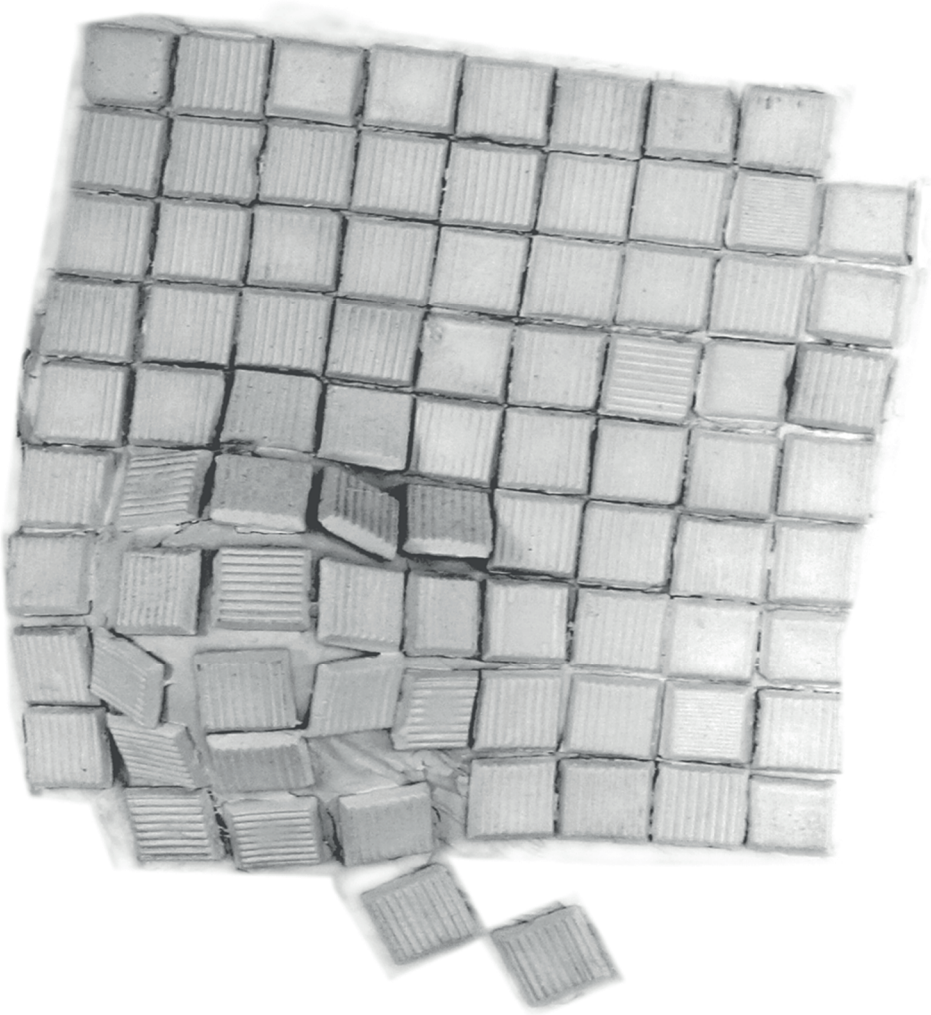
** correlation is significant at the 0.01 level (2-tailed); The table is based on the imputed dataset.

**Table S4.** Test for multicollinearity in model 2[1] based on original data

| | Collinearity Statistics | |
|---|---|---|
| **Covariates** | **Tolerance** | **VIF** |
| ***SEP indicators*** | | |
| Maternal education | 0.781 | 1.281 |
| Paternal education | 0.770 | 1.299 |
| Maternal unemployment | 0.923 | 1.084 |
| Paternal unemployment | 0.951 | 1.051 |
| Household income | 0.701 | 1.426 |
| Receiving benefits | 0.933 | 1.072 |
| Single parenting | 0.797 | 1.255 |
| ***Covariates*** | | |
| Caries experience | 0.958 | 1.044 |
| IOTN-DHC | 0.718 | 1.392 |
| IOTN-AC | 0.717 | 1.395 |
| Self-perceived orthodontic need | 0.832 | 1.203 |
| Gender | 0.985 | 1.016 |
| Age | 0.990 | 1.010 |
| Ethnicity | 0.918 | 1.090 |

[1]Model 2 investigates the Associations of all socioeconomic indicators with OHRQOL at children's age of 10

# Chapter 5
# Characteristics of the individual influencing OHRQoL



# Chapter 5.1

## Associations between objective and subjective orthodontic treatment need related to children's oral health-related quality of life:Comparison between ethnic groups

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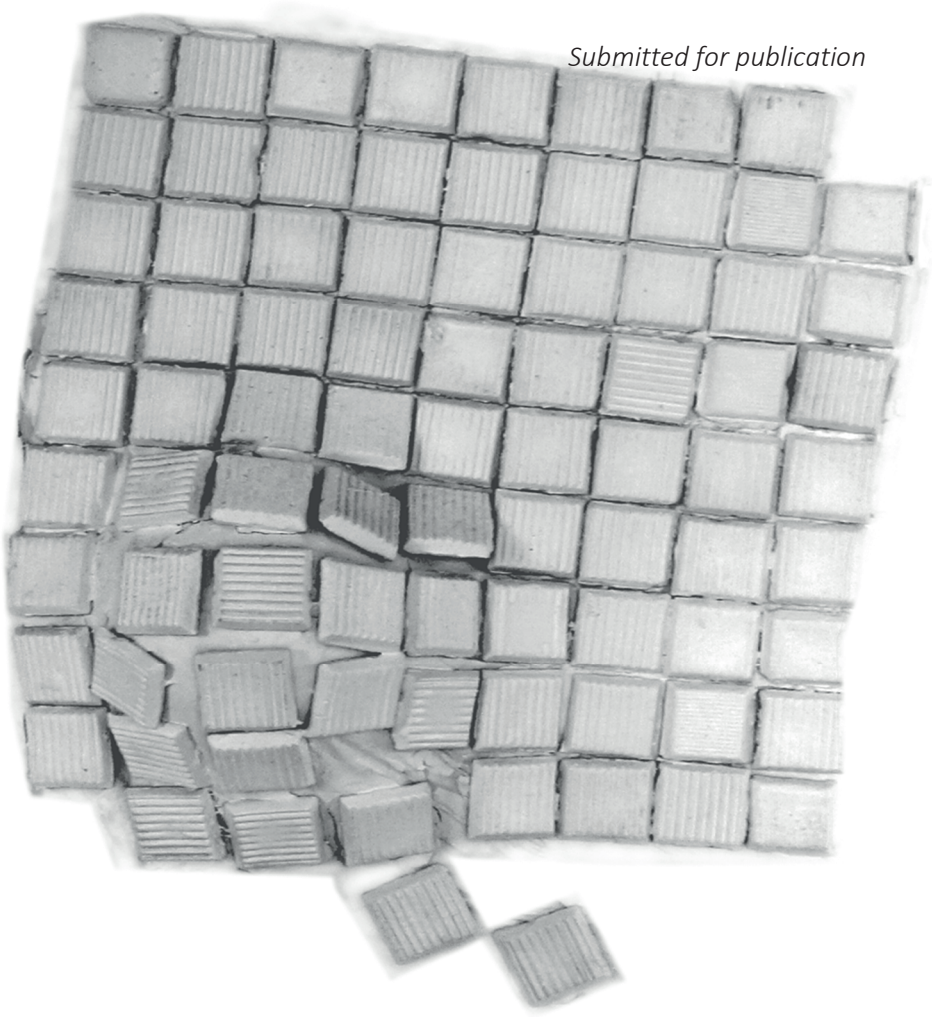
*Submitted for publication*



Abstract

***Introduction*** Ethnic minorities are at a disadvantage with regard to various oral health outcomes. We investigated ethnic differences in objective orthodontic need (OOT) and subjective orthodontic need (SOT), how this differences influence oral health -related quality of life (OHRQoL) and whether these varied among ethnic groups.

***Methods*** This cross-sectional study among n= 2419 9-year old children was part of the Generation R Study, a multi-ethnic cohort study conducted in Rotterdam, The Netherlands. OOT was assessed with the Index of Orthodontic Treatment Need. SOT and OHRQoL were assessed in parental questionnaires. Series of multinomial logistic regression models and weighted least square linear regression models were used to analyze associations between OHRQoL, OOT and SOT.

**Results** Compared to native Dutch children, OHRQoL was significantly lower in children with other ethnic backgrounds. SOT was similar among Dutch and Surinamese children (48.6%, 47.1% resp.), but lower among Turkish (35.8%) and higher among Moroccan (51.8%) children. There were no ethnic differences in OOT. Compared to the Dutch, associations between OOT and SOT were much weaker (or absent) in Turkish, Moroccan and Surinamese children, OOT and SOT tended to influence OHRQoL slightly stronger the ethnic minority groups.

***Conclusion*** Ethnic background explained some of the variability in the association between OOT and SOT, OHRQoL however contributed less to this variability.

*Clinical relevance* This research supports orthodontists in their understanding of and communication with their patients as it highlights the role of (patient-reported) social and individual factors in the assessment of orthodontic treatment need.

**Keywords:** malocclusion, children, quality-of-life, self-perception, ethnic differences

## Introduction

Ethnic minority groups are at a disadvantage with regard to various oral health outcomes. Dental caries, periodontal disease, edentulism and oral cancer are strongly associated with ethnic background (1, 2). These disparities are not only evident in adults, but already exist during childhood. Not only the prevalences of caries but also orthodontic treatment need and dental traumas are reported to vary among children from different ethnic groups (3-6). Generally, children from ethnic minority groups have higher odds for unmet dental care need (7).

Various reasons underlying ethnic differences in oral health have been proposed in the literature. These factors were directly related to oral health beliefs. First and second generation immigrant's brush their teeth more infrequently, have less often a dental insurance and make less use of dental services (8,9). Furthermore, their dental knowledge and dental attitudes appear to be low (10). In addition, there were factors which are rather indirectly related to oral health. For example, ethnic minority groups tend to have higher sugar intake, more frequent eating moments and cultural practices that promote mother to child transmissions of cariogenic bacteria (11-13). Finally, the association between ethnicity and oral health might partly be mediated by socioeconomic status (SES) like low education and lower household income (1, 14).

Based on these reasons not only objectively assessed oral health might be different among ethnic groups, but in particular self-perceived oral health, treatment need and also oral health-related quality of life (OHRQoL) is expected to be different. Especially in the dental subspecialty of orthodontics, where self-perception and aesthetics have a major role, this might become evident. Moreover, ethnicity might explain some of the highly variable association between self-perception of and orthodontist's assessment of treatment need (15). It has been shown, that not only the perception of body image but also how it is evaluated and how related self-esteem develops, differs among ethnic groups (16-18). Therefore, ethnicity might play an important role in the decisions to undergo orthodontic treatment. However, research on ethnic differences in objective orthodontic treatment need (OOT) and subjective orthodontic treatment need (SOT) is very limited.

We investigated in a multi-ethnic population-based cohort study among 2419 school-aged children in the Netherlands, the ethnic differences in the association between OOT and SOT. Furthermore, we explored how different associations between OOT and SOT influence OHRQoL and whether this varied among the different ethnic groups.

## Material and Methods

### *Study design*

This study was part of the Generation R study, a population based multi-ethnic prospective cohort study from fetal life onwards conducted in Rotterdam, The Netherlands. The Generation R Study has been described in detail elsewhere (19). This study was conducted in accordance with the guidelines proposed in the World Medical Association's Declaration of Helsinki and has been approved by the Medical Ethical Committee at the Erasmus University Medical Centre, Rotterdam, The Netherlands (MEC-2012-165). All participants gave written informed consent before any measurement started.

*Study population*

Invitations to participate in the Generation R study were made to all pregnant women in the study area between April 2002 and January 2006. In total, 9778 mothers participated in the first phase of the study. The present study is conducted in the follow-up phase from children's age of 9 years onwards, in which mothers of n = 7393 children participated. For the purpose of the present study we selected children of which information on subjective orthodontic treatment need was available (n = 3783) and which represented the largest ethnic groups (n > 100) in the study population (n = 3048). For the analysis on the association between OOT and SOT we excluded children with missing information on OOT (n = 629). Thus, the final population of this analysis consisted of 2419 subjects: 2036 native Dutch children, 135 Turkish children, 99 Moroccan children and 149 Surinamese children. For the second analysis on the association between OOT and SOT agreement related to OHRQoL, we excluded children with missing information on OHRQoL (n = 7). Thus, the final population of this analysis consisted of 2412 subjects: 2031 native Dutch children, 134 Turkish children, 99 Moroccan children and 148 Surinamese children. In figure 1 the flow chart of both participant selections is presented.

*Ethnic groups*

All children participating in the Generation R Study were born in Rotterdam, the Netherlands. Children's ethnicity was classified based on the ethnic background of their parents (20). Children with parents born in the Netherlands were classified as Dutch. If one of the parents was born in another country, the child was classified as non-Dutch. If the parents were born in different countries, maternal ethnic background defined children's ethnicity, because mothers are most often the primary caregivers and our aim was to take the cultural background of the mothers into account. Maternal ethnic background was assessed by questionnaire at enrolment in the Generation R study and based on the country of birth of the mothers' parents. If both parents of the mother were born abroad, the country of birth of the mothers' mother decided on maternal ethnic background.

*Assessment of OHRQoL, subjective and objective orthodontic treatment need and OHRQoL*

OHRQoL was assessed with a short form of the Child Oral Health Impact Profile (COHIP-ortho) (21). The COHIP-ortho is a questionnaire for parents that measures OHRQoL of the child with 11 questions, covering the different domains of oral health, including social-emotional wellbeing, functional wellbeing, school and peer interaction (Appendix TS1). These questions were answered on a five-point Likert scale. The answers to the questions were given a score from 1 to 5, which were finally summed up for every individual. The total overall score of the COHIP-ortho ranges from 0 to 55 and higher scores correspond to better OHRQoL. Missing values in the responses to the OHRQoL questionnaire (COHIP-ortho) were replaced by the personal mean score of the remaining answers to the questions, as it is proposed by other researchers that used the original version of the COHIP (22). If there were more than 30% of the answers missing, the participant was excluded from the analysis.

SOT was assessed in questionnaires with the question:"Do you think your child needs braces?". In the Netherlands, orthodontic treatment generally starts after the age of

nine, thus the parents might have dealt with the question already to some extent. The response to the question was given by the mothers on a five-point Likert scale from strongly disagree to strongly agree. For the first data analysis following categories for SOT were made: 'No SOT' for children that strongly or somewhat disagreed with the statement. 'Borderline SOT' for children that did not agree but also did not disagree with the statement and 'Definite SOT' for children that somewhat or strongly agreed with the statement. For the second analysis, SOT was categorized into no need ('No SOT' and 'Borderline SOT') versus need ('Definite SOT')

Objective orthodontic treatment need (OOT) was assessed with the Index of orthodontic treatment need (IOTN) from photographic and radiographic records as described elsewhere (23). There are two IOTN components: the Aesthetic Component (AC) and the Dental Health Component (DHC) (24). The IOTN-DHC recognizes five grades of orthodontic treatment need, ranging from no need (1) to very great need (5). For the present study the two lowest categories (1 and 2) were grouped together into little need. For the second analysis the IOTN-DHC was categorized into no need (1, 2, 3) versus need (4, 5). The IOTN-AC component knows a severity range from 1 to 10. As recommended, in this study grade 1 to 4 were considered as no need, 4- 6 were considered borderline need and 7-10 were considered definite aesthetic orthodontic treatment need.

*Covariates*

We considered child's sex and age and family socioeconomic position as potential confounders for the associations between OOT, SOT and OHRQoL among the different ethnic groups. Family socioeconomic position was measured by maternal and paternal education level defined as low for no education finished or primary and secondary phase 1 or 2 finished and high for higher vocational training finished or university or PhD degree finished, by family employment status defined as receiving any benefits or not and by household income (< 2000, 2000€-3200€, > 3200€).

*Statistical analysis*

We used descriptive statistics to characterize the study population. Family socioeconomic factors, child's sex, SOT and OOT were evaluated by ethnic background with the chi-square statistic. Child's age and OHRQoL were evaluated by ethnic background with a one-way ANOVA and the Kruskal-Wallis test.

The association between objective and SOT was investigated with a series of multinomial logistic regression models for each ethnic group. First a basic model was constructed adjusted for child's sex and age only. Model 1 was additionally adjusted for socioeconomic factors, i.e. parental education, employment status and household income. Finally, model 2 was additionally adjusted for the IOTN-AC. Whether the association between OOT and SOT was modified by ethnicity, and thus whether the associations were significantly different among the different ethnic groups was examined by adding an interaction term between ethnic background and OOT to the basic model. The interaction term was considered significant at a p-value $\leq 0.05$.

Series of weighted least square linear regression models were built to analyze the associations between agreement of SOT with OOT and OHRQoL in each ethnic group. The

variable for agreement of SOT with OOT was constructed via a 2x2 table (Appendix table TS4) and had 4 categories: “no SOT and no OOT” (reference), “no SOT but OOT”, “SOT, but no OOT”, “both SOT and OOT”. In the regression models agreement between SOT and OOT was used as independent variable, OHRQoL was the dependent outcome variable. Again, the basic models were adjusted for child’s sex and age only. In the first model we additionally adjusted for socioeconomic factors, i.e. parental education, employment status and income. The second model was additionally adjusted for aesthetic OOT based on the IOTN-AC. Afterwards the linear regression coefficients of the Turkish, the Moroccan and the Surinamese groups were compared with the ones from the Dutch groups by a test for heterogeneity. A difference in effect estimate was considered to be significant at a p-value ≤ 0.05.

Because of the missing data in the potential confounders, we applied a multiple imputation method. Ten imputed data sets were generated using the Markov Chain Monte Carlo (MCMC) method. Imputations were based on the relationships between covariates, determinants and outcomes, but we did not impute missing determinants (SOT, OOT) or outcomes (SOT, OHRQoL). We present the pooled estimates of these data sets as regression coefficients with 95% confidence intervals (β(95% CI)) or as odds ratios and their 95% confidence intervals (OR(95% CI)). All analyses were conducted with the Statistical Package for Social Sciences (SPSS) version 20.0 for windows (SPSS, Inc, Chicago, IL). Generally, we used for all analysis a significance level of $p < 0.05$.

## Results

### *Sample characteristics*

Table 1 shows the family and child characteristics for the different ethnic groups. SOT prevalence among Dutch children was 48.6%. A similar prevalence was observed among Surinamese children (47.1%), whereas a lower prevalence of SOT was observed among Turkish (35.8%) and a higher prevalence among Moroccan (51.8%) children. OHRQoL was relatively high among Dutch children with a median (90% range) of 50.0 (43.0-53.0) and significantly lower in the Turkish (49.0; 38.2-53.0), Moroccan (49.0; 41.8-53.0) and Surinamese children (49.0; 41.0-53.0). Prevalence of OOT based on the IOTN-DHC and IOTN-AC among the Dutch children was 29.5% and 6.2%, respectively, but were not significantly different among Turkish (32.7% and 3.0%), Moroccan (34.2% and 7.9%) and Surinamese children (29.3% and 9.4%). In the total study population, 974 children (40.3%) had “no SOT and no OOT”, 279 children (11.5%) had “no SOT but OOT”, 534 children (22.1%) had “SOT but no OOT” and 632 children (26.1%) had “SOT and OOT” (Appendix table S4).

### *Objective and subjective orthodontic treatment need in different ethnic groups*

Table 2 shows the association between OOT and SOT in the different ethnic groups. Among the Dutch children, having OOT was strongly associated with having borderline SOT (aOR = 1.47 (95%CI: 1.05 - 2.05)) and definite SOT (aOR = 3.82 (95%CI: 2.78 - 5.25)). The association between OOT and SOT were much weaker or absent in the Turkish, Moroccan and Surinamese children. However, the association between OOT and SOT

was only in the Moroccan children significantly different than in Dutch children (p-value = 0.004).

**Table 1.** Sample characteristics by ethnic background (n = 2419)

| | Dutch | Turkish | Moroccan | Surinamese | |
|---|---|---|---|---|---|
| | n = 2036 | n = 135 | n = 99 | n = 149 | p-value |
| *Family characteristics* | | | | | |
| **Maternal education level (%)** | | | | | |
| low | 549 (27.0) | 87 (64.4) | 57 (57.6) | 83 (55.7) | |
| high | 1403 (68.9) | 35 (25.9) | 21 (21.2) | 54 (36.2) | < 0.001 |
| *Missing* (%) | *84 (4.1)* | *13 (9.6)* | *21 (21.2)* | *12 (8.1)* | |
| **Paternal education level (%)** | | | | | |
| low | 564 (27.7) | 71 (52.6) | 56 (56.6) | 76 (51.0) | |
| high | 1325 (65.0) | 36 (26.6) | 17 (17.2) | 41 (27.5) | < 0.001 |
| *Missing* (%) | *147(7.2)* | *28 (20.7)* | *26 (26.3)* | *32 (21.5)* | |
| **Employment status (%)** | | | | | |
| receiving benefits | 1863 (91.5) | 91 (67.4) | 63 (63.6) | 123 (82.6) | |
| receiving no benefits | 140 (6.9) | 34 (25.2) | 30 (30.3) | 23 (15.4) | < 0.001 |
| *Missing* (%) | *33 (1.6)* | *10 (7.4)* | *6 (6.1)* | *3 (2.0)* | |
| **Household income (%)** | | | | | |
| < 2000€ | 171 (8.4) | 55 (40.7) | 50 (50.5) | 50 (33.6) | |
| 2000€-3200€ | 546 (26.8) | 54 (40.0) | 29 (29.3) | 52 (34.9) | |
| > 3200€ | 1205 (59.2) | 12 (8.9) | 9 ( 9.0) | 37 (24.8) | < 0.001 |
| *Missing* (%) | *114 (5.6)* | *14 (10.4)* | *11 (11.1)* | *10 (6.7)* | |
| *Child Characteristics* | | | | | |
| **Sex (%)** | | | | | |
| boys | 1015 (49.9) | 72 (53.3) | 53 (53.5) | 79 (53.0) | |
| girls | 1021 (50.1) | 63 (46.7) | 46 (46.5) | 70 (47.0) | 0.681 |
| **Age** | | | | | |
| Mean (SD) | 9.84 (0.34) | 9.95 (0.47) | 9.92 (0.35) | 9.88 (0.42) | < 0.001 |
| **IOTN -DHC (%)** | | | | | |
| little | 759 (37.3) | 55 (40.7) | 34 (34.3) | 55 (36.9) | |
| moderate | 515 (25.3) | 27 (20.0) | 26 (26.3) | 37 (24.8) | |
| great | 596 (29.3) | 43 (31.9) | 25 (25.3) | 40 (26.8) | |
| very great | 166 (8.2) | 10 (7.4) | 14 (14.1) | 17 (11.4) | 0.446 |
| **IOTN -AC (%)** | | | | | |
| no need | 1126 (55.3) | 81 (60.0) | 57 (57.6) | 81 (54.4) | |
| borderline need | 676 (33.2) | 46 (34.1) | 27 (27.3) | 45 (30.2) | |
| definite need | 162 (8.0) | 5 (3.7) | 9 (9.0) | 18 (12.1) | 0.221 |
| *Missing* (%) | *72 (3.5)* | *30 (22.2)* | *6 (6.1)* | *5 (3.4)* | |
| **Perceived orthodontic need (%)** | | | | | |
| no need | 507 (24.9) | 57 (42.2) | 33 (33.3) | 40 (26.8) | |
| borderline need | 536 (26.3) | 27 (20.0) | 16 (16.2) | 37 (24.8) | |
| definite need | 993 (48.8) | 51 (37.8) | 50 (50.5) | 72 (48.3) | < 0.001 |
| **OHRQoL** | | | | | |
| Median (90% range) | 50.0 (43.0 - 53.0) | 49.0 (38.8 - 53.0) | 49.0 (42.0 - 53.0) | 49.0 (41.0 - 53.0) | 0.013 |
| *Missing (%)* | *5 (0.3)* | *1 (0.7)* | *0 (0.0)* | *1 (0.7)* | |

Based on the original data set. Numbers are presented as absolute numbers for categorical variables or as mean (SD) for normally distributed continuous variables or as median (90%range) for non-normally distributed continuous data. P-values are estimated based on chi-square tests, one-way ANOVAs and Kruskal-Wallis tests.

