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### Temporomandibular disorders and bruxism in children and adolescents

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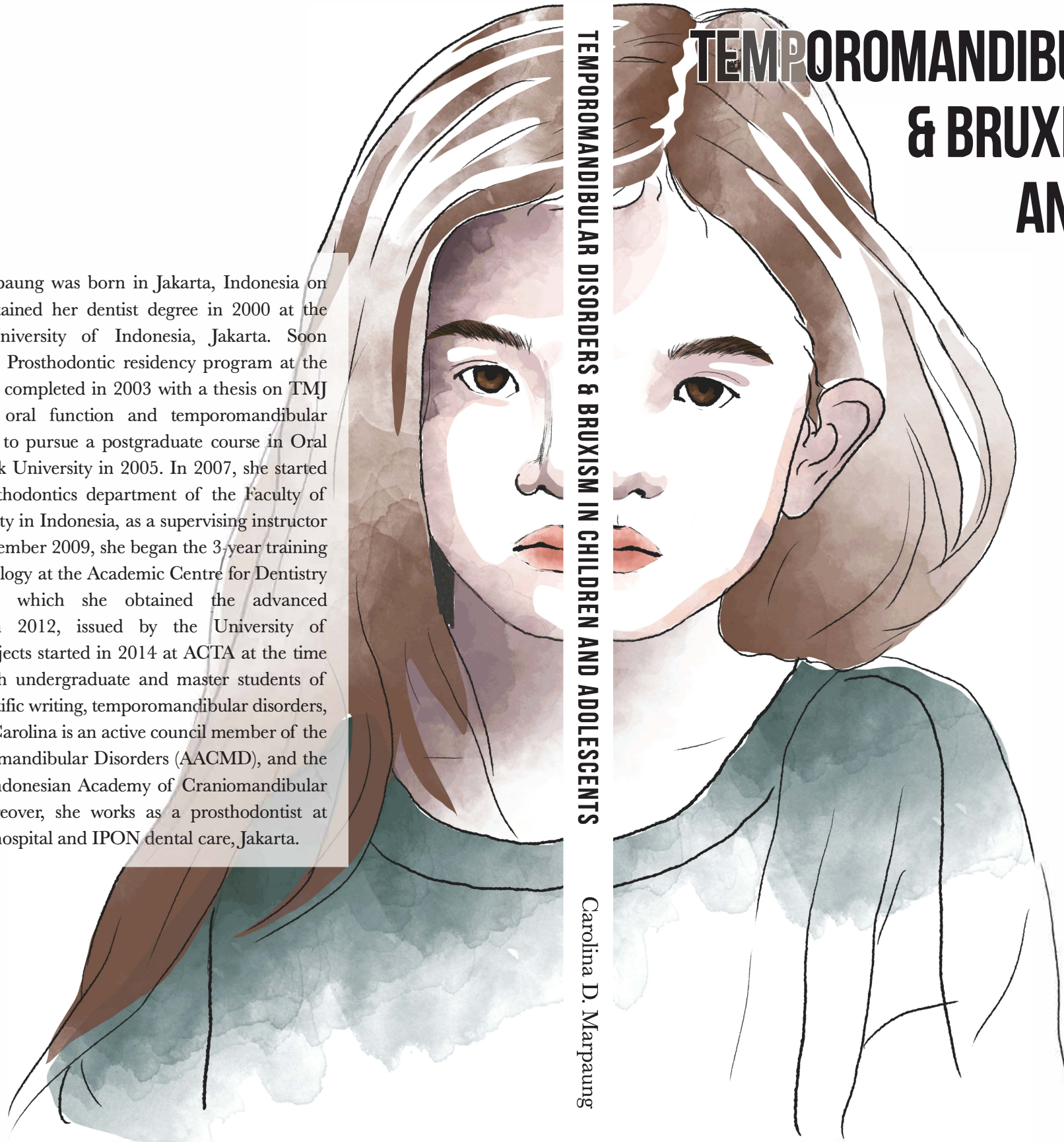
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TEMPOROMANDIBULAR DISORDERS & BRUXISM IN CHILDREN AND ADOLESCENTS

Carolina D. Marpaung

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# Temporomandibular Disorders and Bruxism in Children and Adolescents

Carolina D. Marpaung

This thesis was prepared at the Department of Oral Health Sciences,  
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# Temporomandibular Disorders and Bruxism in Children and Adolescents

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*Everything is only by His grace*

*dedicated to mama*





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# **Chapter 1**

## **General Introduction**

## **Temporomandibular disorders**

### *Background information*

Musculoskeletal disorders are the collective term for a variety of conditions that can affect the muscles, bones, and joints (Campbell 2017). In case the temporomandibular region is affected, a variety of diagnostic terms have been presented in the literature over the years, reflecting the different theories of etiology. In the early 1930s, James Costen, an otolaryngologist, published an article in which he argued that symptoms of pain and dysfunction within the region of the temporomandibular joint (TMJ) were attributed to the loss of vertical dimension of occlusion, leading to compression of the joint structures (Costen 1934). Until today, this process is still known by some as the "Costen syndrome". However, as time progressed, numerous other terms were proposed to describe, somewhat interchangeably, the various disorders in the temporomandibular region. These include, amongst others, temporomandibular joint dysfunction syndrome (Noble 1965), pain dysfunction syndrome (Marbach and Lipton 1987), and facial arthromyalgia (Madland et al. 2000). Currently, the term temporomandibular disorders (TMDs) is recommended by the American Academy of Orofacial Pain as a collective term embracing a number of clinical problems that involve the masticatory muscle, the TMJ, and the associated structures (de Leeuw and Klasser 2013).