**Table 2.** The association between OOT and SOT among different ethnic groups (n = 2419)

| | Subjective orthodontic treatment need (OR (95%CI)) | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **Objective orthodontic treatment** | **Dutch** n = 2036 | | **Turkish** n = 135 | | **Moroccan** n = 99 | | **Surinamese** n = 149 | |
| **Basic model** | borderline | definite | borderline | definite | borderline | definite | borderline | definite |
| no need | ref | ref | ref | ref | ref | ref | ref | ref |
| need | 1.66 (1.23 - 2.25) | 6.02 (4.62 - 7.85) | 0.96 (0.35 - 2.64) | 2.87 (1.30 - 6.35) | 0.54 (0.14 - 2.09) | 1.58 (0.63 - 3.96) | 1.00 (0.35 - 2.85) | 3.33 (1.40- 7.91) |
| **Model 1** | | | | | | | | |
| no need | ref | ref | ref | ref | ref | ref | ref | ref |
| need | 1.67 (1.23 - 2.26) | 6.16 (4.72 - 8.04) | 0.98 (0.34 - 2.80) | 2.78 (1.23 - 6.31) | 0.46 (0.11 - 1.91) | 1.37 (0.52 - 3.62) | 0.84 (0.28 - 2.55) | 2.97 (1.20 - 7.34) |
| **Model 2** | | | | | | | | |
| no need | ref | ref | ref | ref | ref | ref | ref | ref |
| need | 1.46 (1.05 - 2.05) | 3.82 (2.80 - 5.21) | 0.99 (0.33 - 2.95) | 2.30 (0.94 - 5.64) | 0.36 (0.07 - 1.90) | 0.98 (0.34 - 2.83) | 0.65 (0.16 - 2.73) | 1.58 (0.50 - 5.09) |
| ***p-value[a]*** | | | *0.337* | *0.083* | *0.136* | *0.004* | *0.327* | *0.202* |

Data are presented as odds ratios with 95% confidence interval. The basic model is adjusted for age and gender of the child, model 1 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 2 is additionally adjusted for orthodontic treatment need based on the IOTN-AC. [a] p-value for the difference with the Dutch group obtained from interaction term in the model. OOT= Objective orthodontic treatment need, SOT = Subjective orthodontic treatment need.

### *Nonresponse analysis*

Children with missing data on OOT and SOT at the age of 9 years were compared with children who did not have missing data on subjective orthodontic treatment or OHRQoL. Differences were present in all family characteristics and child characteristics, except child's sex and objective orthodontic treatment need (Appendix TS5).

## Discussion

### *Answer to the research question*

OOT measured with the IOTN was similar among children with different ethnic background, but SOT and OHRQoL appeared to be influenced by ethnic background. The association between OOT and SOT was strongly present in Dutch, but much less in children from other ethnic backgrounds. Furthermore, the association between OOT and SOT agreement with OHRQoL were not significantly different among the different ethnic groups. Thus, ethnic background explained some of the variability in the association between SOT and OOT, but OHRQoL contributed less to this variability.

**Table 3.** The association between SOT and OOT agreement with OHRQoL among different ethnic groups (n = 2412)

| | Oral health-related quality of life (ß (95%CI)) | | | | | | |
|---|---|---|---|---|---|---|---|
| **Subjective & objective need** | **Dutch** n = 2031 | **Turkish** n = 134 | | **Moroccan** n = 99 | | **Surinamese** n = 148 | |
| **Basic model** | | | *P value*[a] | | *P value*[a] | | *P value*[a] |
| No SOT and no OOT | ref | ref | | ref | | ref | |
| No SOT, but OOT | -0.46 (-0.84 - -0.07) | 0.04 (-1.30 - 1.37) | *0.48* | -1.24 (-3.67 - 1.19) | *0.53* | 0.28 (-1.61 - 2.17) | *0.45* |
| SOT, but no OOT | -1.14 (-1.47 - -0.81) | -0.95 (-2.72 - 0.82) | *0.84* | 0.06 (-1.47 - 1.60) | *0.13* | -1.01 (-2.65 - 0.64) | *0.88* |
| SOT and OOT | -1.94 (-2.29 - -1.59 ) | -3.43 (-5.79 - -1.06) | *0.09* | -2.09 (-3.73 - -0.46) | *0.86* | -1.47 (-3.09 - 0.16) | *0.58* |
| **Model 1** | | | | | | | |
| No SOT, but OOT | -0.37 (-0.75 - 0.06) | 0.04 (-1.22 - 1.31) | *0.54* | -1.20 (-3.64 - 1.24) | *0.51* | 0.55 (-1.15 - 2.24) | *0.30* |
| SOT, but no OOT | -1.15 (-1.47 - -0.82) | -1.17 (-2.90 - 0.56) | *0.98* | 0.12 (-1.36 - 1.59) | *0.10* | -1.04 (-2.60 - 0.52) | *0.89* |
| SOT and OOT | -1.87 (-2.21 - -1.53) | -3.03 (-5.27 - -0.80) | *0.31* | -2.22 (-3.86 - -0.58) | *0.68* | -1.12 (-2.68 - 0.44) | *0.35* |
| **Model 2** | | | | | | | |
| No SOT, but OOT | -0.25 (-0.63 - 0.13) | 0.24 (-1.20 - 1.67) | *0.51* | -1.05 (-3.54 - 1.44) | *0.53* | 0.25 (-1.65 - 2.16) | *0.61* |
| SOT, but no OOT | -1.11 (-1.44 - -0.79) | -1.24 (-2.96 - 0.48) | *0.88* | 0.17 (-1.32 - 1.65) | *0.10* | -1.15 (-2.72 - 0.43) | *0.96* |
| SOT and OOT | -1.47 (-1.86 - -1.09) | -3.01 (-5.39 - -0.63) | *0.21* | -1.88 (-3.62 - -0.14) | *0.65* | -1.27 (-3.20 - 0.65) | *0.84* |

Data are presented as weighted linear regression coefficients with 95% confidence interval. The basic model is adjusted for age and gender of the child, model 1 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 2 is additionally adjusted for orthodontic treatment need based on the IOTN-AC. [a]p-value for a significance of difference with the Dutch group obtained from test for heterogeneity. SOT = Subjective orthodontic treatment need, OOT = objective orthodontic treatment need, OHRQoL = oral health related quality of life.

### *The relationship between objective and subjective orthodontic treatment need*

Similar to previous studies, we found that the prevalence of SOT was higher than the prevalence of OOT among all children (25-28). This finding suggests, that patients assess their need differently than professionals do. In addition, we found that OOT was not different among ethnic groups, although SOT was different among ethnic groups. This finding shows that patients not only perceive different reasons for SOT than professionals, but also that among patients the importance of certain reasons might be differently perceived. In the present study, this became evident in the association between OOT and SOT, which was only strong among Dutch children. These differences in the association between OOT and SOT origin from individual patient characteristics. Based on our

study, but also suggested by other research, one of these patient characteristics is their ethnic background. In line with our research, Christopherson et al showed ethnic differences in OOT and SOT, where black children wanted braces more and were less satisfied with their smile than white children, although they had less OOT (6).
Obviously not only the severity degree of malocclusions affects subjective need, but also the location of the deviation. Marques et al showed that upper anterior crowding and parents perception mainly determined SOT (28). We did not investigate in detail, whether the different ethnic groups differed in kind of malocclusion. But as the children do not differ in the IOTN-AC, it seems unlikely, that Dutch children suffered more form upper anterior crowding and overjets than from crossbites or deep bites compared to Turkish and Moroccan children.

*The association between subjective orthodontic treatment need and objective orthodontic treatment need with OHRQoL*

Compared to Dutch children, the Turkish, Moroccan and Surinamese children had less subjective orthodontic treatment need and generally lower OHRQoL than Dutch children. Different agreement of SOT and OOT influenced OHRQoL in different dimensions. However, this was not significantly different among ethnic groups. Comparable research about differences in SOT, OOT and OHRQoL is very limited.
We saw that having "SOT and OOT" affected OHRQoL of Turkish children more than other ethnic groups. Moreover, SOT influenced OHRQoL of Turkish children the most (Appendix table S7). This might reflect the barrier which children with an ethnic minority background perceive with regard to the provision of orthodontic and other dental care. On the one hand, a Dutch child doubting its orthodontic treatment need simply goes to the dentist or orthodontist and receives a professional advice. A Turkish child might not get to address the problem promptly, because of financial reasons, less parental support, less availability of dental healthcare providers or any other reason. Thus, Turkish children, especially with minor malocclusions, might feel less supported and have to deal for a longer time with uncertainty about their orthodontic need. On the other hand, children from ethnic minorities might receive orthodontic treatment indeed with more difficulties, when their oral hygiene is more often insufficient as indicated in various studies (8, 9). Orthodontists generally do not start a treatment, if oral hygiene has a higher priority to be addressed. This in turn influences their sense of coherence and dental coping beliefs, which both have been shown in other studies to be strongly associated with OHRQoL (29, 30). Care providers have to inspect thoroughly patient's benefits following orthodontic treatment from several perspectives more than occlusal discrepancy or functional disability. Although SOT affected OHRQoL among all ethnic groups, only Moroccan children tended to perceive a stronger impact of "OOT without SOT" than of "SOT without OOT" on OHRQoL. Possibly, Moroccan children are less aware of their dental needs, which would reflect the limited dental knowledge and lower dental attitudes among this ethnic minority groups (10).
Ethnic disparities in oral health and dental care have widely been investigated. As OHRQoL describes subjective and multidimensional oral health, encompassing the physical and psychological health state as well as the functioning in social interaction, the concept is very suitable to measure the perceived oral health of individuals (31). Also in

the present study, OOT lowered OHRQoL more in Turkish and Moroccan children than in Dutch children (Appendix TS6). Therefore, we consider the lowered OHRQoL among children with ethnic backgrounds in line with other research showing ethnic differences in dental insurances and provision of needed dental care (7). We showed additionally that these differences are actually perceived by individuals and have impact on their quality of life.

*Considerations to explain ethnic differences*

Next to clinical factors, other reasons influencing SOT are supposed in the literature. These factors are diverse and include severity of malocclusion, dissatisfaction with dental appearance, self-esteem, dental referrals, parental concern, social norms, gender, age, and intellectual level (15, 7, 32). The influence of these factors depend on the cultural and social characteristics of the individual and thus might correlate with the ethnic differences in OHRQoL, OOT and SOT. Ethnicity as we defined and used in this study is a complex concept which covers elements of culture, religion and migration history (33). We already discussed the clinical as well as environmental factors. Besides we adjusted our analysis for gender, age, social class and intellectual level, thus the influence on our results of these more environmental variables should be limited. The cultural factors, as (dis-)satisfaction with dental appearance and social norms, remain as theoretical explanation for the ethnic differences we found in this study.

The literature on ethnic differences in dissatisfaction with dental appearance is very limited. However, dissatisfaction with dental appearance might be related to self-esteem which is a recurring topic in orthodontic research as well as in research on ethnic differences. The relationship between self-esteem, orthodontic treatment need and OHRQoL has been investigated by several studies. It has been shown that self-esteem and OHRQoL are positively related (34, 35), however there seems no relation between OOT and self-esteem (35, 36). The differences in self-esteem among ethnicities is less ambiguously described. For example, Caucasians and Hispanics show worse body image or lower and less stable self-esteem than African and Asians (16, 18). Further research is needed to evaluate how complex ethnic differences in self-esteem are related to the associations between OOT, SOT and OHRQoL.

To our knowledge, no studies investigated social norms regarding orthodontics among different ethnic groups. However several studies indicate that ethnic minority groups have poorer dental knowledge, lower dental attitude and make less use of preventive dental care (13, 10, 12). We can only speculate that Dutch children visit the orthodontist earlier because they go more often to the dentist who refers them, and therefore they have more subjective treatment need in relation to OOT than the other ethnic groups in our sample. Research on the determinants of ethnic differences in the associations between OOT, SOT and OHRQoL is strongly recommended, because it would also further elucidate the determinants of the highly variable association between SOT and OOT.

*Strength & limitations*

Many studies up to now showed that there is a relationship between treatment need and treatment demand. Although this relationship is not one-to-one, the differences in

association are rarely addressed. As one of the first, we were able to show in a large multi-ethnic population-based cohort study among n=2419 children that parts of the variability between OOT and SOT can be explained by ethnicity.
Still our results have to be seen in the light of some limitations. Information bias might have affected the results of our study, because instead of asking the children in this study, the parents were asked to provide the information on SOT and OHRQoL. This approach was based on several studies that found parents to be good proxies for children's OHRQoL (37-39). Parents neither over- nor underestimate their children's SOT systematically (15). The non-response analysis showed that data on SOT and OHRQoL were more often missing in children from low socioeconomic position. Selection bias might have affected our results, when the association between OOT, SOT and OHRQoL differed between participants and non-participants. However, this seems unlikely. Although we adjusted for several confounders, our analysis might be affected from residual confounding. Especially, OHRQoL might be influenced by other oral diseases than malocclusion, and we were not able to correct our analysis for this, because these data were not available. Finally, we only included the major ethnic groups of our study population into our analysis, because of sample size reasons. Therefore we cannot make any conclusions on other ethnic minority groups, which are also represented in the general population. In addition, the Surinamese group in our study was heterogeneous, as they included Surinamese-Hindustani originating from India and Surinamese-Creoles originating from Africa. We did not specify these subgroups, because of the small sample size. Still the sample size of our ethnic groups was much smaller than the sample size of the control group, the Dutch children. However, as the ethnic groups are still representative this might only has affected the precision of our results and might reflect why some of our results were not significant.

*Implication for clinical practice and future research*
Malocclusion and OOT can be considered a public health problem as it is common in a large proportion of the population. In the Netherlands as in other western countries, around 60% of young adults receive any kind of orthodontic treatment (40, 41). Yet, nothing is known about the composition of this group with regard to their ethnic background. As there were no differences among ethnic groups in OOT, but absolutely in how they perceive their need, knowledge on the provision of treatment to all ethnic groups is especially important to prevent inequalities in dental and particular in orthodontic care.
In addition, this study adds to the understanding of the role of psychosocial factors in orthodontic treatment need. Our results support the suggestion of other studies to use patient reported measures complementary to clinical diagnosis in a higher degree than it is common today (42, 43).
Last, this study highlights the importance to research ethnic disparities in oral health care. Children from ethnic minorities seem to have generally lower OHRQoL than native Dutch children. As children with a compromised start to oral health are much more likely to follow a trajectory which will lead to poor oral health in adulthood, studies focusing on determinants of ethnic disparities in children's oral health are highly rec-

ommended. In this way effective preventive strategies can be developed to raise awareness among care providers and policy makers.

### *Conclusion*

In summary, whereas there were no differences in OOT between native Dutch children and children from ethnic minority groups, the associations among OOT, SOT and OHRQoL differed. Because Turkish, Moroccan and Surinamese children already tend to have generally lower OHRQoL compared to native Dutch children, the ethnic differences in the association between OOT, SOT and OHRQoL need more attention in clinical practice and oral health research.

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## Supplemental Material

**Table S1.** The 11 items of the COHIP-ortho, used in the present study

During the past 3 month, how often has your child:

1. had pain in his/her teeth/toothache
2. had crooked teeth or spaces between his/her teeth
3. had discolored teeth or spots on his/her teeth
4. had bad breath
5. had bleeding gums
6. had difficulty eating foods he/she would like to because of his/her teeth, mouth, or face
7. felt worried or anxious because of his/her teeth mouth or face
8. not wanted to speak/ read out loud in class because of his/her teeth, mouth, or face
9. been teased, bullied or called names by other children because of his/ her teeth, mouth, or face
10. felt that he/she was attractive (good looking) because of his/ her teeth, mouth, or face
11. had difficulty saying certain words because of his/her teeth or mouth

**Table S2.** The association between ethnicity and subjective orthodontic treatment need

| | Basic model | | | Model 1 | | | Model 2 | |
|---|---|---|---|---|---|---|---|---|
| | borderline | definite | | borderline | definite | | borderline | definite |
| **Dutch** | ref | | Ref | | | ref | | |
| **Turkish** | ref | 0.42 (0.27 - 0.66) | 0.41 (0.28 - 0.58) | ref | 0.46 (0.29 - 0.73) | 0.46 (0.31 - 0.67) | ref | 0.46 (0.29 - 0.73) | 0.46 (0.31 - 0.70) |
| **Moroccan** | ref | 0.46 (0.26 - 0.82) | 0.81 (0.53 - 1.24) | ref | 0.50 (0.27 - 0.91) | 0.94 (0.60 - 1.47) | ref | 0.49 (0.27 - 0.91) | 0.99 (0.61 - 1.59) |
| **Surinamese** | ref | 0.80 (0.53 - 1.21) | 0.85 (0.60 - 1.20) | ref | 0.85 (0.56 - 1.30) | 0.93 (0.65 - 1.33) | ref | 0.86 (0.56 - 1.32) | 0.92 (0.63 - 1.36) |

Data are presented as odds ratios wit 95% confidence interval. The basic model is adjusted for age and gender of the child, model 2 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 3 is additionally adjusted for orthodontic treatment need based on the IOTN-DHC and IOTN-AC

**Table S3.** The association between ethnicity and OHRQoL

| | Basic model | Model 1 | Model 2 |
|---|---|---|---|
| **Dutch** | Ref | Ref | Ref |
| **Turkish** | -1.06 (-1.73 - -0.39) | -0.75 (-1.43 - -0.63) | -0.82 (-1.48 - -0.15) |
| **Moroccan** | -0.46 (-1.09 - 0.16) | -0.21 (-0.86 - 0.45) | -0.26 (-0.91 - 0.36) |
| **Surinamese** | -0.82 (-1.41 - -0.22) | -0.62 (-1.22 - -0.03) | -0.69 (-1.27 - -0.09) |

Data are presented as weighted linear regression coefficients wit 95% confidence interval. The basic model is adjusted for age and gender of the child, model 2 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 3 is additionally adjusted for orthodontic treatment need based on the IOTN-DHC and IOTN-AC

**Table S4.** Agreement between OOT and SOT in the study population

| | Subjective orthodontic treatment need | |
|---|---|---|
| **Objective orthodontic treatment need** | No need (n (%)) | Need (n (%)) |
| No need (n (%)) | 974 (40.3) | 534 (22.1) |
| Need (n (%)) | 279 (11.5) | 632 (26.1) |

Numbers are presented as absolute numbers with percentages of the total group. SOT = Subjective orthodontic treatment need, OOT = objective orthodontic treatment need

**Table S5.** Non-response analysis (n = 7393)

| | Included | Excluded | p-value |
|---|---|---|---|
| | n = 2419 | n = 4974 | |
| *Family characteristics* | | | |
| **Maternal education level (%)** | | | |
| low | 776 (32.1) | 1649 (33.2) | |
| high | 1513 (62.5) | 1881 (37.8) | < 0.001 |
| *Missing* | 130 (5.3) | 1444 (29.0) | |
| **Paternal education level (%)** | | | |
| low | 767 (31.7) | 1433 (28.8) | |
| high | 1419 (58.7) | 1705 (34.3) | < 0.001 |
| *Missing* | 233 (9.6) | 1836 (36.9) | |
| **Employment status (%)** | | | |
| receiving benefits | 227 (9.3) | 352 (7.1) | |
| receiving no benefits | 2140 (88.4) | 2132 (42.9) | < 0.001 |
| *Missing* | 52(2.1) | 2490 (50.1) | |
| **Household income (%)** | | | |
| < 2000€ | 326 (13.5) | 651 (13.1) | |
| 2000€-3200€ | 681 (28.2) | 759 (15.3) | |
| > 3200€ | 1263 (52.2) | 965 (19.4) | < 0.001 |
| *Missing* | 149 (6.1) | 2599 (52.3) | |
| *Child Characteristics* | | | |
| **Sex (%)** | | | |
| boys | 1219 (50.4) | 2488 (50.0) | |
| girls | 1200 (49.6) | 2486 (50.0) | 0.758 |
| | 0 (0.0) | 0 (0.0) | |
| **Ethnicity (%)** | | | |
| Dutch | 2036 (84.2) | 2156 (43.3) | |
| Turkish | 135 (5.6) | 385 (7.7) | |
| Moroccan | 99 (4.0) | 314 (6.3) | |
| Surinamese | 149 (6.1) | 368 (7.4) | < 0.001 |
| *Missing* | 0 (0.0) | 1751 (35.2) | |
| **Age** | | | |
| Mean (SD) | 9.85 (0.35) | 9.88 (0.40) | 0.002 |
| *Missing* | 0 (0.0) | 0 (0.0) | |
| **IOTN -DHC (%)** | | | |
| little | 903 (37.3) | 848 (17.0) | |
| moderate | 605 (25.0) | 531 (10.7) | |
| great | 704 (29.1) | 663 (13.3) | |
| very great | 207 (8.6) | 224 (4.5) | 0.327 |
| *Missing* | 0 (0.0) | 2708 (54.4) | |
| **IOTN -AC (%)** | | | |
| no need | 1345 (55.6) | 1254 (25.2) | |
| borderline need | 794 (32.8) | 747 (15.0) | |
| definite need | 194 (8.0) | 189 (3.7) | 0.921 |
| *Missing* | 2333 (96.4) | 2784 (56.1) | |
| **Perceived orthodontic need (%)** | | | |
| no need | 637 (26.3) | 413 (8.3) | |
| borderline need | 616 (25.5) | 344 (6.9) | |
| definite need | 1166 (48.2) | 607 (12.2) | 0.024 |
| *Missing* | 0 (0.0) | 3610 (72.6) | |
| **OHRQoL** | | | |
| Median (90% range) | 50.0 (43.0 - 53.0) | 50.0 (42.0 - 53.0) | 0.011 |
| *Missing (%)* | 7 (0.3) | 3590(72.2) | |

Numbers are presented as absolute numbers for categorical variables or as mean (SD) for normally distributed continuous variables or as median (90%range) for non-normally distributed continuous data. P-values are estimated based on chi-square tests, one-way ANOVAs and Kruskal-Wallis tests.

**Table S6.** The association between OOT and OHRQoL among different ethnic groups (n = 2419)

| | Dutch | Turkish | | Moroccan | | Surinamese | |
|---|---|---|---|---|---|---|---|
| | n = 2036 | n = 135 | *p-value*[1] | n = 99 | *p-value*[1] | n = 149 | *p-value*[1] |
| **Basic model** | | | | | | | |
| no need | ref | ref | | ref | | ref | |
| need | -1.10 (-1.39 - -0.82) | -1.61 (- 3.06 - -0.16) | *0.49* | -1.73 (-3.02 - -0.44) | *0.35* | -0.59 (- 1.89 - -0.72) | *0.11* |
| **Model 1** | | | | | | | |
| no need | ref | ref | | ref | | ref | |
| need | -1.10 (-1.38 - -0.81) | -1.03 (-2.41 - 0.35) | *0.92* | -1.84 (-3.15 - -0.53) | *0.28* | -0.17 (-1.41 - 1.07) | *0.15* |
| **Model 2** | | | | | | | |
| no need | ref | ref | | ref | | ref | |
| need | -0.64 (-0.97 - -0.30) | -0.66 (-2.13 - 0.82) | *0.97* | -1.76 (-3.18 - -0.35) | *0.13* | -0.08 (-1.65 - 1.50) | *0.49* |

Data are presented as weighted linear regression coefficients wit 95% confidence interval. The basic model is adjusted for age and gender of the child, model 1 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 2 is additionally adjusted for orthodontic treatment need based on the IOTN-AC. [1]p-value for a significance of difference with the Dutch group obtained from test for heterogeneity. OOT = objective orthodontic treatment need

**Table S7.** The association between SOT and OHRQoL among different ethnic groups (n = 3040)

| | Oral health-related quality of life (ß (95%CI)) | | | | | | |
|---|---|---|---|---|---|---|---|
| **Subjective orthodontic treatment need** | **Dutch** n = 2573 | **Turkish** n = 162 | | **Moroccan** n = 114 | | **Surinamese** n = 191 | |
| **Basic model** | | | *p-value*[a] | | *p-value*[a] | | *p-value*[a] |
| borderline | -0.48 (-0.74 - 0.22) | -1.28 (-2.61 - 0.06) | *0.27* | -1.03 (-3.05 - 1.00) | *0.60* | -0.49 (-1.83 - 0.86) | *0.98* |
| definite | -1.69 (-1.94 - -1.43) | -2.79 (-4.29 - -1.30) | *0.15* | -1.10 (-2.04 - 0.20) | *0.31* | -1.64 (-2.81 - -0.47) | *0.93* |
| **Model 1** | | | | | | | |
| borderline | -0.49 (-0.74 - -0.23) | -1.28 (-2.47 - -0.09) | *0.19* | -0.84 (-2.89 - 1.20) | *0.73* | -0.48 (-1.76 - 0.82) | *0.98* |
| definite | -1.68 (-1.93 - -1.43) | -2.17 (-3.54 - -0.81) | *0.49* | -1.06 (-2.40 - 0.29) | *0.38* | -1.66 (-2.76 - 0.56) | *0.98* |
| **Model 2** | | | | | | | |
| borderline | -0.44 (-0.69 - -0.18) | -1.19 (-2.52 - 0.12) | *0.02* | -1.66 -3.75 - 0.44 | *0.05* | -0.45 (-1.75 - 0.85) | *0.18* |
| definite | -1.47 (-1.74 - -1.20) | -2.00 (-3.37 - -0.63) | *0.45* | -0.94 (-2.14 - 0.26) | *0.39* | -1.68 (-2.82 - -0.54) | *0.72* |

Data are presented as weighted linear regression coefficients with 95% confidence interval. The basic model is adjusted for age and gender of the child, model 1 is additionally adjusted for the following socioeconomic indicators: maternal and paternal education level, employment status and household income; and model 2 is additionally adjusted for orthodontic treatment need based on the IOTN-DHC and IOTN-AC. [a]p-value for a significance of difference with the Dutch group obtained from test for heterogeneity. SOT = Subjective orthodontic treatment need, OHRQoL = oral health related quality of life

# Chapter 5.2

## Influence of self-esteem on self-perceived orthodontic treatment need and oral health-related quality of life

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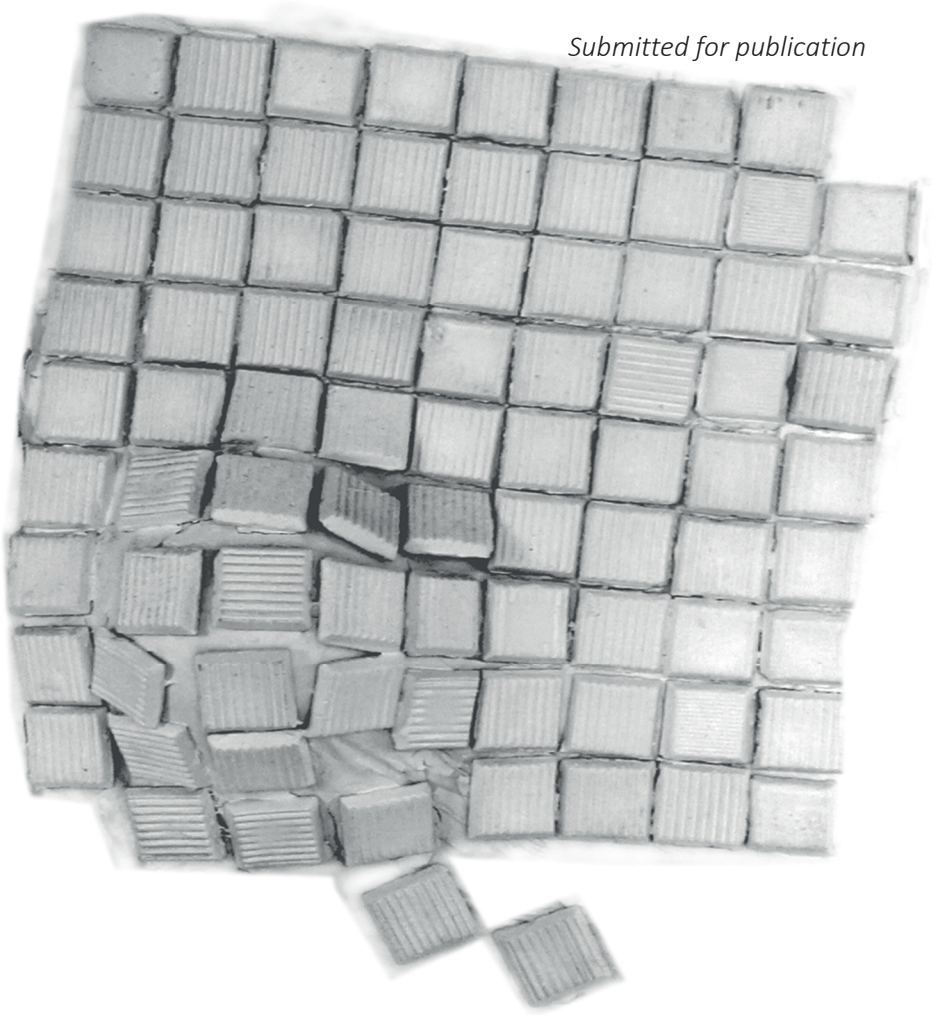

Abstract

***Background*** Self-esteem (SE) is suggested to influence the relationship between orthodontic treatment need and oral health related quality of life (OHRQoL), but evidence is limited. The aim of the present study was to investigate the role of self-esteem in the relationship between subjective orthodontic treatment need (SOT) and OHRQoL in children.

***Methods*** This cross-sectional study was embedded in the Generation R Study, a multi-ethnic population based cohort. OHRQoL, measured with the Child Oral Health Impact Profile-ortho, and SOT were assessed within parental questionnaires. SE was measured with a modified version of the Harter's self-perception profile rated by the children. We evaluated the mediating role of SE in the association between SOT and OHRQoL with linear regression models. Furthermore, we investigated whether there is a difference in this association between children with high and low SE.

***Results*** In total, 3849 children participated in the study. SOT was significantly inversely associated with OHRQoL (borderline: β(95%CI) = -0.55 (-0.77, -0.33); definite: -1.65 (-1.87, -1.54)). Children with lower SE scores showed stronger relationships between borderline and definite SOT with OHRQoL (β (95%CI) = -0.56 (-0.81, -0.31) resp. -1.68 (-1.94, -1.42)) than children with higher SE scores did (β (95%CI) = -0.51 (-0.97, -0.04) resp. -1.43 (-1.90, -0.95)).

***Conclusion*** SE has no mediating role in the association between SOT and OHRQoL. However, SE modifies the relationship between SOT and OHRQoL. Work still needs to be done to find an explanation for the effect modification by SE in the relationship between subjective health perceptions and OHRQoL.

***Keywords:*** quality-of-life, self-esteem, subjective need, malocclusions, children

## Introduction

Oral health related quality of life (OHRQoL) is the most commonly used patient reported outcome measure in dental research (1). It measures the subjective impact of one's own oral health on daily life in different domains, including functional limitations, social emotional wellbeing, school performance and peer interaction (2). Especially in the dental field of orthodontics, OHRQoL gained increasing importance to supplement ordinary objective clinical measures (3). Because objective clinical measures often cannot explain the demand for orthodontic treatment need, OHRQoL is a valuable complementary measure to understand some of the variation between subjective and objective orthodontic treatment need.

A useful framework for research on OHRQoL is provided by the Wilson and Cleary model (4). Based on this model, biological/physical variables influence OHRQoL via symptom status, functional status and general oral health perception, the latter giving the subjective rating to the model. Moreover, this pathway is influenced by environmental factors, like social economic position, and individual characteristics, like self-esteem (SE). SE is described as the subjective ability to deal with the environment and is impacted by the interactions with others (5). In contrast to OHRQoL, SE is considered to be a stable construct (6).

In the case of orthodontics, the association between biological/physical variables and OHRQoL has been extensively investigated (7-9). Children with malocclusions perceive significant impacts on OHRQoL (7). Also, different studies investigated the role of SE in the relationship between malocclusion and OHRQoL. It has been shown that OHRQoL is positively associated with SE (10, 11). However, orthodontic treatment could not be proven to advance SE, neither had children with malocclusions consistently lower SE (12-15). In contrast, the association between self-perceived orthodontic treatment need and OHRQoL is less widely investigated and to our knowledge, the role of self-esteem has not at all been investigated in the association between subjective orthodontic treatment need (SOT) and OHRQoL. According to the Wilson and Cleary Model SE possibly influences the relation between biological/physical variables and OHRQoL, presumably by acting on general oral health perception and OHRQoL, rather than the functional or symptoms status (see figure 1). Therefore, the aim of the present study was to investigate the role of self-esteem in the relationship between SOT, as one representative for oral health perception, and OHRQoL among 9 year old children living in Rotterdam, The Netherlands.

## Material & Methods

### *Study design and study population*

The study was performed within the Generation R Study, which is a prospective multiethnic population-based cohort in Rotterdam, The Netherlands. Details of the Generation R Study have been extensively described elsewhere (16, 17). The Generation R Study was approved by the Medical Ethics Committee of the Erasmus University Medical Centre (MEC- MEC-2012-165). All participants provided written informed consent before data collection started.

All pregnant women which had a delivery date between April, 1 2002 and January 31, 2006 living in the study area were invited to participate in the study. In total, n = 8548 participants were eligible to participate in the study phase from children's age of 9 years onwards, of which n = 7393 participants gave full consent for participation. Data on children's OHRQoL and SOT assessed at the age of 9 was available from n = 3849, which compromise the study population for the present study.

*Oral health-related quality of life*

OHRQoL of the children at the age of 9 was assessed by questionnaires, which were sent to the mothers of the children. OHRQoL was measured with an 11-item version of the Children's Oral Health Impact Profile (COHIP). This version of the COHIP has been validated to measure OHRQoL related to malocclusions (18). The questions of the short COHIP version were answered on a five-point Likert scale and covered five domains of children's oral health: oral symptoms, functional well-being, emotional well-being, school and peer interaction. All answers were added up to a final OHRQoL score (range 11-55 points), with the highest score indicating the best quality of life. Missing values in the responses to the questionnaire were replaced by the personal mean score of the remaining answers, as proposed by other researchers using the original version of the COHIP (19). If there were more than 30% of the answers missing, the participant was excluded from the analysis. The individual questions of the 11-item version of the COHIP are presented in the appendix (Table S1).

*Self-perceived orthodontic treatment need*

Self-perceived orthodontic treatment need was measured with the question "Do you want your child to get braces?". This question was also included in the maternal questionnaires. The mother answered the question on a five point Likert scale, with answer possibilities ranging from "strongly disagree" to "strongly agree". For the data analysis answers are categorized into self-perceived need (strongly / somewhat agree), borderline self-perceived need (do not agree/ do not disagree) vs no need (strongly / somewhat disagree).

*Self-esteem*

Self-esteem was assessed in questionnaires sent directly to the children. For this an adapted question format of the Harter's self-perception profile according to Wichstrom (1995) was used (20). Because Wichstrom (1995) adapted the adolescent version of the Harter's self-perception profile, however, we studied younger children, we used the question format as Wichstrom suggested, but applied this to the validated self-perception profile for children (CBSK in Dutch) (21). Four subscales of the CBSK were used: school competence (5 items), social acceptance (5 items), athletic competence (3 items) and physical appearance (3 items). One item from the physical appearance scale and one from the school competence scale of the CBSK, because of spatial limitations and those items seemed to be most redundant. We did however add two items, as global indicators of self-worth. Also, slight adaptions of wording were made, to make the questionnaire more up to date. In addition, we revised the four point coding into a three point coding system. That is, because it has been established by Achenbach that

variability of items scores is higher when a three point coding system is used (22). Thus, the children answered the questions of the CBSK with one of the three options: 'not true', 'somewhat true' or 'true'. All answers were added up to a final SE score (range 18 - 54 points) or SE subscale score respectively, with the highest score indicating the highest SE. Missing values in the responses to the CBSK were replaced by the mean score of the remaining answers for the particular subscale. If there were more than 30% of the answers missing per subscale the SE score was coded as missing value. The overall SE score was categorized into high and low based on a 20 % cut-off at a SE score of 28.0. The individual items of the adapted format of the Harter's self-perception profile are presented in the appendix (Table S2).

*Covariates*

Potential confounding factors were considered from three domains: social economic position, individual child characteristics and clinical variables. Social economic position was captured with maternal and paternal education level (high: higher vocational training, university or PhD degree vs. low: no education, primary school, lower or intermediate vocational training, general school or first year of higher vocational training), with netto household income (≤ 2000€ vs. > 2000€), and maternal marital status (married, registered partnership, living together vs. no partner all, partner with whom I do not live). Individual child characteristics covered age, gender and ethnicity of the child. Children's ethnic background was based on the ethnicity of their mother, because this takes into account their cultural background as mothers are most often the primary caregivers. Maternal ethnic background was based on the country of birth of the mothers parents and a mother was considered non-native Dutch if at least one of her parents were born in another country. Finally following clinical variables were considered: caries experience (diseased, missing and filled teeth (dmft) index = 0 vs. dmft index > 0), orthodontic treatment need based on the Dental Health Component (IOTN-DHC) and Aesthetic component (IOTN-DHC) of the Index of orthodontic treatment need (no need (IOTN-DHC ≤ 3) vs. need (IOTN-DHC > 3) and no need (IOTN-AC 1-4) vs. borderline need (IOTN-AC 5-7) vs. need (IOTN-AC 8-10)), tooth brushing frequency (once or less a day vs. twice or more a day) and dental visits (more than one year ago vs. less than one year ago). The dmft index and IOTN-AC as well as IOTN-DHC have been assessed from photographic and radiographic records, which has been extensively described elsewhere (23, 24). All covariates were assessed, or verified, at the children's age of 9 years, except for maternal and paternal education level, marital status and caries experience, which were assessed at the children's age of 5 years.

*Statistical analysis*

Differences in sample characteristics among children with no, borderline or definite SOT were evaluated with Chi-square tests for categorical data and Kruskall-Wallis-tests or analysis of variance for continuous data. Then, Spearman correlations analysis were conducted between SOT and the SE overall score as well as the SE subscale scores (Appendix Table S4), and overall SE with SOT as well as IOTN-AC (Appendix table S5). The difference in OHRQoL according to high and low overall SE was evaluated with a Mann-Whitney-U-test (Appendix table S6).

Finally, we used linear regression models with weighted least squares to evaluate the role of SE in the association between SOT and OHRQoL.

**Table 1.** Characteristics of the study population (n = 3849)

| | Subjective orthodontic need | | | |
|---|---|---|---|---|
| | **No** n = 1075 | **Borderline** n = 980 | **Yes** n = 1794 | p-value |
| *Family characteristics* | | | | |
| **Maternal education level** | | | | |
| Low (n (%)) | 385 (35.8) | 298 (30.4) | 576 (32.1) | |
| High (n (%)) | 609 (56.7) | 616 (62.9) | 1115 (62.2) | 0.011 |
| **Paternal education level** | | | | |
| Low (n (%)) | 358 (33.3) | 283 (28.9) | 572 (31.9) | |
| High (n (%)) | 572 (53.2) | 566 (57.8) | 1017 (56.7) | 0.077 |
| **Household income** | | | | |
| ≤ 2000€ (n (%)) | 184 (17.1) | 144 (14.7) | 240 (13.4) | |
| > 2000€ (n (%)) | 749 (69.7) | 693 (70.7) | 1291 (72.0) | 0.036 |
| **Marital status** | | | | |
| Married (n (%)) | 889 (82.7) | 809 (82.6) | 1505 (83.9) | |
| No partner (n (%)) | 107 (10.0) | 103 (10.5) | 178 (9.9) | 0.852 |
| *Child characteristics* | | | | |
| **Age** | | | | |
| mean±SD | 9.87±0.37 | 9.82±0.34 | 9.86±0.37 | 0.007 |
| **Gender** | | | | |
| Boy (n (%)) | 577 (53.7) | 510 (52.0) | 827 (46.1) | |
| Girl (n (%)) | 498 (46.3) | 470 (48.0) | 967 (53.9) | 0.000 |
| **Ethnicity** | | | | |
| native Dutch (n (%)) | 671 (62.4) | 676 (69.0) | 1267 (70.6) | |
| non Dutch (n (%)) | 388 (36.1) | 285 (29.1) | 501 (27.9) | 0.000 |
| **Caries experience[2]** | | | | |
| 0 (n (%)) | 585 (54.4) | 562 (57.3) | 1011 (56.4) | |
| > 0 (n (%)) | 195 (18.4) | 175 (17.9) | 340 (19.0) | 0.759 |
| **Tooth brushing** | | | | |
| Once or less a day (n (%)) | 214 (19.9) | 167 (17.0) | 287 (16.0) | |
| Twice or more a day (n (%)) | 854 (79.4) | 808 (82.4) | 1498 (83.5) | 0.025 |
| **Dental visits** | | | | |
| > 1 year ago (n (%)) | 26 (2.4) | 15 (1.5) | 33 (1.8) | |
| < 1year ago (n (%)) | 1047 (97.4) | 958 (97.8) | 1756 (97.9) | 0.329 |
| **Aesthetic orthodontic need** | | | | |
| No (n (%)) | 604 (56.2) | 512 (52.3) | 568 (31.7) | |
| Borderline (n (%)) | 178 (16.6) | 232 (23.7) | 588 (32.8) | |
| Yes (n (%)) | 17 (1.6) | 20 (2.0) | 208 (11.6) | 0.000 |
| **Objective orthodontic need** | | | | |
| No (n (%)) | 656 (61.0) | 587 (59.9) | 648 (36.1) | |
| Yes (n (%)) | 170 (15.8) | 205 (20.9) | 764 (42.6) | 0.000 |
| **OHRQoL** | | | | |
| median (90% range) | 51.0 (45.0 - 53.0) | 50.0 (44.0 - 53.0) | 49.0 (41.0 - 52.0) | 0.000 |
| **SE overall** | | | | |
| median (90% range) | 47.0 (37.0 - 52.0) | 46.0 (38.0 - 51.0) | 46.0 (37.0 - 52.0) | 0.171 |

Data may not add up to n = 3849, because they are based on the non - imputed data set. Missing values: maternal education: 6.4%, paternal education level: 12.5%, household income: 14.2%, marital status: 6.7%, ethnicity: 1.6%, caries experience: 25.5%, toothbrushing: 0.5%, dental visits: 0.1%, aesthetic orthodontic need: 23.9%, objective orthodontic need: 21.3, SE total: 6.4%; p-value is based on chi -square test for categorical data and UNIANOVA or Kruskall- Wallis-test for continuous data.

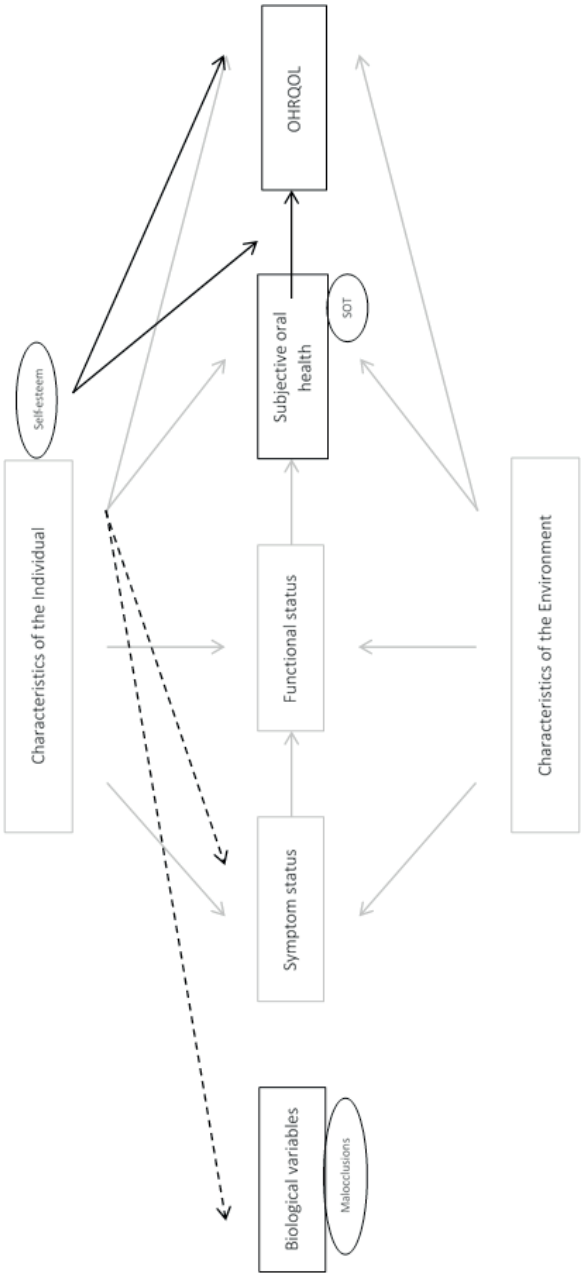


**Figure 1.** Relationships between objective and subjective oral health measures based on the Wilson and Cleary model. Grey lines indicate relationships suggested by Wilson and Cleary, Black lines indicate the associations investigated in this study, and dotted black lines indicates relationships investigated in other studies, but not proven yet. Self-esteem is depicted as one of the individual characteristics in the model, malocclusions is depicted as a biological/physiological variable.

Generally, we built 3 different models with SOT as determinant and OHRQoL as outcome variable; A basic model adjusted for gender and age only, model 1 additionally adjusted for paternal education level, household income and marital status, and model 2 additionally adjusted for caries experience, IOTN-AC and IOTN-DHC. We included the confounding variables into the model based on the association between the covariates with SOT, OHRQoL and self-esteem. In another step, overall SE was added to each model to assess the extra amount of variance explained for OHRQoL ($R^2$ change) and to evaluate the significance of this change. We also calculated the percentage change in estimate after adding SE to the model for borderline and definite SOT (($\beta_{model}$ - $\beta_{model+SE}$)/($\beta_{model}$)). Finally, we evaluated whether there is a difference in the association between SOT and OHRQoL between children with high and low SE by evaluating interaction terms between SOT and SE in the model and presenting a stratified analysis. We built interaction terms separately for the borderline self-perceived need and definite self-perceived need group with SE (continuous variable). The association between SOT and OHRQoL was also presented stratified for high and low SE. Because we had to deal with missing data in the covariates and determinant variable, a multiple imputation was applied. For this, 10 imputed datasets were generated by using a fully conditional specified model, which takes into account the uncertainty of the data. We present the pooled estimates from these 10 dataset as betas with 95% confidence intervals (β (95%CI)). For all analysis, a p-value < 0.05 was considered to be significant. Analyses were performed in SPSS 21.0 (IBM Statistics Inc, Chicago, IL, USA).

### *Non-response analysis*

Children which were excluded from the study, because of loss to follow up or missing data on OHRQoL (n = 4752) were compared with children included into the study (n =3796) using chi-square tests and t-tests. The excluded population had more often a low maternal and paternal education level, low household income and were more often single parenting, from ethnic minorities and with a higher caries prevalence (all p-values < 0.001). The non-response analysis is presented in the Appendix (Table S7).

## Results

### *Sample characteristics*

In table 1 the family and child characteristics of the study population are presented by SOT. Of all participating children 1075 had no SOT (27.9%), 980 had borderline SOT (25.5%) and 1794 had definite SOT (46.6%). Parents from children with SOT were higher educated (p-values = 0.011/0.077) and had a higher household income (p-value = 0.036). Furthermore, children with SOT were more often female (p-value < 0.001), native Dutch (p-value < 0.001), brushed their teeth more often (p-value = 0.025), had more often an unfavorable IOTN-AC grade (p-value < 0.001), were more often in need for objective orthodontic treatment (p-value < 0.001) and had lower OHRQoL (p-value < 0.001) than children without or with borderline SOT. There were no significant differences in the other sample characteristics among the SOT groups.

### *Self-esteem in the association between subjective orthodontic treatment need and OHRQoL*

SOT was significantly inversely associated with OHRQoL based on the fully adjusted model (borderline need: β (95%CI) = -0.55 (-0.77, -0.33); definite need: β (95%CI) = -1.61 (-1.87, -1.42)).

SE was not significantly different between the groups based on SOT (p-value = 0.171, table 1). Furthermore, adding SE to the model on the association between SOT and OHRQoL did not attenuate or strengthen the association between SOT and OHRQoL with more than 10% (appendix table S4). However, adding SE to the model on the association between SOT and OHRQoL improved the model significantly (p-values < 0.001, table 2). In the fully adjusted model on SOT and OHRQoL, SE was significantly positively associated with OHRQoL (β (95%CI) = 0.08 (0.06, 0.11)).

### *Subjective orthodontic treatment need associated with OHRQoL stratified by self-esteem*

After stratification for low and high SE, the association between SOT and OHRQoL appeared to be modified by children's SE (table 3). Based on the fully adjusted model, the association between borderline SOT and OHRQoL children was little but significantly stronger in children with low SE (β (95%CI) = -0.56 (-0.81, -0.31)) than in children with high SE (β (95%CI) = -0.51 (-0.97, -0.04)) (p-value = 0.02). In contrast, the association between definite SOT and OHRQoL was more profound, but non-significantly stronger in children with low SE (β (95%CI) = -1.68 (-1.94, -1.42)) than in children with high SE (β (95%CI) = -1.43 (-1.90, -0.95)) (p-value = 0.28)).

**Table 2.** Associations between SOT[1] and OHRQoL[2] by subjective orthodontic treatment need2 and the role of SE[3] in this association (n = 3849)

| | | | OHRQoL[2] (β (95% CI)[4] | | |
|---|---|---|---|---|---|
| | | | Basic model | Model 1 | Model 2 |
| **Step 1** | **Subjective orthodontic need** | | | | |
| | | borderline | -0.54 (-0.77 - -0.31) | -0.60 (-0.80 - -0.36) | -0.55 (-0.77 - -0.33) |
| | | yes | -1.77 (-2.00 - -1.54) | -1.78 (-2.00 - -1.57) | -1.65 (-1.87 - -1.42) |
| **Step 2** | **Subjective orthodontic need** | | | | |
| | | borderline | -0.51 (-0.73 - -0.30) | -0.55 (-0.77 - -0.34) | -0.53 (-0.74 - -0.31) |
| | | yes | -1.71 (-1.93 - -1.49) | -1.74 (-1.95 - -1.53) | -1.61 (-1.84 - -1.39) |
| | SE | | 0.10 (0.07 - 0.12) | 0.09 (0.06 - 0.11) | 0.08 (0.06 - 0.11) |
| ***$R^2$ change[5]*** | | | 0.02 | 0.02 | 0.01 |
| ***p-value[6]*** | | | < 0.001 | < 0.001 | < 0.001 |

[1] SOT = subjective orthodontic treatment need [2]OHRQoL = oral health related quality of life;;[3]SE= self-esteem;[4] beta and 95% confidence interval (β (95% CI)) obtained from weighted least square linear regression models. [5]Change in $R^2$ between step 1 (SE not included) and step 2 (SE included), [6]p-value for significance of $R^2$ change. Basic model adjusted for age and gender only, model 1 additionally adjusted for paternal education level, household income and ethnicity; model 2 additionally adjusted for caries experience, aesthetic orthodontic need and objective orthodontic need.

**Table 3.** Association between subjective orthodontic treatment and OHRQoL by SE (n = 3849)

| | | Low SE | High SE |
|---|---|---|---|
| | | N = 3146 | N = 703 |
| | | β (95% CI) | β (95% CI) |
| **Basic model** | | | |
| | borderline | -0.52 (-0.79 - -0.26) | -0.56 (-1.04 - -0.08) |
| | yes | -1.81 (-2.06 - -1.55) | -1.48 (-1.95 - -1.01) |
| **Model 1** | | | |
| | borderline | -0.58 (-0.83 - -0.33) | -0.52 (-0.98 - -0.06) |
| | yes | -1.85 (-2.09 - -1.60) | -1.43 (-1.88 - -0.98) |
| **Model 2** | | | |
| | borderline | -0.56 (-0.81 - -0.31) | -0.51 (-0.97 - -0.04) |
| | yes | -1.68 (-1.94 - -1.42) | -1.43 (-1.90 - -0.95) |
| **p-value for borderline*** | | 0.020 | |
| **p-value for yes*** | | 0.280 | |

[1] OHRQoL=oral health related quality of life; [2]SE= self-esteem; Beta and 95% confidence interval (β (95% CI)) obtained from weighted least square linear regression models. Basic model adjusted for age and gender only, model 1 additionally adjusted for paternal education level, household income, marital status and ethnicity; model 2 additionally adjusted for caries experience, aesthetic orthodontic need and objective orthodontic treatment need. * obtained from interaction term entered into the basic model.

## Discussion

### *Short summary of the results*

The results of our study suggest a different role of SE than expected. SE did not mediate or confound the association between SOT and OHRQoL, thus SOT did not influence OHRQoL via SE. However, SE is a determinant for OHRQoL that modified the association between SOT and OHRQoL.

### *Interpretation of results in relation to the literature*

In line with other studies, we found a significant relationship between SE and OHRQoL (10, 11). This confirmed, that SE is one of the psychosocial determinants of OHRQoL as proposed by the Wilson & Cleary model and described by many other authors (4, 25). Based on the Wilson and Cleary model malocclusion influences OHRQoL via symptom status, functional status and general oral health perception and this pathway in turn should be affected by self- esteem (4). However, there is no evidence confirming the relevance of SE in the association between malocclusion and OHRQoL (10, 26). Malocclusions seemed unrelated to SE and thus doubts arose whether SE is one of the individual characteristics determining OHRQoL, especially in the dental subspecialty of orthodontics. The present study investigated the confounding and mediating role of SE in the relationship between SOT and OHRQoL, which might be different to the role of SE in the association between malocclusion and OHRQoL (see figure 1). Still, SE was also unrelated to SOT and also did not change the effect estimates between SOT and OHRQoL with more than 10 percent. Thus, SE did neither mediate nor confound the association between SOT and OHRQoL. However, we proved that SE is still a determinant for OHRQoL. Based on our results, SE might influence OHRQoL in two ways, name-

ly on the one hand directly and on the other hand as modifier in the association with SOT, as children with a lower SE score showed a significantly stronger relationship between borderline SOT and OHRQoL than children with a higher SE score. Our findings might explain why other researches did not find a mediating role of SE between objective orthodontic variables and OHRQoL. However, this might be unique for the orthodontic field, as for example the relationship between caries and OHRQoL is mediated through self-esteem and other psychological variables (27, 28).
In contrast to OHRQoL, which is considered to have a dynamic, context-specific character, SE is a relatively stable construct [6]. SE is a personal resource that facilitates coping with less favorable conditions, such as poor oral health [9, 26]. Therefore, it seems not only coherent that OHRQoL is correlated with SE in our study as well as in other studies, but also that malocclusions are unrelated to SE. High SE is a psychological resource that protects individuals from the effects of deleterious oral conditions, but still children with low SE might be more focused on their malocclusion [9]. In line with this, the present finding suggest a modifying role of SE on the relationship between SOT and OHRQoL. The absence of an association between SOT and SE, however, appeared rather surprising, because earlier studies found a relationship between SE and the way people are satisfied with their faces; those with higher SE showed less frequent impacts from their malocclusion, suggesting less self-perceived orthodontic treatment need (26, 29). First, the relationship between SOT and objective orthodontic treatment need might explain the absence of an association between SOT and SE, because SE has been shown to be unrelated with orthodontic treatment seeking (30). Possibly SOT is more related to advices from the professional or comparison with friends and family or even objective orthodontic treatment need than expected, and less to emotional impacts than suggested. Secondly, the impacts of SE on OHRQoL might be much less related to malocclusions but rather reflect other (oral) conditions (27, 28, 31).

*Limitation and Strength*

Some limitations of the study have to be considered. First, the OHRQoL questionnaire as well as SOT were assessed with questionnaires addressed to the mothers instead of the children themselves. This might have led to information bias, however several studies discussed maternal reports regarding patient reported oral health outcome measures as valid proxies for children reports (32-34). Second, in the non-response analysis, data were more often missing in children from low socioeconomic position and with caries. This could have caused selection bias, when the association between SOT and OHRQoL and the role of SE in this association is different between the included and the excluded population. However, the conclusion of our findings did not change after adjusting our analysis for socioeconomic status and oral conditions and therefore a selection bias in the present study seems unlikely. Third, although we adjusted our analysis for several factors that are thought to influence OHRQoL, residual confounding might have affected our results as it is a general thread to observational studies. Finally, SE was the only psychological factor investigated in the present study, thus we cannot say anything about the influence of other factors related to the children's psychological profile on OHRQoL. However, several studies suggested the relationship between other psychological factors, like sense of coherence, health locus of control and coping beliefs with

oral health (related quality of life) (25, 35). Yet, to our knowledge, this is the first study, which investigates the role of SE in the association between SOT and OHRQoL. The major strength of the study is, that we made use of a large population based sample including n = 3976 children, instead of using a small selected clinical sample. Furthermore, we were able to combine objective clinical measures as well as questionnaire data in this study.

*Implications of the result for research and practice*

Orthodontics is a major oral health problem among children and adolescent, as more than half of the young adolescents have received orthodontic treatment (36-38). As the relationship between subjective and objective orthodontic treatment need is very inconsistent, many different reasons unrelated to the severity of malocclusions seem to exist why to seek or not to seek orthodontic treatment. The present study clearly indicates that clinical measures are not sufficient to assess the impacts of malocclusions and the objective need for treatment, but subjective measures like OHRQoL need to be included as well. As care givers are not only interested in aligning their patient's teeth, but also in improving their OHRQoL, it is important for them to understand the relationships between clinical indicators and psychological indicators on OHRQoL. The present study is also important for future oral health research, as we support the idea to take SE into consideration when investigating relationships regarding emotional impacts of oral health and OHRQoL, however not to expect relationships with SE among clinical measures and their social impacts.

*Conclusion*

From the results obtained, we confirm that SE is a relevant determinant of OHRQoL as proposed by the Wilson and Cleary model, which describes the pathway between biological/physical variables, in this case malocclusions, and OHRQoL. Whereas other studies already suggested SE to be unrelated to malocclusions but to be associated with OHRQoL, we showed that SE is also unrelated to SOT. Our findings, however, suggest that SE modifies the relationship between SOT and OHRQoL, which has not been established before. Work still needs to be done to understand and explain the role of SE for OHRQoL, as such as well as in relation to oral health perceptions.

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## Supplemental material

**Table S1.** The 11 items of the Cohip-ortho, used in the present study

| |
|---|
| During the past 3 month, how often has your child: |
| 12. had pain in his/her teeth/toothache |
| 13. had crooked teeth or spaces between his/her teeth |
| 14. had discolored teeth or spots on his/her teeth |
| 15. had bad breath |
| 16. had bleeding gums |
| 17. had difficulty eating foods he/she would like to because of his/her teeth, mouth, or face |
| 18. felt worried or anxious because of his/her teeth mouth or face |
| 19. not wanted to speak/ read out loud in class because of his/her teeth, mouth, or face |
| 20. been teased, bullied or called names by other children because of his/ her teeth, mouth, or face |
| 21. felt that he/she was attractive (good looking) because of his/ her teeth, mouth, or face |
| 22. had difficulty saying certain words because of his/her teeth or mouth |

**Table S2.** The 18 items of the adapted Harter's Self-perception Profile

| | |
|---|---|
| **School competence** | |
| | 1. I feel just as smart as others my age |
| | 2. I am pretty slow finishing my school work |
| | 3. I have trouble figuring out the right answers in school |
| | 4. I do very well at school |
| | 5. I often forget what I have learned |
| **Social acceptance** | |
| | 6. I find it hard to make friends |
| | 7. I have a lot of friends |
| | 8. I often do things on my own |
| | 9. I feel I am socially accepted by people my age |
| | 10. I am popular with others my age |
| **Athletic competence** | |
| | 11. I do very well at all kinds of sports |
| | 12. I am better than others my age at sports |
| | 13. I often look others doing sports rather than do it |
| **Physical appearance** | |
| | 14. I wish my body was different |
| | 15. I wish my physical appearance was different |
| | 16. I think I am good looking |
| **Global self-worth** | |
| | 17. I am often disappointed in myself |
| | 18. I like the kind of person I am |

**Tabel S3.** Non-response analysis among all participants participating in the Generation R study phase at children's age of 9-years (n = 7393)

| | Excluded population | Included population | |
|---|---|---|---|
| | N = 3544 | N = 3849 | p-value |
| *Family characteristics* | | | |
| **Maternal education level** | | | |
| Low | 1158 | 1267 | |
| High | 1044 | 2350 | 0.000 |
| **Paternal education level** | | | |
| Low | 980 | 1220 | |
| High | 962 | 2162 | 0.000 |
| **Household income** | | | |
| ≤ 2000€ | 367 | 576 | |
| > 2000€ | 916 | 2739 | 0.000 |
| **Marital status** | | | |
| married | 1821 | 3216 | |
| no partner | 395 | 393 | 0.000 |
| *Child characteristics* | | | |
| **Age** | | | |
| mean±SD | 9.85±0.36 | 10.09±0.47 | |
| **Gender** | | | |
| Boy | 1783 | 1923 | |
| Girl | 1738 | 1949 | 0.409 |
| **Ethnicity** | | | |
| native Dutch | 1566 | 2626 | |
| non Dutch | 1664 | 1184 | 0.000 |
| **Caries experience[2]** | | | |
| 0 | 1288 | 2167 | |
| > 0 | 777 | 713 | 0.000 |
| **Tooth brushing** | | | |
| Once or less a day | 3 | 671 | |
| Twice or more a day | 20 | 3178 | 0.580 |
| **Dental visits** | | | |
| > 1 year ago | 1 | 74 | |
| < 1year ago | 21 | 3783 | 0.372 |
| **Aesthetic orthodontic need** | | | |
| No | 908 | 1691 | |
| Borderline | 535 | 1006 | |
| Yes | 136 | 247 | 0.958 |
| Objective orthodontic need | | | |
| No | 985 | 1902 | |
| Yes | 652 | 1146 | 0.134 |
| **Subjective orthodontic need** | | | |
| no | 1 | 1075 | |
| borderline | 2 | 980 | |
| yes | 8 | 1794 | 0.199 |

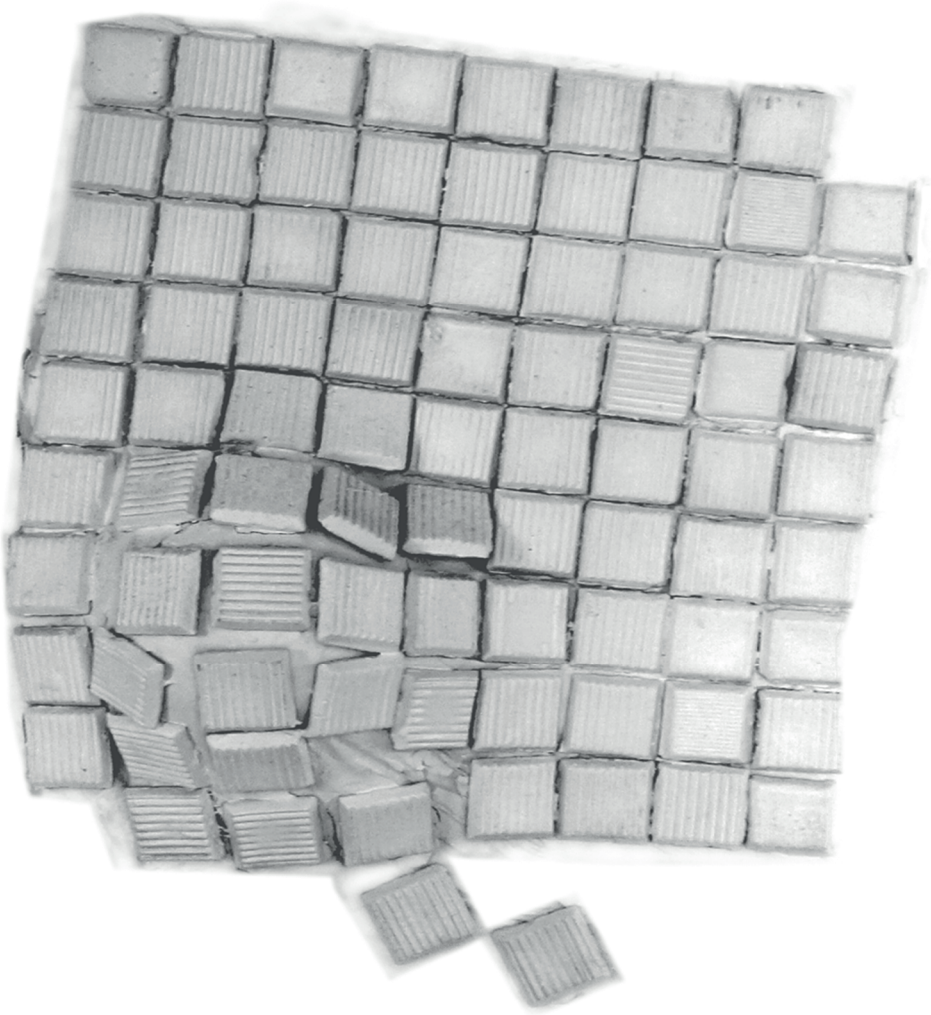
p-value is based on chi -square test for categorical data and t-test or Mann-Whitney-U test for continuous data. OHRQoL = oral health related quality of life, dmft= Diseased, missing and filled teeth index, SE=self-esteem

**Table S4.** Percentage change in estimate after adding SE to the model for borderline and definite SOT

| | %* | |
|---|---|---|
| | **borderline** | **yes** |
| **Basic model** | 5.5 | 3.4 |
| **Model 1** | 8.3 | 2.3 |
| **Model 2** | 3.6 | 2.4 |

*calculated as ((βmodel - βmodel+SE)/(βmodel))

# Chapter 6
# General discussion



## General discussion

### Introduction

To understand the relationships among measures of objective and subjective oral health with OHRQoL related to the orthodontic field, we developed methods to assess children's OHRQoL and orthodontic need in large scale epidemiologic studies and we examined the influence of (non-) clinical variables on these relationships. In the following chapter we report the main findings of the studies presented in this thesis. Subsequently, we discuss the relevant methodological consideration of the studies and place the studies in their context. What are the implications of the findings for patients, clinicians, researchers and policymakers? Finally, we provide unanswered questions for future research and finish with a general conclusion. An overview of the main findings of the studies presented in this thesis is given in table 1.

### Statement of the principal findings

#### *Methods to conduct large scale orthodontic and OHRQoL research*

In chapter 2.1, we developed and validated, based on the Children Oral Health Impact Profile (COHIP-38) a short OHRQoL measure, the COHIP-ortho, applicable in 8-13 year old children. Our results showed that the 11- item version performed as good as the original 38 - item version with regard to psychometric properties in an orthodontic study sample.

In chapter 2.2 we described and investigated the validity and reliability of a new method for systematic, accurate and credible data collection for orthodontic treatment need. For this we used the Index of Orthodontic Treatment Need (IOTN), applied to a combination of 2D and 3D photographs and radiographs (orthopantomograms and cephalograms). The results suggested that compared to the assessment of the IOTN on plaster casts, the assessment of the IOTN on a combination of 2D- pictures, 3D- pictures and radiographs was fair. Whereas orthodontic treatment need in general terms could be assessed from 2D and 3D pictures only, the combination with radiographs improved the assessment and enabled a valid IOTN scoring.

#### *Biological and Physiological factors influencing OHRQoL*

In chapter 3.1, 3.2 and 3.3 we describe the relationship between oral health status and OHRQoL. First, we investigated in a systematic review the association between malocclusions as objectively assessed orthodontic treatment need, which is one of the most common oral health problems in childhood, and OHRQoL, presented in chapter 3.1. This study showed that children with malocclusion have significant lower OHRQoL than children without malocclusion, although the impact of malocclusions on OHRQoL seemed rather small.

Next to objectively assessed orthodontic treatment need, the association between subjective orthodontic treatment need and OHRQoL was investigated and showed that self-perceived orthodontic treatment need is associated with poor OHRQoL in children. Moreover, the more orthodontic treatment need was perceived by the children, the

lower was their OHRQoL score, independently of their objectively assessed orthodontic treatment need.
Finally, we also investigated the association between caries experience and OHRQoL. The results of this study showed that early caries experience and later OHRQoL are inversely related. Compared to children with a healthy dentition, children with severe caries at the age of 6 were significantly more likely to have poor OHRQoL at the age of 10.

### *Characteristics if the environment influencing OHRQoL*

Chapter 4 describes the association between socioeconomic position and OHRQoL. In this study, we used various socioeconomic indicators to depict the family's socioeconomic position of the children. Children from parents with low educational level, from unemployed parents, children from families with low household income or with benefit dependency as well as children from a single-parenting household had consistently poorer OHRQoL. This association was almost completely independent of objective oral health indicators like caries experience and orthodontic treatment need.

**Table 1.** Overview of main results

| Chapter | Main exposures | Main Outcomes | Main results |
|---|---|---|---|
| **2. Methods to conduct large scale orthodontic and OHRQoL research** | | | |
| 2.1 | Not applicable | OHRQoL | The COHIP-ortho is a good questionnaire to be used in research on children's OHRQoL. |
| 2.2 | Not applicable | Orthodontic treatment need | The use of 2D and 3D pictures is suitable to assess orthodontic need in scientific studies, but the method improves when 2D and 3D pictures are combined with radiographs. |
| **3. Biological and Physiological factors influencing OHRQoL** | | | |
| 3.1 | Malocclusions | OHRQoL | Children with malocclusion have significant lower OHRQoL. The strength of the association varies among age and cultural differences. |
| 3.2 | Subjective orthodontic treatment need | OHRQoL | Self-perceived orthodontic treatment need is associated with poor OHRQoL in children, independently of their objective oral health status. |
| 3.3 | Caries experience | OHRQoL | Early caries experience and later OHRQoL in childhood are inversely related. |
| **4. Characteristics of the environment influencing OHRQoL** | | | |
| 4.1 | Different SEP indicators | OHRQoL | Children from a lower family SEP had significantly lower OHRQoL independent of their objective oral health status |
| **5. Characteristics of the individual influencing OHRQoL** | | | |
| 5.1 | Objective orthodontic treatment need, Ethnic background | Subjective orthodontic treatment need, OHRQoL | objective orthodontic treatment need was not different among children from different ethnic groups. However, children from ethnic minority groups tend to have lower OHRQoL |
| 5.2 | Subjective orthodontic treatment need, Self-esteem | OHRQoL | Children with low self-esteem show a stronger association between subjective orthodontic treatment need and OHRQoL than children with high self-esteem |

OHRQoL = oral health-related quality of life
SEP = socio-economic position

### *Characteristics of the individual influencing OHRQoL*

In chapter 3.1, chapter 3.2, chapter 5.1 and chapter 5.2 individual factors influencing OHRQoL were discussed. In the systematic review we showed that age of the children

had a major influence on the association between malocclusions and OHRQoL. The older children were, the bigger was the negative impact of their malocclusion on OHRQoL. The findings of the study presented in chapter 3.2 suggest that the association between definite subjective orthodontic treatment need and OHRQoL was stronger in girls, whereas borderline subjective orthodontic treatment need affected OHRQoL more in boys than in girls.
Ethnic background, or cultural differences, were discussed in chapter 3.1 and in chapter 5.1. The findings of the systematic review and meta-analysis showed that there were differences in the association of malocclusion and OHRQoL among the countries of study conduction. In the Generation R Study, we did not find ethnic differences in objective orthodontic treatment need among children. And although we found differences in subjective orthodontic treatment need and OHRQoL among children with different ethnic backgrounds, the associations between agreement of objective and subjective orthodontic treatment need with OHRQoL were not significantly different. Still children from ethnic minority groups tended to have lower OHRQoL than native Dutch children.
Finally, we evaluated the role of self-esteem in the association between subjective orthodontic treatment need and OHRQoL and investigated whether there is a difference in this association between children with high and low self-esteem. Self-esteem did not play a significant role in the association between subjective orthodontic treatment need and OHRQoL. However, children with low self-esteem showed a stronger association between subjective orthodontic treatment need and OHRQoL than children with high self-esteem.

## Strength and weaknesses of the included studies

### *Study design*

The studies described in this thesis do all have an observational design. This means, that no causation can be inferred from the studies presented. Observational studies do not allow to make unexposed and exposed groups similar by randomization with regard to factors, which could influence the outcome of interest. Therefore, observational studies are prone to confounding. However, in observational studies it is possible to adjust associations for possible confounding in the analysis phase and the studies presented in this thesis were adjusted for a wide range of potential confounding factors. Still, residual confounding might have occurred due to poorly measured or unmeasured confounding factors.
Most of the studies presented in this thesis were embedded in the Generation R Study, a prospective, multi-ethnic population-based birth cohort. In a prospective cohort study, a group of people is classified according to a common experience or condition. This group is followed over time and can be compared on several outcomes. The advantage of this design is that various health outcomes stemming from a single exposure can be studied, like for example socioeconomic position. However, this design is only well suited to study common outcomes. Objective as well as subjective oral health measures, like malocclusion, dental caries, subjective treatment need and OHRQoL are very common oral conditions during childhood and are therefore well suited to be studied within cohort studies.

Almost all studies were conducted following a cross -sectional approach. In cross-sectional analysis the exposure and outcome are assessed at the same time, which can make it difficult to determine the time order of events. When the particular outcome affects the supposed exposure, reverse causation cannot be excluded. Sometimes, the relation between outcome and exposure can also be bidirectional. The bias of reverse causation may especially be possible in the study about subjective orthodontic treatment need and OHRQoL as well as the study that's investigates the role of self-esteem in this association, because these are all somehow psychological, self-perceived and subjective measures. Little is known yet on the association between subjective orthodontic treatment need and OHRQoL and also in our study it is not conclusive which direction this association has. Moreover, the answer to the question depends on how subjective orthodontic treatment need (and OHRQoL) is defined. From the perspective of the Wilson and Cleary Model adapted for OHRQoL, subjective orthodontic treatment need and self-esteem determine (or are part of) OHRQoL (1). Moreover self-esteem is a relative stable construct, a personal resource that might help to cope with poor oral conditions (2, 3). Therefore, it is rather unlikely, that OHRQoL with its dynamic, context-specific character is influenced by self-esteem (4). However, it is also likely, that these relationships are bidirectional. Reverse causation is not a likely explanation for the remaining studies presented in this thesis, as the outcome, i.e. children's OHRQoL, nearly impossibly causes objective oral health measures, i.e. objective orthodontic treatment need, malocclusions, dental caries, or a low socioeconomic position of the parents.

### *Bias*

The studies presented in this thesis are all prone to different kind of biases. A bias is a systematic error that affects the result of the study. Biases in observational studies are commonly categorized into selection bias, information bias and confounding.

*Selection bias*

Within population-based cohort studies, selection bias can occur, when the association between the exposure and outcome is different in the included study population and the excluded study population, which was initially eligible to be included. This selection bias can occur either at the start of the cohort study or during follow -up. Even though the initial participation rate of the Generation R Study was relatively high with 61%. This was not random as participants were more often higher educated, more often native Dutch and healthier compared to the source population (5). Still, selection bias only occurs when participation is associated with both the outcome and the exposure. As the studies presented in this thesis study outcomes that occur 10 years after inclusion of participants, initial participation cannot be associated with the outcomes of interest. Therefore, the selection bias due to non-participation in this thesis is considered to be unlikely. Furthermore, studies that describe effects of selective initial participation in similar large birth cohorts showed that associations are only slightly influenced by selection bias due to initial participation and that selection bias due loss to follow up is of major concern (6-8). Selection bias during follow-up of the cohort study is related to study dropout or to non-response to questionnaires and the visits to the focus center. Loss to follow-up in the Generation R Study during the first and second postnatal phases

was relatively low, with follow-up rates over 80% (5). However, the studies presented in this thesis are mainly conducted with data obtained in the third postnatal phase of the Generation R Study, which has a follow-up rate around 70%. But, the main outcome of the presented studies was based on questionnaire data, which non-response was also in the first two phases the major source of missing data. This has unfortunately lead to a higher loss to follow-up for the analysis presented in this thesis. We conducted non-response analyzed to determine differences in characteristics between the included and excluded study subjects. Generally, data on OHRQoL, subjective and objective orthodontic treatment need as well as caries experience were more often missing in children from parents with lower socio-economic status (based on parental education, parental employment, household income and benefit dependency). Similar to the selection bias due to selective initial participation, selection bias due to loss to follow up only occurs when it is related to the outcome and exposure. However, as we do not have information on the outcome in the excluded population, this is problematic to ascertain. Furthermore, we applied multiple imputation of missing data to reduce the risk of selection bias. Thus, the 95% confidence intervals that are presented in this thesis represent the uncertainty associated with the missing values. Hence, we consider selection bias due to non-response unlikely.

Although in a totally different way, also the meta-analysis on the association between malocclusions and OHRQoL is prone to selection bias. Selection bias in meta-analysis and systematic reviews occurs due to selective inclusion of studies. This specific selection bias is called publication bias. However, we tried to include a maximum of studies, independent of study size, methodological quality and dichotomous or continuous analysis. Furthermore, we investigated publication bias visually with funnel plots, which did not give any indication for bias (9).

*Information bias*

The data collected within the Generation R Study and used throughout this thesis were mainly collected in parental questionnaires (OHRQoL, subjective orthodontic treatment need, socioeconomic indicators and ethnic background). Only children's self-esteem was collected within questionnaires directly addressed to the children. Furthermore, data on objective oral health variables were obtained from photographic records. All of these assessment methods are prone to information bias due to measurement error leading to misclassification. Two types of misclassification do exist: non-differential and differential. Non-differential misclassification, or random misclassification, occurs when misclassification of the outcome, exposure or covariate is unrelated to the other variables under study. A non-differential measurement error would have attenuated the results of our studies and is possible to have occurred, for example due to data entry mistakes in the objective oral health data. In general, the assessment of objective oral health measures from photographic records might be limited and causing a non-differential measurement error, since oral examination is the gold standard. However, intra-oral and extra-oral photographs, in combination with 3D-pictures and radiographs have been validated to be suitable for scientific studies (10, 11).

Differential misclassification is present when misclassification of the outcome, exposure or covariate is unrelated to the other variables under study. For example, if answering

the items of the OHRQoL questionnaire has influenced the answers to the questions about subjective orthodontic treatment need or if socioeconomically deprived participants were less able to understand the OHRQoL items, than differential misclassification might have occurred, leading to an under- or overestimation of the true associations. However, little is known about the influence of low socioeconomic position on the ability to answer questionnaires or the influence of previous questions on answering other questions. Therefore, a differential misclassification error in the presented studies is difficult to ascertain.

*Confounding*
As previously mentioned, confounding is a major threat to observational studies. Confounding variables are factors that blur the associations between exposure and outcome variables. The effect of confounding is unpredictable as it can strengthen or attenuate associations or can even make it dubious. To be a confounding variable, a covariate has to meet following requirements: it needs to be associated with the outcome, it needs to be associated with the exposure and it is not an intermediary factor, which means it should not be on the causal pathway between exposure and outcome (12). In all studies presented in this thesis we adjusted the analysis for potential confounders. The selection of confounding variables was based on the previous literature, associations with the outcome or exposure, or due to a change in the effect estimate of 10% (13). Still, residual confounding might be present in our analyses, as it is generally possible in observational studies, for example by unmeasured oral health variables.

## *Measurements*

*OHRQoL*
*OHRQoL is defined as "*a multidimensional construct that reflects (among other things) people's comfort when eating, sleeping, and engaging in social interaction; their self-esteem; and their satisfaction with respect to their oral health" (14). Many different measures are developed to assess children's OHRQoL. The assessment of OHRQoL in the Generation R Study was done using the COHIP-ortho, described in chapter 2.1. Although this measure has been validated, there were limitations to this questionnaire. The major issue is, that children's OHRQoL was assessed in parental questionnaires. Accordance between parental and child reports in the assessment of OHRQoL has widely been described in the literature and especially in orthodontics parents are seen as valid proxies for the assessment of their children's OHRQoL (15-18). Generally, when discrepancies between parents and child reports were found, children tend to report poorer OHRQoL than expected by the parents. In this case, the associations presented in this thesis would rather be underestimated than overestimated.
In OHRQoL research it is important to question whether we measure what we want to measure. The multidimensional, dynamic, context-specific character of OHRQoL challenges this question (4). Although research on OHRQoL has enormously increased in the recent years, there is still no consensus on what OHRQoL exactly is (19). A good framework to understand influences on and parts of OHRQoL is given by the Wilson and Cleary Model that incorporates biological, social, psychological and cultural factors (1,

20). Also, the OHRQoL questionnaire should include items on all of these different factors. The basis for the COHIP-ortho was its original 38-item version. The development of the original COHIP has extensively been described in the literature (21). In addition, it has been validated in various oral health patient groups as well as the general population (22, 23). The COHIP covers among others all oral health domains relevant to OHRQoL as described by the Wilson and Cleary Model: oral health (biological and physiological variables), functional well-being, social-emotional wellbeing and self-image (oral health perception).
Another question is whether the different items in an OHRQoL questionnaire should be differently weighted, as biological, social, psychological and cultural factors could influence OHRQoL with different impact. In this case, weighting items could improve the psychometric properties of the questionnaire. The studies presented in this thesis make no use of weighted items. Weighted items were shown to improve particular OHRQoL measures (24). However, whether weighting of items would have improved the COHIP-ortho is unclear. Other studies in turn indicated that self-weighting of items did not improve the performance in terms of validity and reliability of overall scores from OHRQoL questionnaires (25).

*Subjective orthodontic treatment need*
Within in the Generation R Study, subjective orthodontic treatment need was assessed in parental questionnaires with the question: "Do you think you child needs braces?" This question was answered on a five point Likert-scale from strongly agree to strongly disagree. This assessment method of subjective orthodontic treatment need has two major drawbacks. First, we do not ask the children themselves but again ask the parents, which might have led to information bias. Second, we did not consider whether the children already might have orthodontic treatment, which definitely would have influenced the answer to this question. Still, in the Netherlands it is rather uncommon that children start their orthodontic treatment before the age of 10. Besides these limitations, our approach has also advantages to the approaches of other studies. Different studies have used OHRQoL as a surrogate for subjective treatment need (26, 27). From the perspective of the Wilson and Cleary Model, where health perceptions are only part or one of the determinants of OHRQoL, this is wrong. Other studies have used aesthetics as surrogate for subjective orthodontic treatment need (26-28). But also aesthetics are limited in their ability to reflect subjective orthodontic treatment need, as aesthetics and perceived treatment need are not one-to-one related (27, 29).

*Objective oral health measures*
Objective oral health measures collected within the Generation R Study and used in the present thesis include malocclusions and dental caries.
Malocclusions were assessed following the Index of Orthodontic Treatment Need (IOTN) that is composed of the dental health component (DHC) and the aesthetic component (AC) (30). Using this index enables a systematic, accurate and credible data collection of orthodontic information. The IOTN is a valuable tool for scientific studies as it only measures the worst feature of the occlusion and intra-examiner as well as inter-examiner variabilities are low (31-33). However, instead of applying this index during

oral examination or on plaster casts, it was applied to photographic records, including 2D pictures, 3D pictures and radiographs (chapter 2.2). Although this approach was shown to be appropriate for scientific studies and the sensitivity as well as specificity of assessing orthodontic need was satisfactory, it is possible that this approach has induced some non-differential measurement error. The assessment of orthodontic treatment need with the IOTN has some limitations, which are not related to the photographic approach exclusively. First, certain occlusal traits reduce the reliability of the Index, because these are more difficult to ascertain (34). Second, it is necessary to achieve the correct patient positioning to evaluate the occlusion correctly (35). Finally, the sufficient removal of saliva from the teeth is necessary to prevent underexposure and unwanted light reflections (35). Of course, because the IOTN data collection was embedded in a large scale cohort study, the Generation R Study, for logistic reasons we were not able control continuously for this factors, which additionally might have led to some misclassification of orthodontic treatment need. Despite these concerns, this appeared to be the most appropriate approach to assess orthodontic treatment need in a large scale cohort study.
Dental caries was assessed with the decayed, missing and filled teeth Index (dmft) (36). Also this Index was assessed from intraoral photographs instead of direct oral examination as described in other studies (10). Scoring dental caries per tooth on intraoral photographs was shown to have a high sensitivity and specificity (10). Of course compared to the gold standard (oral examination) this approach is limited, although suitable for scientific studies (10, 37). Thus, the assessment of the dmft score in this study might be affected by non-differential measurement error leading to an underestimation of the associations between caries experience and OHRQoL.

*Socioeconomic position*
With socioeconomic position the "social and economic factors that influence which positions individuals or groups hold within the structure of a society" are meant (38). It is a complex, multidimensional construct which is commonly captured in health research by different indicators from various levels, i.e. individual level, household level and neighborhood level (39-41). Which indicators are chosen should be based on the research question (39-41). As we were interested in the general association between family SEP and children's OHRQoL, we used different indicators. As children however are difficult to be classified according to the commonly used socioeconomic indicators, they were classified based on their parents socioeconomic position. Consequently, we call children's socioeconomic position "family socioeconomic position". In chapter 4.1 we used the following indicators to depict family socioeconomic position: parental education level, parental employment status, net household income, benefit dependency, single parenting and ethnicity. The first three indicators are traditional socioeconomic indicators in health research, but we added the others because these are socioeconomic indicators which were associated with oral health research in previous studies (40-42). Using many different socioeconomic indicators could have led to different associations with OHRQoL, however this did not happen in the study on social inequalities in children's OHRQoL. The different indicators were correlated but also reflect the multiple dimensions of the complex construct of socioeconomic position. Still the strength of

associations between socioeconomic indicators and OHRQoL slightly varied, which is why selected only certain indicators to adjust the analyses for the effects of socioeconomic position in chapter 3.2, chapter 3.3, chapter 5.1 and chapter 5.2. First, we used level of education. Level of education is relatively easy to measure, relatively stable over time and an appropriate indicator for socioeconomic position in The Netherlands (43). Still, information on income is a more sensitive issue and might be prone to a higher non-response. As in contrast to educational level, income is a less stable variable, therefore we used the information obtained most closely to the assessment of OHRQoL, thus also assessed at children's age of 10. However, it is suggested that educational level influences oral health and oral health behavior through knowledge, skills and acceptance of oral health education (39-42). Second, we used household income. This indicator reflects, other than education level, the material resources of the family. With regard to dentistry, and especially orthodontics, material resources of the family might have a major impact on the access to care, because in the Netherlands dental insurance cannot be taken for granted. General pediatric dental care is generally covered, however for parents is not. If parents do not make use of dental care, their children will also be less likely to do so. In addition, orthodontic care in the Netherlands is only covered in certain circumstances, depending on the severity of malocclusion and the individual insurance package.

*Ethnicity*

In the studies presented in this thesis, we defined ethnicity based on the country of birth, which is the standard classification employed by Statistics Netherlands (44, 45). As ethnicity is a complex concept, with several definitions based on shared origins, culture, traditions, language or religious traditions and also linking geographical areas, ethnicity can also be measured in other ways, for example genetically based or by self-identification (45, 46).

In the present thesis, a child was considered non-native Dutch if at least one of the parents was born abroad (47). When both parents were born abroad, the ethnic background of the mother determined ethnicity of the child, because mothers are most often the primary caregivers and we aimed to take the cultural background into account. In chapter 5.1 we were able to distinguish between four ethnic groups, i.e. native Dutch, Turkish, Moroccan and Surinamese. These ethnicities represent the largest ethnic minority groups in the Netherlands (48). We did not distinguish between first, second or third generation immigrants, which might have classified people into one group, although they feel different in culture and ethnic identity. In chapter 3.2 and 5.2 we adjusted our analysis for ethnicity instead of investigating ethnic differences. In these studies, we only distinguished between non-native and native children.

Ethnicity and socioeconomic position are related and therefore should be mutually adjusted for in the analyses (49, 50). However, correlation between ethnicity and the different socioeconomic indicators was not higher than the correlation among the different socioeconomic indicators. Therefore, we considered ethnicity as a socioeconomic indicator in chapter 5.1. In oral health research socio-demographics are often combined socio-economic indicators and have been shown to be similar related to oral health outcomes (42, 51).

*Self-esteem*

Self-esteem is described as the self-perceived ability to deal with the environment and is highly influenced by interaction with others, nevertheless it is considered to be a stable construct (3, 52). It is a psychological resource that might protect from the effects of deleterious oral health conditions (4). Self-esteem can be measured indirect and directly (53, 54). Indirect measures have the advantage, that study participants are not aware of the process of assessment. Direct measures are prone to information bias, because respondents might answer social desirable (55, 56). Still, direct measures are more commonly used as they are easier to apply especially within large scale cohort studies. For the study in Chapter 5.3 self-esteem was assessed directly.

Self-esteem was the only variable in the Generation R Study and used in this thesis, which was assessed in questionnaires directly sent to the children. Self-esteem was assessed with a modified version of the Harter's self-perception profile. The adaption was based on Wichstrom's (1995) suggestions, but applied to the validated self-perception profile for children (57, 58). Next to the deletion of some redundant items, we changed the answer coding of the items from a four point coding into a three point coding to increase the variability of item scores (59). For this adapted scale no clinically important differences have been established (58). This made it difficult to interpret the obtained data. Therefore, we investigated differences in oral health between children with rather low or rather high self-esteem, based on upper 20% cut-off within the study population.

## *Statistical analysis*

*Assessing determinants of OHRQoL*

In chapter 3.1, chapter 3.2, chapter 3.3, chapter 4.1 and chapter 5.2 determinants of OHRQoL were assessed. OHRQoL is a continuous variable, however simply building ordinary linear regression models with OHRQoL as outcome variable is not possible. To build linear regression models the following assumptions need to be fulfilled: linearity of the outcome variable related to the predictor, normality of the sampling distribution and the residuals in a population, homogeneity of variances across all levels of the predictor and independence of the observations (60). In evaluating the association of biological/physiological factors as well as individual factors and OHRQoL, we assumed that OHRQoL was inversely linear related to all predictors. We also confirmed this assumption with trend analyses, by treating the predictor variables as a continuous term. Also, we considered all the observations on OHRQoL as independent. The major threat to the independency of observation in the studies presented in this thesis is, that we did not exclude twins from the analysis. However, we consider the dependence in OHRQoL between twins as unlikely and moreover the number of twins in our study population was very limited (3.0%). Although OHRQoL is not a normally distributed variable and also the residuals in the population were not perfectly normal distributed, the assumption of normality was not the major threat to the models with OHRQoL as outcome variable, because of the central limit theorem. This theorem means that there are a variety of situations in which we can assume normality regardless of the shape of our sample data. One of these situations is the case of large sample, which we definitely

had with sample sizes varying between 2419 and 3796 children in the studies presented in this thesis. However, we could not fulfill the assumption of homoscedasticity. We approached this potential bias in two ways. In chapter 3.2, chapter 4.1, chapter 5.1 and chapter 5.2 we used weighted least squares linear regression models and in chapter 3.3 we categorized OHRQoL into quartiles and built logistic regression models.

*Missing data*

A general concern in large scale observational studies is the large amount of missing data. Different patterns of missing data can occur in research: information might missing completely at random, at random or not at random. Because it is difficult to ascertain which missing value pattern is present, the choice how to deal with the missing data in the studies presented in this thesis was based on assumptions (61).We considered the patterns of missing values in all studies to be missing at random, which means that the missing of values was related to variables that are assessed in the study and included into the statistical models. Generally data were more often missing in participants with unfavorable socioeconomic status. The recommended method to deal with data missing at random is the multiple imputation method (61, 62). There are different suggestions how often a dataset should be imputed to obtain a valid pooled estimate. We imputed 5 (chapter 3.3) or 10 times (chapter 3.2, chapter 4.1, chapter 5.1, chapter 5.2), depending on whether the pooled estimate did not changed with more imputations. The imputations were based on the relations between all variables in the model, however main determinants and outcomes were generally not imputed.

## Interpretation and possible mechanisms of the findings

### *Associations between objective and subjective oral health measures with OHRQoL*

An important aim of the thesis was to investigate the relation between oral health conditions, more precisely malocclusions, and OHRQoL. The studies presented in this thesis showed that all biological/physiological variables, or clinical variables, assessed were related to children's OHRQoL. The meta-analysis in chapter 3.1 revealed that malocclusions were associated with poorer children's OHRQoL, however also showed that this relationship is very variable among different studies. As OHRQoL is considered a multidimensional, dynamic concept, this was already suggested by others (20). The negative influence of malocclusions on OHRQoL has several underlying understandings. The first one is pain related to temporomandibular dysfunction, which is less likely in children, and dental traumas, which is more likely in children (63-65). Second, malocclusion can cause functional limitation, like chewing and speaking problems (65, 66). Pain represents the symptoms status and functional problems represent the functional status in the Wilson and Cleary Model via which malocclusions are suggested to influence OHRQoL. However recently, researchers report more often about the emotional influences of malocclusions on OHRQoL, including laughing, feeling ashamed or having reduced self-esteem (65, 67-69). Emotional status is not directly a factor in the Wilson and Cleary model (1). Still, oral symptoms, functional well-being and emotional wellbe-

ing are domains of OHRQoL. Unfortunately we could not investigate on which specific OHRQoL domain malocclusion act. But the influence of self-esteem, as emotional factor, or individual factor following the Wilson and Cleary Model, is discussed in the subsequent paragraph.

Caries is the most important preventable oral disease during childhood, with a prevalence of 60- 90 % worldwide (70). Also dental caries was associated with poorer OHRQoL (chapter 3.3). Several other studies have shown the relationship between caries and children's OHRQoL in cross-sectional studies, however we showed a longitudinal influence of dental caries on children's OHRQoL (71-74). The longitudinal relationship between dental caries and children's OHRQoL has two theoretical explanations. First, caries experience at a young age is a strong predictor for later caries (75-77). Second, children remembering severe dental caries (treatment) remain to perceive poorer OHRQoL. Most likely it is a combination of both, which would be represented in the Wilson and Cleary Model by biological/physiological status and subjective oral health.

The subjective oral health measure investigated in this thesis was subjective orthodontic treatment need (chapter 3.2). More self-perceived subjective orthodontic treatment need was associated with poorer OHRQoL independent of their occlusion. To our knowledge, no comparable studies have been conducted. However, when subjective orthodontic treatment need is related to dental aesthetics, our findings are in line with studies investigating the relationship between dental aesthetics and OHRQoL (26, 27, 29, 78). Indeed, our findings indicated that the association between subjective orthodontic treatment need and OHRQoL is rather modified by aesthetic impairments than by dental health impairments. This can be an explanation for the observation that subjective and objective orthodontic treatment need often differ (79). Still, aesthetics are limited in their ability to assess subjective orthodontic treatment need (27).

### *Influences on the relationship between objective and subjective orthodontic treatment need and OHRQoL*

A second important aim of the thesis was to investigate the influence of non-clinical variables on OHRQoL. In chapter 4.1, we showed that environmental characteristics of the child, i.e. family socioeconomic position significantly influenced children's OHRQoL. Many studies suggest that the influence of low socioeconomic position on oral health and OHRQoL is due to unfavorable oral health knowledge and oral health behavior (80). Although we were not able to test this, we suggest that there is a multidimensional influence of socioeconomic position on children's OHRQoL. Based on our results, maternal education level, which might correlate with oral health knowledge and oral behavior the most, was less associated with OHRQoL than parental employment status and household income, which might be more correlated with dental insurance and resources for dental treatment (81, 82). Furthermore, the effect of socioeconomic position might also be mediated by characteristics of the individual.

In chapter 3.1, chapter 3.2, chapter 5.1 and chapter 5.2, we investigated how individual characteristics of the child, i.e. age, gender, ethnicity/cultural differences and self-esteem are related to children's OHRQoL. In the systematic review, children's age was shown to have a major influence on the association between malocclusions and OHRQoL. The older the children were the more their malocclusion affected their

OHRQoL (83). Also, in other studies age emerged consistently as determinant of OHRQoL, however whether this relationship is consistent at all ages remains unclear (74, 84-86). Differences for gender were seen in the study in subjective orthodontic treatment need and OHRQoL. Generally, it is suggested that girls perceive lower OHRQoL than boys, however research on gender differences in oral health is very limited (26, 29). Our research did not allow to conclude whether self-perceived orthodontic treatment need affects OHRQoL stronger in girls than in boys, still we saw a significant difference in gender. In line with other studies, girls might be more conscious about their appearance, whereas boys might be more aware of their malocclusion (29, 87). Likely, age plays a role for gender differences in OHRQoL research as well. Furthermore, we saw that the relationship between malocclusions and children's OHRQoL varied among different countries, which we interpreted as cultural differences. The influence of malocclusions on OHRQoL could be influenced by local health care systems (88). But moreover, cultural differences in OHRQoL should be expected considering the World Health Organization's definition of quality of life: '[quality of life is the] individual's perception of his/her position in life in the context of culture and value systems in which they live […] (88). In line, we showed ethnic differences in subjective orthodontic treatment need and OHRQoL. Other studies also reported ethnic differences in OHRQoL might also partly be due to differences on socioeconomic status (42). Furthermore, also self-esteem might mediate the ethnic differences in oral health variables (67, 90-92). In concordance with other research, we found that self-esteem was significantly related with OHRQoL (67, 93, 94). Different to the relationship between caries and OHRQoL, self-esteem did not mediate the association between malocclusions and OHRQoL (2, 67, 95, 96). However, we showed that self-esteem did modify the relationship between subjective orthodontic treatment need and OHRQoL. Children with low self-esteem might suffer more from their malocclusion (4). Still, the direct influence of self-esteem on OHRQoL might be more related to other oral health fields than orthodontics (2, 67, 94-97). In general, like proposed by the Wilson Cleary Model adapted for OHRQoL, all non-clinical factors seem to be interrelated in their influence on OHRQoL.

### Implications for patients, clinicians, researchers and policymakers

Research and interventions in oral health are of major public health interest. First, because the need for oral health treatment in children, like orthodontics and other health problems, is highly prevalent in western countries. In the Netherlands, around 70% of the young adults have had orthodontic treatment and also the prevalence of other oral diseases among children is not negligible, especially because most of them should be preventable (85, 98). Second, in oral health treatment, especially orthodontics, high costs are involved. The evidence from this present thesis should be used to inform social policies and programs explicitly by considering characteristics of the individual and its environment as mechanisms for enhancing oral health and OHRQoL of the population.

The results of this thesis promote a more active role of the patient in oral health care. All chapters highlight the importance of OHRQoL, either to replace or to complement conventional objective oral health measures in clinical practice as well as oral health research. Oral health needs from the perspective of patients appear increasingly im-

portant, because patients and professionals judgements of health care needs differ. For example, whereas orthodontic treatment provision nowadays is largely based on functional status or oral symptoms, the findings from chapter 3.2 suggest that self-perceived problems like avoiding to smile or to speak might particularly be associated with lower OHRQoL and thus should rather be considered in decisions on orthodontic treatment need.

Furthermore, the studies presented in this thesis highlight the importance to promote good oral health during childhood, because those who get a compromised start to oral health are much more likely to follow a trajectory which will lead to poor oral health (-related quality of life) later. The development of oral health disparities, due to for example socio-economic or ethnic disadvantage, starts already during childhood and tends to persist and increase through life and needs therefore to be tackled as early as possible (99).

Research on the different determinants of OHRQoL and the effect of these determinants on the relationships between clinical variables and OHRQoL, as conducted in this thesis, also supports an effective communication between oral health care provider and the patient. For example, the findings in chapter 3.2 suggest that girls and boys might be different approached in orthodontic practice to improve compliance with the treatment.

Still, because the impact of oral diseases on OHRQoL are not consistent, different determinants on oral health need to be understood in order to make on the one hand correct treatment decisions, on the other hand to develop effective oral health education, oral health promotion and policy decisions. A few of these factors were identified in the present thesis and will help to design proper targeted multicomponent interventions. These interventions to improve children's oral health, OHRQoL and oral health care should generally target on many different factors. It is known from other public health areas, that those multicomponent interventions are most effective (100). In summary, the studies presented in this thesis are relevant to orthodontics, oral epidemiology and community dentistry. Still, they also build a basis for further research on orthodontic and other oral health care topics.

### Unanswered questions for future research

We noticed the increasing research interest in the modifying role of non-clinical factors in the association between subjective and objective oral health measures with OHRQoL. The theoretical model by Wilson and Cleary (1995), that guided the presented research, helped to identify variables that determine objective and subjective oral health. We recommend this model for further research that might guide, develop and evaluate oral health promotion interventions.

In the present thesis we investigated different non-clinical factors in the association between subjective and objective oral health measures with children's OHRQoL. However, we only looked at a limited number of environmental characteristics (socioeconomic indicators) and individual characteristics (age, gender, ethnic background, self-esteem). The relationship between further environmental characteristics, like influences of the family, friends or healthcare providers and the home, neighborhood or school setting, and children's OHRQoL should be researched as well. Also further individual

characteristics that possible influence children's OHRQoL, like social support, perceived stress and personality traits such as sense of coherence, should be studied. Finally, these studies could investigate many different factors influencing OHRQoL simultaneously by using structural equation modelling as the primary analytical approach.
While we investigated the relationship between different socioeconomic indicators and OHRQoL, we still don't know the pathways of socioeconomic inequalities in children's OHRQoL. The same accounts for ethnic or cultural inequalities in children's OHRQoL. Furthermore, we discovered that gender based differences are rarely addressed in oral health research. Because we found gender differences in children's OHRQoL we highly recommend more attention to this topic. Finally, the work presented in this thesis indicated that future studies need to focus on both cross-sectional and longitudinal effects of oral health variables on OHRQoL separately as well as simultaneously. Then, not only the impact of oral diseases are of interest, but also the impact of dental treatments, like different kinds of braces, should be studied with regard to future subjective and objective oral health measures. This can be realized by studies with longer follow up periods and repeated oral health measurements within different age groups.

## General conclusion

Based on the findings presented in this thesis it can be concluded that, the COHIP-ortho is a good questionnaire to be used in research on children's OHRQoL, especially related to orthodontics. In dental practice, this instrument can easily be combined with objective measures and in epidemiological studies it can be integrated into health surveys. To determine orthodontic treatment need with the IOTN in scientific studies, the use of 2D and 3D pictures are suitable. However, the assessment of orthodontic treatment need is much better when 2D and 3D pictures are combined with radiographs. Subsequently, both methods can contribute to an increase in the knowledge of patient's perspective on oral health and related well-being obtained from large scale epidemiologic studies.
Objective oral health measures, like malocclusions and caries experience, have a significant albeit small impact on children's OHRQoL. The impacts of these clinical, or biological/physiological, variables might not only occur on the short-term, but impacts on OHRQoL might also occur on the long-term. In addition, children with self-perceived orthodontic treatment need experience poorer OHRQoL independent of their objective oral health measures.
Finally, also non-clinical variables, like characteristics of the environment and characteristics of the individual affect OHRQoL as well as the relation between objective oral health measures, subjective oral health measures and OHRQoL. First, girls perceived more impact of malocclusions on OHRQoL than boys and generally the impact of malocclusions on OHRQoL increased with age. Second, children from low family socioeconomic position and non-native ethnic background have poorer OHRQoL. In addition, the associations between objective and subjective orthodontic treatment need with OHRQoL was different between native Dutch children and children with an ethnic minority background. Third, children's self-esteem modifies the relation between subjective oral health measures, i.e. self-perceived orthodontic treatment need, and their OHRQoL. Future research should continue to bring us closer to the exact relations between children's OHRQoL, subjective and objective (orthodontic) oral health measures.

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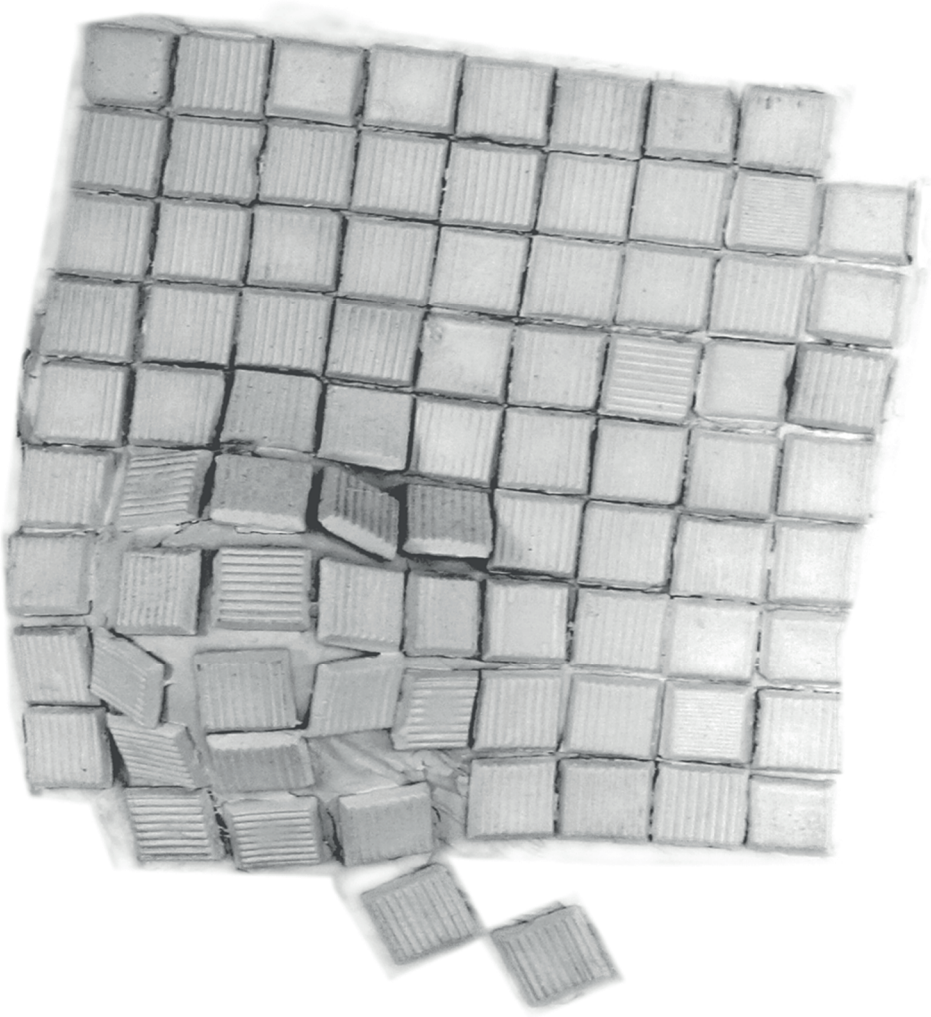
# Chapter 7
Summary



## Summary

In **chapter 1** we gave an introduction to the research presented in this thesis. Oral health can be measured objectively and subjectively. However, subjective and objective (orthodontic) oral health measures are not concurrent. The concept of oral health-related quality of life (OHRQoL) was introduced in the dental literature, basically because OHRQoL aims to capture subjective oral health in an objective way. OHRQoL has been defined as "*a multidimensional construct that reflects among other things peoples comfort when eating, sleeping and engaging in social interaction; their satisfaction with respect to their oral health".* Whether OHRQoL cannot simply explain the inconclusive relation between subjective and objective oral health measures is still unclear. However, OHRQoL is very suitable to measure (the perceived) oral health of individuals. It is particular important to study OHRQoL as well as subjective and objective oral health measures related to the orthodontic field in children. First, because the majority of the orthodontic patient groups are children and second, oral health during childhood is strongly related to oral health in later life and thus needs to be addressed as early as possible. Therefore we aimed in this thesis to understand the relationships among measures of objective oral health outcomes, subjective oral health outcomes and OHRQoL specifically related to the orthodontic field by investigating biological/ physiological determinants, environmental determinants and individual determinants of children's OHRQoL.

In **chapter 2** we validated two methods for large-scale epidemiological research on children's OHRQoL, in particular for the orthodontic field.

We presented a shortened OHRQoL measure, which was derived from the Child Oral Health Impact Profile (COHIP). The use of OHRQoL measures in research and clinical practices has been limited by their length involving a lot of time and carrying a large response burden. We performed a cross-sectional study to compare OHRQoL data obtained with the original COHIP, with 38 items, and OHRQoL data obtained with the COHIP-ortho, with 11 questions. We showed that the performance of the COHIP-ortho was comparable with the COHIP regarding the construct and criterion validity, score distributions and internal validity. The COHIP-ortho has been used throughout the thesis as OHRQoL measure.

The other validation concerned the use of photographic records, i.e. 3D extra-oral pictures, 2D extra-oral photos, lateral cephalograms and orthopantomograms to assess the Index of Orthodontic Treatment Need (IOTN) in children. The IOTN is a commonly used occlusal index to obtain systematic, accurate and credible orthodontic data. Currently, oral examination and plaster casts are seen as the gold standard to assess the IOTN. However both have disadvantages as they are time-consuming and requiring a lot of storage place. In this study we assessed the IOTN first on plaster casts, second on a combination of 2D-photos and 3D-pictures and third on a combination of 2D-pictures, 3D-pictures and radiographs. After assessing agreement of the different methods it appeared that a combination between 2D and 3D-pictures can be used to assess orthodontic treatment need in general terms, but that the individual IOTN grades have only been sufficiently assessed when 2D and 3D-pictures were combined with radiographs.

Throughout the rest of this thesis we use the latter method to assess the IOTN in children.

In chapter 3 we assessed the relationship between different biological/ physiological variables, thus objective oral health measures, and subjective oral health measures with children's OHRQoL within the Generation R Study.

We started with a systematic review and meta-analysis of the literature on the impact of malocclusions on children's OHRQoL. The pooled results show that there was a clear inverse association of malocclusion with OHRQoL, although the impact children perceived was small. Furthermore, we saw that the effect of malocclusions on OHRQoL varied by the age of children and the country of study conduction. However, this evidence was solely based on cross-sectional studies. Further research is needed to explain remaining heterogeneity in the association of malocclusion with OHRQoL and to evaluate the causality of the association.

In the next study we described the association of subjective orthodontic treatment need and children's OHRQoL. Subjective orthodontic treatment need was assessed in parental questionnaires with the question "Do you think your child needs braces? Based on the answer to this question children had no subjective need, borderline subjective need and definite subjective need. From linear regression models we concluded that OHRQoL is poorer in children with self-perceived orthodontic need. Furthermore, we saw that the association between definite subjective treatment need and OHRQoL was stronger in girls, whereas the association between borderline subjective orthodontic treatment need and OHRQoL was stronger in boys. Further research is needed to prove the role of these individual factors on the association between subjective oral health measures and OHRQoL.

Then we assessed the relation between dental caries at children's age of 6 with OHRQoL at the children's age of 10. Caries is an objective oral health measure that influence influences OHRQoL and might therefore interfere in the relationship between subjective and objective orthodontic measures with OHRQoL. Caries experience was assessed with the diseased, missed and filled teeth index. With multinomial logistic regression analysis we showed that children with severe caries at the age of 6 were significantly more likely to have lower OHRQoL at the age of 10 than children without caries experience at the age of 6, however actual differences in OHRQoL among different caries severity groups was rather small. We explained the relationship between caries experience and later OHRQoL, firstly with the fact that caries experience is a strong predictor for later caries experience and secondly with children's memory on pain and treatment having a lasting impact on OHRQoL. Further research is needed to evaluate the background and causality of the association between early caries and later OHRQoL.

In chapter 4 we evaluated the associations of environmental characteristics with children's OHRQoL within the Generation R Study.

One environmental characteristic relates to the social inequalities in children's OHRQoL. Because it is difficult to assess children's socioeconomic status directly we measured their family socioeconomic position (SEP) with the following indicators: maternal and paternal education level, maternal and paternal employment status, net household income, receiving benefits and single parenting. With linear regression analysis we

showed that all family SEP indicators were consistently associated with OHRQoL. Moreover, we showed that this association remained independent of their objective oral health status. Given these disparities, interventions and policies promoting good oral health and wellbeing should target children from low socioeconomic position. Still, more research is needed to understand the pathways of social inequalities in children's OHRQoL.

In chapter 5 we aimed to assess the associations between individual characteristics and children's OHRQoL within the Generation R Study.

First, we evaluated differences among children with different ethnic backgrounds in the associations between objective and subjective orthodontic treatment need related to their OHRQoL. It is generally known that ethnic minorities are at a disadvantage with regard to oral health. Still, children with Turkish, Moroccan or Surinamese background had similar orthodontic treatment need as native Dutch children. However, the association between subjective and objective orthodontic treatment need was much weaker in children with ethnic background than in native Dutch children. Still, OHRQoL was the highest in Dutch children, supported by a stronger impact of subjective and objective orthodontic treatment need on OHRQoL in children with ethnic background. Based on the results more attention should be payed to ethnic differences in clinical practice, however further research is needed to understand the pathways underlying ethnic differences in subjective and objective oral health.

Furthermore, we aimed to describe the role of self-esteem in the relationship between subjective orthodontic treatment need and OHRQoL. Self-esteem is a personal resource that facilitates coping with less favorable conditions and was measured with a modified version of the Harter's self-perception profile. With linear regression analysis we revealed that self-esteem had no confounding or mediating role in the association between subjective orthodontic treatment need and OHRQoL. However, self-esteem modified the relationship between subjective orthodontic treatment need and OHRQoL, as this association was different between children with high and children with low self-esteem. Work still needs to be done to understand and explain the role of self-esteem for OHRQoL, as such as well as in relation to oral health perceptions.

In chapter 6, a general discussion regarding the studies combined in this thesis was presented. This discussion started with a summary of the principal findings of the different chapters followed by the discussion of the strength and limitations of the conducted research, considering the methods with regard to design (observational, mostly cross-sectional), measurements (parental reports, no direct oral examinations) and analyses (particularities of OHRQoL data, missing data). After evaluating strength and limitations of the presented findings, we provide interpretations and possible mechanisms of the findings. Subsequently, we discussed the possible implications of our findings for patients, clinicians, researchers and policymakers. Finally, we presented unanswered questions for future research on subjective and objective oral health measure as well as OHRQoL in children. The discussion finished with the following conclusions based on this thesis: First, the COHIP-ortho and the assessment of orthodontic treatment need on photographic and radiographic records are valuable methods for large-scale oral health research with low response burden for participants. Second, objective oral health measures have a significant albeit small impact on children's OHRQoL. Self-

perceived oral health has a similar impact on children's OHRQoL. Third, children from low socioeconomic position have poorer OHRQoL. Fourth, individual characteristics of children, like gender, age, ethnicity and self-esteem are determinants of children's OHRQoL, and influence the associations between subjective and objective (orthodontic) oral health measures.

## Samenvatting

In **hoofdstuk 1** hebben wij een introductie van het onderzoek in dit proefschrift gegeven. Mondgezondheid kan worden gemeten vanuit het perspectief van de zorgverlener (objectief) en vanuit het perspectief van de patiënt (subjectief). Echter, objectieve en subjectieve mondgezondheid komen niet goed overeen. Het concept mondgezondheidsgerelateerde levenskwaliteit (OHRQoL) is geïntroduceerd in de tandheelkundige literatuur, voornamelijk omdat OHRQoL er naar streeft subjectieve mondgezondheid vast te leggen op een objectieve manier. OHRQoL is gedefinieerd als "een multidimensionaal construct dat onder meer het comfort tijdens het eten, slapen en in interactie met anderen reflecteert; dus de tevredenheid ten opzichte van de mondgezondheid." Of OHRQoL de zwakke relatie tussen subjectieve en objectieve mondgezondheid eenvoudig kan verklaren is nog onduidelijk. Maar OHRQoL is ook geschikt om de (zelfervaren) mondgezondheid te meten. Als het om orthodontie gaat, is het bijzonder belangrijk om OHRQoL alsook objectieve en subjectieve mondgezondheidmaten bij kinderen te meten. In eerste plaats, omdat de meeste orthodontiepatiënten kinderen zijn. Daarnaast, omdat mondgezondheid tijdens de kindertijd sterk gerelateerd is aan de mondgezondheid in het latere leven en daarom zo vroeg mogelijk moet worden aangepakt.
Daarom richt dit proefschrift zich erop de relaties tussen maten voor objectieve mondgezondheid, subjectieve mondgezondheid en OHRQoL, in het bijzonder in verband met orthodontie, in kaart te brengen. Dit is gedaan door biologische/ fysiologische determinanten, determinanten van de omgeving en individuele determinanten van OHRQoL in kinderen te bestuderen.
In **hoofdstuk 2** hebben wij twee methoden voor grootschalig onderzoek naar OHRQoL en orthodontische behandelbehoefte bij kinderen gevalideerd.
Wij presenteren een ingekorte OHRQoL-vragenlijst, die afkomstig is van de "Child Oral Health Impact Profile" (COHIP). Tot nu is het gebruik van vragenlijsten gericht op OHRQoL in de praktijk erg beperkt geweest door de lengte, de tijd die het kost om ze in te vullen en de dusdanige belasting voor de respondent. Wij hebben een cross-sectionele studie uitgevoerd om gegevens over OHRQoL, verkregen met de originele COHIP (38 vragen), te vergelijken met de gegevens verkregen door de ingekorte COHIP-ortho (11 vragen). Wij hebben laten zien dat de resultaten van de COHIP-ortho en de COHIP-38 vergelijkbaar zijn op het gebied van construct- en criteriumvaliditeit, verdeling van de punten en de interne validiteit. In dit proefschrift wordt verder alleen de COHIP-ortho gebruikt om OHRQoL bij kinderen te meten. Hierna valideerden wij het gebruik van foto's, dat wil zeggen 3D-foto's, 2D-foto's en röntgenopnames, om de "Index of Orthodontic Treatment Need" (IOTN) bij kinderen vast te stellen. De IOTN is een occlusie-index om systematische, accurate en betrouwbare orthodontische gegevens te verkrijgen. Tot nu waren gebitsafdrukken en het onderzoek direct in de mond de gouden standaard om de IOTN te meten. Echter, beide manieren hebben nadelen omdat zij veel tijd kosten of veel opslagruimte vragen. In deze studie hebben wij de IOTN eerst op gebitsmodellen, dan op een combinatie van 2D- en 3D-fotos en vervolgens op een combinatie van 2D-, 3D- en röntgenfoto's gemeten. Nadat we de overeenkomst tussen deze drie manieren hebben bekeken, blijkt dat de 2D- en 3D- foto's voldoende zijn om een algemene orthodontische behandelbehoefte vast te stellen, maar ook kan de IOTN

alleen goed beoordeeld worden als 2D- en 3D-foto's worden gecombineerd met röntgenfoto's. In dit proefschrift wordt verder alleen een combinatie van 2D-, 3D- en röntgenfoto's gebruikt om de orthodontische behandelingsbehoefte bij kinderen te meten.
In **hoofdstuk 3** schatten wij de relatie tussen verschillende biologische en fysiologische variabelen, dat wil zeggen objectieve mondgezondheidsmaten, en subjectieve gezondheidsmaten met OHRQoL van kinderen in de Generation R Study.
Wij hebben een systematische literatuurstudie en meta-analyse over de impact van malocclusies op OHRQoL van kinderen uitgevoerd. De gecombineerde resultaten laten zien dat er een duidelijk negatief verband tussen malocclusies en OHRQoL is, ook al is de impact die kinderen ervaren klein. Verder hebben wij gezien dat de relatie tussen malocclusies en OHRQoL varieerd afhankelijk van de leeftijd van kinderen en het land waarin de individuele studie is uitgevoerd. Echter, het bewijs voor het gevonden verband is enkel gebaseerd op cross-sectionele studies. Meer onderzoek is nodig om de overige heterogeniteit in de associatie tussen malocclusies en OHRQoL te verklaren en de causaliteit van het verband te evalueren.
Wij beschrijven ook het verband tussen subjectieve orthodontische behandelbehoefte en OHRQoL van kinderen. Subjectieve orthodontische behandelbehoefte hebben wij gemeten in vragenlijsten met volgende vraag gericht aan de moeder: "Denkt u dat uw kind een beugel nodig heeft?". Gebaseerd op de antwoorden op deze vraag zijn de kinderen verdeeld in de volgende categorieën: "geen subjectieve behandelbehoefte", "twijfelachtige subjectieve behandelbehoefte" en "definitieve subjectieve behandelbehoefte". Met behulp van lineaire regressiemodellen hebben wij geconcludeerd dat kinderen met een subjectieve behandelbehoefte lagere OHRQoL hebben dan kinderen zonder subjectieve behandelbehoefte. Bovendien hebben wij gezien dat het verband tussen een definitieve subjectieve behandelbehoefte en OHQOL bij meisjes sterker uis, terwijl het verband tussen een twijfelachtige subjectieve behandelbehoefte en OHRQoL juist bij jongens sterker is. Meer onderzoek is nodig om de rol van individuele factoren in de relatie tussen subjectieve behandelbehoefte en OHRQoL aan te tonen.
Tot slot hebben wij de relatie tussen cariës op 6-jarige leeftijd en OHRQoL op 10-jarige leeftijd onderzocht. Cariëservaring hebben wij gemeten met de "decayed, missing and filled teeth"-index (dmft). Met behulp van multinomiale logistische regressie hebben wij aangetoond dat kinderen met ernstige cariës op 6-jarige leeftijd meer kans hadden om een lagere OHRQoL op 10-jarige leeftijd te hebben dan kinderen met een gaaf gebit op 6-jarige leeftijd, ook al was het verschil in OHRQoL klein. Wij verklaarden de relatie met twee theorieën: cariës op jonge leeftijd is een sterke voorspeller van cariës op latere leeftijd of de herinnering aan pijn en de behandeling hebben mogelijk een langdurige invloed op de OHRQoL van kinderen. Meer onderzoek is nodig om de achtergrond en causaliteit van dit verband te bewijzen.
In **hoofdstuk 4** hebben wij de relatie tussen omgevingsfactoren en OHRQoL van kinderen in de Generation R Study geëvalueerd.
Hiervoor beschrijven wij sociale ongelijkheden in OHRQoL van kinderen. Omdat het moeilijk is om de sociaaleconomische status van kinderen direct te meten, hebben wij de sociaaleconomische positie (SEP) van de familie met de volgende indicatoren gemeten: de beroepssituatie van moeder en vader, het opleidingsniveau van moeder en vader, het netto huishoudinkomen, uitkeringsafhankelijkheid en éénouderschap. Met

lineaire regressieanalyse hebben wij laten zien dat alle SEP-indicatoren consistent zijn geassocieerd met OHRQoL van kinderen. Bovendien zien wij dat deze relaties onafhankelijk zijn van de objectieve mondgezondheid van de kinderen. Gezien deze verschillen moeten interventies en beleid ter bevordering van de mondgezondheid zich richten op kinderen van lagere sociaaleconomische status. Echter, er is meer onderzoek nodig om de achterliggende oorzaken van sociale ongelijkheden in OHRQoL van kinderen te begrijpen.

In **hoofdstuk 5** richten wij ons op de associaties tussen individuele kenmerken en OHRQoL van kinderen in de Generation R Study.

Wij evalueren de objectieve en subjectieve behandelbehoefte, gerelateerd aan OHRQoL bij kinderen van verschillende etnische achtergrond. Het is bekend dat etnische minderheden in het nadeel zijn met betrekking tot hun mondgezondheid. In onze studie zagen wij dat Turkse, Marokkaanse en Surinaamse kinderen een vergelijkbare objectieve behandelbehoefte hadden als Nederlandse kinderen. Echter, Turkse, Marokkaanse en Surinaamse kinderen laten een veel zwakker verband tussen objectieve en subjectieve orthodontische behandelbehoefte zien dan Nederlandse kinderen. Desondanks was OHRQoL het hoogst in Nederlandse kinderen, wat wellicht komt door de sterkere invloed van subjectieve en objectieve orthodontische behandelbehoefte op OHRQoL bij kinderen met een etnische achtergrond. Gebaseerd op deze studie moet er meer aandacht gegeven worden aan etnische verschillen in de praktijk. Echter, er is meer onderzoek nodig om de achterliggende redenen te begrijpen die voor etnische verschillen in subjectieve en objectieve mondgezondheid zorgen.

Vervolgens richten wij ons op de rol van het gevoel van eigenwaarde in de relatie tussen subjectieve orthodontische behandelbehoefte en OHRQoL. Het gevoel van eigenwaarde is een persoonlijke “resource” die het omgaan met moeilijke of pijnlijke situaties makkelijker maakt. In onze studie wordt eigenwaarde gemeten met een aangepaste versie van de “Competentiebelevingsschaal voor Kinderen”. Met behulp van lineaire regressieanalyse hebben wij laten zien, dat het gevoel van eigenwaarde de associatie tussen subjectieve orthodontische behandelbehoefte en OHRQoL noch vertroebeld (engl: confound), noch dat het verband tussen subjectieve orthodontische behandelbehoefte en OHRQoL via het gevoel van eigenwaarde wordt verklaard (engl: mediate). De relatie tussen subjectieve orthodontische behandelbehoefte en OHRQoL is echter wel verschillend tussen kinderen met een hoger gevoel van eigenwaarde en kinderen met een lager gevoel van eigenwaarde. Er moet nog veel onderzoek worden gedaan om de rol van eigenwaarde voor OHRQoL als zodanig, maar ook in relatie tot percepties van de mondgezondheid, te begrijpen en uit te leggen.

Tot slot geven wij in **hoofdstuk 6** een algemene discussie over het onderzoek gepresenteerd in dit proefschrift. Deze discussie begint met een samenvatting van de bevindingen uit de verschillende hoofdstukken gevolgd door een uiteenzetting van de sterke en zwakke punten van het onderzoek, in het bijzonder de opzet van het onderzoek (observationeel, meestal dwarsdoorsnedes), de metingen (rapportages van ouders, geen direct mondonderzoek) en de statistische analyses (bijzonderheden van OHRQoL gegevens, ontbrekende waarden). Aansluitend geven wij interpretaties van, en mogelijke mechanismen achter de gevonden resultaten. Vervolgens bespreken wij de mogelijke implicaties van onze bevindingen voor patiënten, tandartsen, onderzoekers en beleids-

makers. Tot slot geven wij mogelijke onderzoeksvragen voor toekomstig onderzoek naar subjectieve en objectieve mondgezondheid evenals OHRQoL bij kinderen. De discussie eindigt met de volgende conclusies voortkomend uit dit proefschrift: De COHIP-ortho en de beoordeling van een orthodontische behandelbehoefte op fotografische en radiografische verslagen zijn waardevolle methoden voor grootschalige onderzoek naar de mondgezondheid bij kinderen. Objectieve mondgezondheidsmaten hebben een kleine maar toch duidelijke impact op de OHRQoL van kinderen. Zelfervaren mondgezondheid heeft een soortgelijk effect op de OHRQoL van kinderen. Kinderen van een lagere sociaaleconomische positie hebben een verminderde OHRQoL. Tot slot, individuele kenmerken van kinderen zoals geslacht, leeftijd, etniciteit en het gevoel van eigenwaarde zijn determinanten van OHRQoL van kinderen, en zijn van invloed op het verband tussen subjectieve en objectieve (orthodontische) mondgezondheidsmaten.

## Zusammenfassung

In **Kapitel 1** wird eine Einleitung zu dem hiesigen Forschungsprojekt gegeben. Da subjektive und objektive (kieferorthopädische) Mundgesundheit in geringem Maße korrelieren, wird als Erklärungsversuch das Konzept der mundgesundheitsbezogenen Lebensqualität hinzugezogen (Engl.: oral health-related quality of life (OHRQoL)). OHRQoL wird definiert als „ein mehrdimensionales Konstrukt, das unter anderem das Wohlbefinden von Menschen beim Essen, Schlafen und in der sozialen Interaktion wiederspiegelt; d.h. ihre Zufriedenheit bezogen auf ihre Mundgesundheit". OHRQoL wird auch als ein Maß zur Messung von Mundgesundheit genommen. Deutlich wird jedoch, dass OHRQoL diesen Zusammenhang zwischen subjektiver und objektiver Mundgesundheit nicht hinreichend erklärt. Insbesondere bei Kindern ist es im Bereich der Kieferorthopädie relevant, die OHRQoL zu erfassen. Einerseits sind die Patienten im kieferorthopädischen Bereich mehrheitlich Kinder. Andererseits übt die Mundgesundheit bereits in jungen Jahren einen starken Einfluss auf die Mundgesundheit im Erwachsenenalter aus. Daher ist es das Ziel der vorliegenden Dissertation, den Zusammenhang zwischen objektiver und subjektiver Mundgesundheit im Bereich der Kieferorthopädie unter Einbezug der OHRQoL zu untersuchen. Diesbezüglich untersuchen wir biologische/ physiologische Faktoren, Umgebungsfaktoren und individuelle/ persönliche Faktoren von OHRQoL bei Kindern.

In **Kapitel 2** werden zwei Methoden für groß angelegte Forschungsprojekte über OHRQoL bei Kindern im Bereich der Kieferorthopädie validiert. Wir präsentieren einen verkürzten OHRQoL-Fragebogen des Child Oral Health Impact Profile (COHIP). OHRQoL-Fragebögen werden bisher nur begrenzt in Zahnarztpraxen und zahnmedizinischer Forschung eingesetzt, da sie generell sehr lang sind und dadurch eine hohe Bearbeitungszeit beanspruchen. Wir führten eine Querschnittstudie durch, um die Daten aus der ursprünglichen COHIP-Version (38 Fragen) mit den Daten aus der verkürzten COHIP-ortho-Version (11 Fragen) zu vergleichen. Das Ergebnis dieses Vergleichs zeigt, dass die Daten der beiden Fragebögen in Bezug auf die Konstruktvalidität, Kriteriumsvalidität, Punkteverteilung und internen Validität, nahezu übereinstimmen. In den folgenden Kapiteln wird daher der COHIP-ortho-Fragebogen benutzt um die OHRQoL bei Kindern zu erfassen.

Außerdem evaluieren wir den Nutzen von 2D, 3D und Röntgenfotos zur Erfassung des Index of Orthodontic Treatment Need (IOTN). Der IOTN ist ein okklusaler Index zur systematischen, akkuraten und zuverlässigen Erfassung kieferorthopädischer Daten. Derzeit sind Gipsabdrücke des Gebisses und direkte orale Untersuchungen der goldene Standard zur Bestimmung des IOTN. Diese beiden Methoden haben jedoch auch Nachteile, bspw. weil sie sehr zeitaufwendig sind und viel Platz zur Datenaufbewahrung brauchen. In der hiesigen Studie wird der IOTN zunächst auf der Grundlage von Gipsabdrücken bestimmt, dann auf der Grundlage von 2D und 3D Fotos und letztlich werden 2D, 3D und Röntgenfotos kombiniert. Beim Abgleichen der Übereinstimmungen dieser verschiedenen Methoden stellte sich heraus, dass die Kombination von 2D und 3D Fotos gut geeignet ist zur Feststellung generellen kieferorthopädischen Behandlungsbedarfs. Eine hinreichende Bestimmung des IOTN erfolgt jedoch lediglich durch die Kom-

bination von 2D, 3D und Röntgenfotos, weshalb im weiteren Verlauf der Studie ausschließlich letztere Methode zur Bestimmung des IOTN bei Kindern angewandt wird.
In **Kapitel 3** wird der Zusammenhang zwischen verschiedenen biologischen/ physiologischen Variablen, also objektiver Mundgesundheit, mit der OHRQoL bei Kindern im Rahmen der Generation R Studie untersucht. Zunächst führen wir eine systematische Literaturstudie und Metaanalyse der Literatur über den Einfluss von Okklusionsstörungen auf die OHRQoL von Kindern aus. Die gebündelten Ergebnisse zeigen einen eindeutig negativen Zusammenhang zwischen Okklusionsstörungen und OHRQoL bei Kindern, auch wenn dieser Einfluss von den Kindern selbst als eher klein wahrgenommen wird. Des Weiteren weisen die Ergebnisse darauf hin, dass Okklusionsstörungen in unterschiedlichen Ausprägungen auftreten, je nach Altersgruppe und Herkunftsland, in dem die Studie durchgeführt wurde. Diese Aussage basiert jedoch nur auf Querschnittstudien. Weiterführende Forschungsarbeiten sind dringend indiziert, um die starke Heterogenität und die Assoziation zwischen Okklusionsstörungen und OHRQoL zu erklären und einen kausalen Zusammenhang herzustellen.
Außerdem beschreiben wir den Zusammenhang zwischen subjektiver kieferorthopädischer Behandlungsbedürftigkeit und der OHRQoL bei Kindern. Das subjektive Behandlungsbedürfnis wurde gemessen indem die Eltern gefragt wurden: „Denken Sie, dass ihr Kind eine kieferorthopädische Behandlung benötigt?“. Basierend auf den Antworten zu dieser Frage wurden die Kinder in drei Kategorien eingeteilt, nämlich nicht-behandlungsbedürftig, fraglich-behandlungsbedürftig und definitiv-behandlungsbedürftig. Anhand einer linearen Regressionsanalyse wird deutlich, dass die OHRQoL bei Kindern, die sich als subjektiv kieferorthopädisch behandlungsbedürftig erleben, eingeschränkt ist. Des Weiteren wird deutlich, dass es einen stärkeren Zusammenhang zwischen definitivem Behandlungsbedürfnis und OHRQoL bei Mädchen gibt, wohingegen der Zusammenhang zwischen fraglichem Behandlungsbedürfnis und OHRQoL bei Jungen stärker ausgeprägt ist. Es ist daher dringend indiziert, dass der Zusammenhang zwischen Geschlecht und individuellen Faktoren mit subjektiver Mundgesundheit und OHRQoL tiefergehend erforscht wird.
Schließlich wird der Zusammenhang zwischen Zahnkaries bei Kindern im Alter von sechs Jahren mit der OHRQoL im Alter von zehn Jahren untersucht. Der Zustand des Zahnkaries wird anhand des Diseased-, Missing- oder Filled-Teeth Index beurteilt. Mittels einer multinomialen logistischen Regressionsanalyse zeigen wir, dass Kinder, die im Alter von sechs Jahren schweren Karies aufweisen, deutlich häufiger eine niedrigere OHRQoL im Alter von zehn Jahren haben, wobei diese absoluten Unterschiede in der OHRQoL eher gering sind zwischen Kindern ohne Karies, leichtem Karies und schwerem Karies. Diesen Zusammenhang zwischen dem Auftreten von Karies und der OHRQoL erklären wir uns dadurch, dass zum einen Karies in jungen Jahren Einfluss hat auf Karies im späteren Alter, und zum anderen dass die Erinnerung der Kinder an die damit einhergehenden Schmerzen und die unangenehme Behandlung einen nachhaltigen Einfluss auf die OHRQoL hat. Weiterführende Forschung ist notwendig, um die Hintergründe und Kausalitäten des Zusammenhangs zwischen frühem Karies und der späteren OHRQoL zu untersuchen.
In **Kapitel 4** wird der Zusammenhang zwischen Umweltmerkmalen und OHRQoL bei Kindern in der Generation R Studie beurteilt. Im Zuge dessen beschreiben wir soziale

Unterschiede von Kindern und deren OHRQoL. Da es schwierig ist, den sozial-ökonomischen Status von Kindern als solchen zu beurteilen, messen wir die sozio-ökonomische Lage (socio-economic position, SEP) der Familie anhand der folgenden Indikatoren: mütterliches und väterliches Bildungsniveau, mütterlicher und väterlicher Beziehungsstatus, Haushaltsnettoeinkommen, Abhängigkeit von Sozialhilfeleistungen und Alleinerziehung. Durch eine lineare Regressionsanalyse zeigen wir, dass alle SEP Indikatoren einheitlich mit OHRQoL verbunden sind. Des Weiteren wird deutlich, dass dieser Zusammenhang unabhängig vom tatsächlichen Zustand der Mundgesundheit zu sein scheint. In Anbetracht dieser Unterschiede sollten sich Interventionen und Maßnahmen zur Förderung einer guten Mundgesundheit und dem mundgesundheitlichen Wohlbefinden auf Kinder in einer niedrigeren sozio-ökonomischen Lage richten. Jedoch ist hier weitere Forschung indiziert, um die Hintergründe des Zusammenhangs zwischen sozialer Ungleichheit und OHRQoL bei Kindern zu verstehen.

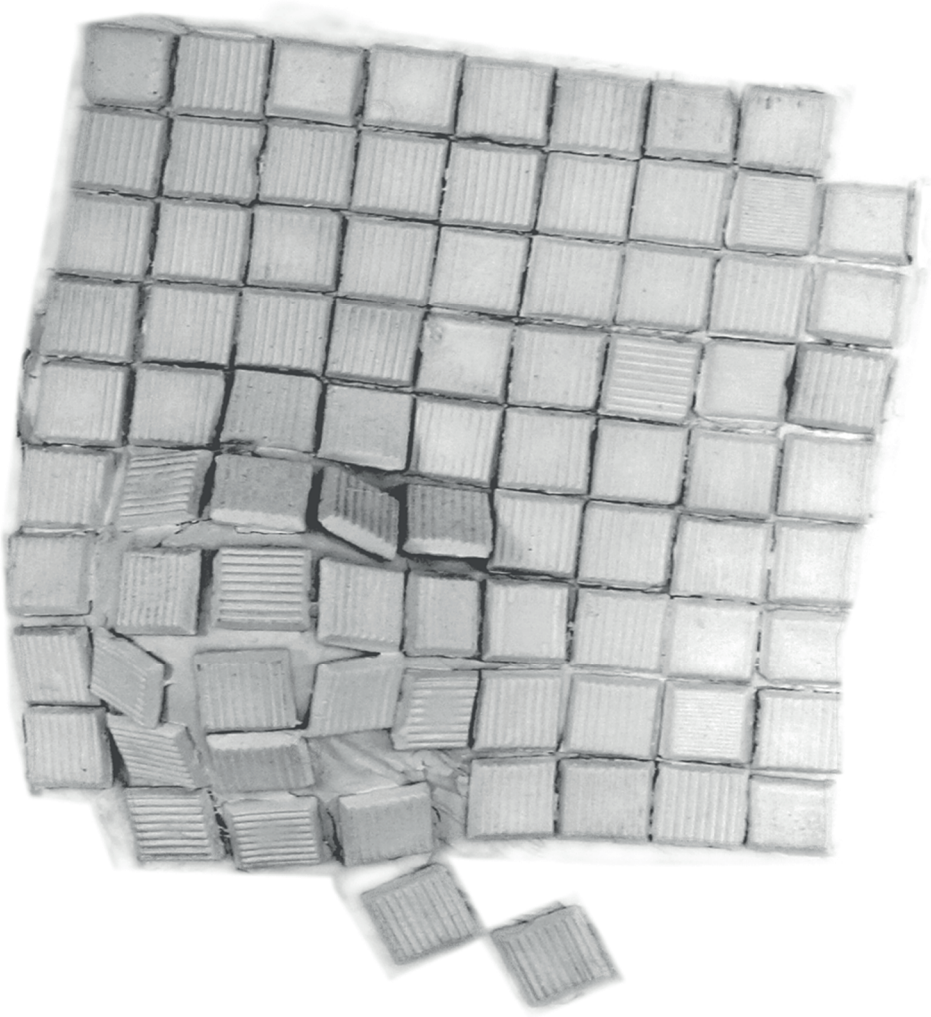
In **Kapitel 5** wird angestrebt, den Zusammenhang zwischen individuellen Merkmalen und OHRQoL von Kindern aus der Generation R Studie zu bewerten. Zunächst werden die Unterschiede zwischen Kindern verschiedener ethnischer Herkunft und deren subjektiver und objektiver kieferorthopädischer Behandlungsbedürftigkeit, sowie die OHRQoL bewertet. Es ist allgemein bekannt, dass ethnische Minderheiten im Hinblick auf die Mundgesundheit benachteiligt sind. Dennoch brauchen türkische, marokkanische und surinamische Kinder ebenso oft eine kieferorthopädische Behandlung wie niederländische Kinder. Allerdings war der Zusammenhang zwischen objektivem und subjektivem Behandlungsbedürfnis dieser Kinder deutlich geringer als bei einheimischen Kindern. OHRQOL war am höchsten bei niederländischen Kindern, was zu dem Befund passt, dass subjektives und objektives kieferorthopädisches Behandlungsbedürfnis einen größeren Einfluss auf OHRQoL hat bei Kindern mit ethnischer Herkunft. Vor dem Hintergrund dieser Ergebnisse sollte in zahnärztlichen Praxen den ethnischen Unterschieden mehr Beachtung geschenkt werden. Weitere Forschung ist notwendig, um die Hintergründe ethnischer Differenzen der subjektiven und objektiven Mundgesundheit zu verstehen.

Dann wird der Einfluss von Selbstwertgefühl auf den Zusammenhang von subjektivem kieferorthopädischem Behandlungsbedürfnis und OHRQoL bei Kindern beschrieben. Selbstwertgefühl ist eine Ressource, die es einem erleichtert, weniger günstige Situationen zu meistern. Wir messen Selbstwertgefühl mit einer modifizierten Version des Harter's Self-Perception Profiles-Fragebogen für Kinder. Anhand linearer Regressionsanalyse zeigen wir, dass Selbstwertgefühl den Zusammenhang zwischen subjektivem Behandlungsbedürfnis und OHRQoL weder trübt, noch ein Mediator dafür ist. Allerdings modifiziert Selbstwertgefühl den Zusammenhang zwischen subjektivem Behandlungsbedürfnis und OHRQoL, denn die Zusammenhänge differieren zwischen Kindern mit hohem und Kindern mit geringem Selbstwertgefühl. Weiterführende Studien sind notwendig, um die Rolle von Selbstwertgefühl auf die OHRQoL als solches, aber auch in Bezug auf die Wahrnehmung der eigenen Mundgesundheit, zu verstehen und zu erklären.

In **Kapitel 6** findet die allgemeine Diskussion der o.g. Studien statt. Es beginnt mit der Zusammenfassung der wichtigsten Ergebnisse, anschließend werden die starken und schwachen Punkte der Studien besprochen, insbesondere bezogen auf Studiendesign (observierend, meist im Querschnittsformat), Messungen (Angaben der Eltern, keine

direkten oralen Untersuchungen) und statistische Analysen (Besonderheiten der OHRQoL Daten, fehlende Daten). Im Anschluss daran präsentieren wir Interpretationen und mögliche Implikationen der Ergebnisse. So werden mögliche Auswirkungen auf Patienten, Ärzte, Forscher und politische Entscheidungsträger besprochen. Letztlich formulieren wir ungeklärte Fragen in Hinblick auf zukünftige Forschung zur subjektiven und objektiven Mundgesundheit und OHRQoL bei Kindern. Die Diskussion endet mit den folgenden Schlussfolgerungen auf dem Boden der hiesigen Dissertation: Erstens, der COHIP-ortho und die Beurteilung kieferorthopädischen Behandlungsbedürfnisses mittels 2D, 3D und Röntgenfotos bieten wertvolle Methoden mit geringem Zeitaufwand für groß aufgesetzte Forschungsprojekte über Mundgesundheit bei Kindern. Zweitens, objektive Mundgesundheitsmaße haben einen deutlichen, wenn auch geringen Einfluss auf die OHRQoL bei Kindern. Ebenso ist es bei der subjektiven Einschätzung von Mundgesundheit und den Einfluss auf OHRQoL bei Kindern. Drittens, Kinder mit einem niedrigeren sozioökonomischen Status haben eine geringere OHRQoL. Viertens, individuelle Merkmale von Kindern, wie Geschlecht, Alter, ethnische Herkunft und Selbstwertgefühl sind letztlich Bestandteile der OHRQoL bei Kindern, und beeinflussen den Zusammenhang zwischen subjektiven und objektiven (kieferorthopädischen) Mundgesundheitsmaßen.

# Chapter 8
Appendices



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## Publication List

**Kragt L**, Jaddoe VWV, Wolvius, Ongkosuwito EM. The association of subjective orthodontic treatment need with oral health-related quality of life. *Community Dent Oral Epidemiol, accepted*

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*Submitted*

**Kragt L**, Wolvius EB, Raat H, Jaddoe VWV, Ongkosuwito EM. Social inequalities in children's oral health related quality of life: The Generation R Study. *Submitted for publication*

**Kragt L**, Wolvius EB, Jaddoe VWV, Ongkosuwito EM. Associations between objective and subjective orthodontic treatment need related to children's oral health-related quality of life: Comparison between ethnic groups. *Submitted for publication*

**Kragt L**, Wolvius EB, Jaddoe VWV, Tiemeier H, Ongkosuwito EM. Influence of self-esteem on self-perceived orthodontic treatment need and oral health-related quality of life. *Submitted for publication*

**Kragt L**, Moen M, van den Hoogenband CR. Optimizing oral health among Dutch elite athletes for Rio 2016. *Submitted for publication*

Van der Tas JT, Bertens LCM, Elfrink MEC, Veerkamp JS, Jaddoe VWV, Raat H, Moll HA, Ongkosuwito EM, Wolvius EB, **Kragt L**. Social inequalities in dental caries at the age of six, Rotterdam. The Generation R Study. *Submitted for publication*

Dhamo B, Elezi B, **Kragt L**, Wolvius EB, Ongkosuwito EM. Does dental caries affect dental development in children and adolescents? *Submitted for publication*

Dahmo B, **Kragt L**, Jaddoe VWV, Wolvius EB, Ongkosuwito EM. Ethnic differences in dental development: The Generation R Study. *Submitted for publication*

Vermeij-Keers C, Rozendaal AM, Luijsterburg AJM, Latief BS, Lekkas C, **Kragt L**, Ongkosuwito EM. Classification of cleft lip and alveolus in adult unoperated patients: a new embryological approach. *Submitted for publication*

Choi TM, **Kragt L**, Goos JAC, Mathijssen IMJ, Wolvius EB, Ongkosuwito EM. Dental arch morphology and intermaxillary relationship in Saethre-Chotzen syndrome and Muenke syndrome. *Submitted for publication*

Asllanaj B, **Kragt L**, Voshol I, Koudstaal M, Kuijpers MA, Xi T , Bergé SJ, Vermeij-Keers C, Ongkosuwito EM. Dentition patterns in children with unilateral cleft lip and cleft alveolus. *Submitted for publication*

## PhD portfolio

| | |
|---|---|
| **Name PhD student:** | Lea Kragt- de Roos |
| **Erasmus MC Department:** | Oral & Maxillofacial Surgery, Special Dental Care and Orthodontics/ The Generation R Study Group |
| **Research School:** | Netherlands Institute for Health Sciences (NIHES) |
| **PhD period:** | January 2013 - December 2016 |
| **Promotors:** | Prof. Dr. Eppo B Wolvius |
| **Copromotor:** | Dr Edwin M Ongkosuwito |

| PhD training, teaching activities and other activities | Year | Workload (ECTS) |
|---|---|---|
| **1. PhD training** | | |
| ***Master degree Heath Sciences, specialization Clinical Epidemiology, NIHES, Erasmus University Rotterdam, The Netherlands*** | | |
| Principles of Research in Medicine | 2013 | 0.7 |
| Clinical Decision Analysis | 2013 | 0.7 |
| Methods of Clinical Research | 2013 | 0.7 |
| Methods of Public Health Research | 2013 | 0.7 |
| Health Economics | 2013 | 0.7 |
| Methods of Health Service Research | 2014 | 0.7 |
| Causal Inference | 2014 | 0.7 |
| Social Epidemiology | 2014 | 0.7 |
| Markers and Prognostic Research | 2014 | 0.7 |
| The Practice of Epidemiological Analysis | 2015 | 0.7 |
| Logistic regression | 2015 | 1.4 |
| Study Design | 2013 | 4.3 |
| Biostatistical Methods 1: Basic Principles | 2013 | 5.7 |
| Clinical Epidemiology | 2014 | 5.7 |
| Methodological Topics in Epidemiologic Research | 2014 | 1.4 |
| Biostatistical Methods 2: Classical Regression Models | 2015 | 4.3 |
| Diagnostic Research | 2014 | 0.9 |
| Principles of Epidemiologic Data-analysis | 2014 | 0.7 |
| Environmental Epidemiology | 2014 | 1.1 |
| Quality of Life measurement | 2013 | 0.9 |
| Health services: research and Practice | 2014 | 0.9 |
| From Problem to Solution in Public Health | 2014 | 1.1 |
| Courses for the Quantitative Researcher | 2015 | 1.4 |
| ***Extracurricular courses, Erasmus University Rotterdam/ Erasmus University Medical Center, Rotterdam, The Netherlands*** | | |
| Minicursus Methodologie van patiëntgebonden Onderzoek en Voorbereiding van Subsidievragen | 2013 | 0.6 |
| Werken met Endnote | 2013 | 0.3 |
| Basiscursus Regelgeving en Organisaties voor klinisch Onderzoek (BROK) | 2013 | 1.0 |
| Stralingshygiëne 5R | 2013 | 0.3 |
| Systematisch Literatuuronderzoek in Pubmed | 2014 | 0.2 |
| Wetenschappelijke Integriteit | 2014 | 0.3 |
| Scientific writing | 2015 | 3.0 |
| ***Extracurricular courses, abroad*** | | |
| Courses in the use of occlusal indices, Cardiff, Great Britain | 2013 | 1.0 |
| Dentistry for non-dentists, Academic Centrum voor Tandheelkunde Amsterdam (ACTA), The Netherlands | 2014 | 0.3 |
| ***Seminars and Workshops*** | | |
| Workshop occlusal Index, Rotterdam, the Netherlands. | 2014 | 0.7 |
| Generation R research meetings | 2013-16 | 1.0 |
| Seminars Department of Public Health | 2015-16 | 0.5 |
| Research meetings Department of Oral & Maxillofacial Surgery, Special Dental Care and Orthodontics | 2013-15 | 1.0 |

| | | |
|---|---|---|
| ***(Inter)national conferences and presentations*** | | |
| 90th European Congress of Orthodontic Society, Warsaw, Poland. Oral presentation | 2014 | 0.7 |
| International Association for Dental research General Session, Boston, United States. Oral presentation | 2015 | 0.7 |
| International Center for oral health inequalities research & policy Launch conference, London, Great Britain. Poster presentation | 2015 | 0.3 |
| Nederlandse Vereniging voor Orthodontisten Najaarsvergadering, Zwolle, The Netherlands. Oral presentation | 2015 | 0.5 |
| Generation R research meeting. Oral presentation | 2015 | 0.5 |
| Research meetings Department of Oral & Maxillofacial Surgery, Special Dental Care and Orthodontics. Oral presentation | 2015 | 0.5 |
| Nederlandse vereniging van specialisten in de dentomaxillaire orthopaedie Studieclub DMO, Nijmegen, The Netherlands. Oral presentation | 2016 | 0.7 |
| | | |
| **2. Teaching activities** | | |
| Supervision of master student 'Dentition patterns in children with unilateral cleft lip and cleft alveolus' | 2014-2015 | 4.0 |
| Supervision of Master student 'Ethnic disparities in dental caries among six-year-old children in the Netherlands / Social inequalities in dental caries at the age of six, Rotterdam. The Generation R Study' | 2015-2016 | 4.0 |
| Supervision of master student 'Malocclusions and OHRQoL' | 2016 | 1.0 |
| | | |
| **3. Other activities** | | |
| Peer review for Quality of Life Research, Community Dentistry and Oral Epidemiology, International Journal of Pediatric Dentistry, Transplant International | 2014-16 | 1.5 |
| Wetenschapsknooppunt Erasmus University Rotterdam, Van piep tot stok, gastles in basisschoolklassen, Rotterdam, The Netherlands | 2014 | 0.7 |
| Development of Action plan for Scientific Integrity Department of Oral and Maxillofacial Surgery, Special Dental Care, Orthodontics | 2015 | 1.0 |

## About the author

Lea Kragt was born on July 22nd 1986 in Hannover, Germany. In 2005, she completed her high school degree at the Integrierte Gesamtschule Linden, Hannover, Germany. One year later she started to study mathematics at the Technical University Berlin, Germany, but switched in 2007 to Wageningen University, the Netherlands to start her study in Nutrition and Health Sciences. In 2010 she obtained her Bachelor's degree in Nutrition and Health and started her Master's program in Nutrition and Health Sciences combined with a Minor program in Communication and Policy Making. In 2012, she obtained her Master's (including Minor) degree with the specialization in Public Health and Epidemiology. As part of her education, Lea conducted research at the Department of Human Nutrition, Wageningen University and the Louis Bolk Institute, Driebergen-Zeist, the Netherlands. In 2013, she started her PhD project at the Generation R Study Group and the Department of Oral and Maxillofacial Surgery, Special Dental Care and Orthodontics at the Erasmus University Medical Center in Rotterdam, the Netherland, the results of which are presented in this thesis. In 2015, as part of her PhD program, she obtained a second Master's degree in Clinical Epidemiology at the National Institute for Health Sciences in Rotterdam. Lea lives in Rotterdam, together with her husband Jelle de Roos and son Till.

## Words of Gratitude

*Acti labores iucundi*

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