It was not until 1992 that reference standard diagnoses for TMDs were established by consensus, known as the Research Diagnostic Criteria for TMD (RDC/TMD) (Dworkin and LeResche 1992). These criteria were intended to be the first step towards improved TMD classification, and were further developed into the Diagnostic Criteria for TMD (DC/TMD) (Schiffman et al. 2014). The current criteria classify TMDs into TMJ disorders, masticatory muscle disorders, headache attributed to TMDs, and disorders of the surrounding structures (Schiffman et al.

2014). The clinical manifestations of TMDs can be either pain, sound, movement interference, or any combination of the three (de Leeuw and Klasser 2013). Partly owing to differences in descriptive terminologies, combined with a lack of homogeneity in diagnostic criteria, prevalence rates of the various manifestations of TMDs vary widely between studies. The literature seems to be more consistent, however, regarding the occurrence of the two main types of TMDs: TMD pain and TMJ sounds (Manfredini and Guarda Nardini 2010). The prevalence of pain-related TMDs in the general population is generally assumed to be around 10% (de Leeuw and Klasser 2013; LeResche 1997; Manfredini et al. 2011), whereas this is around 25% for TMJ sounds (da Silva et al. 2016; Naeije et al. 2013).

### *Consequences and impact*

Pain-related TMDs have shown to be the second most common musculoskeletal pain condition (after chronic low back pain) (National Institute of Dental and Craniofacial Research 2013). TMD pain can originate from the TMJs, but more frequently the masticatory muscles are involved (Okeson and de Leeuw 2011; Zakrzewska 2013). Besides functional limitations, TMD pain can cause physical and psychological distress in adults (Barros Vde et al. 2009; Rodrigues et al. 2015), due to which patients take time off from work, or are unable to carry out their normal activities (Macfarlane et al. 2002).

TMJ sounds occur at some point of the mouth opening and/or closing movement, and are usually described as clicking, popping, grating, or crepitus (McNeill 1993). One of the most common sounds is due to a disc that is displaced to the anterior part of the condyle in closed mouth position. In most cases, the displaced disc reduces (restores) its normal, physiological relationship with the condyle on mouth opening, which results in a clicking sound. During closing, the disc slips off

the condyle, giving rise to a softer clicking sound (Naeije et al. 2013). This condition is known as anterior disc displacement with reduction (ADDR). ADDR is considered a stable and painless condition that usually causes little or no discomfort to the patient (Kimos et al. 2009; Lundh et al. 1987; Sato et al. 2003). However, there are reports that in some patients, an ADDR may cause a more serious complaint in case the disc reduction gets hampered, known as anterior disc displacement without reduction. The latter can be accompanied by limited mouth opening and joint pain (Kalaykova et al. 2011a; Lundh et al. 1987; Westesson and Lundh 1989).

## **Bruxism**

Bruxism has been suggested as one of the causative factors of TMDs, even though the final proof for such causality is lacking (Lobbezoo and Lavigne 1997; Manfredini and Lobbezoo 2010). Bruxism is defined as a repetitive jaw muscle activity characterized by clenching or grinding of the teeth and/or bracing or thrusting of the mandible (Lobbezoo et al. 2013). Despite being often grouped together and generically referred to as 'bruxism', it has two circadian manifestations. Depending on the time of the day on which bruxism activities occur, the activity is either defined as awake bruxism or as sleep bruxism (Klasser et al. 2015; Paesani 2010). Although the effects of both conditions on the masticatory structures may be the same, both forms of bruxism are suspected to have different etiopathogeneses (Manfredini and Lobbezoo 2010; Paesani 2010). Sleep bruxism is now considered to be primarily a sleep-related movement disorder with a yet to be determined multifactorial aetiology involving complex multisystem physiological processes (Klasser et al. 2015; Paesani 2010). Of these processes, several studies have indicated that sleep bruxism is part of an arousal response, and is related to disturbances in the central dopaminergic system (Lobbezoo and Naeije 2001; Shetty et al. 2010). It mainly involves grinding and clenching activities (De Laat and Macaluso 2002). On the other hand, it is

assumed that awake bruxism is a semi voluntary ‘clenching’ activity, and is thought to be related with psychological factors such as life stress and anxiety (Manfredini and Lobbezoo 2009; Shetty et al. 2010).

Besides TMDs, both awake bruxism and sleep bruxism have been associated with several clinical problems, such as tooth wear and failing dental restorative treatments (Lobbezoo et al. 2006; Lavigne et al. 2007; Paesani et al. 2013). On the other hand, it has been suggested that sleep bruxism can have beneficial effects as well, like preserving upper airway patency during sleep, and maintaining salivary lubrication of the alimentary tract during sleep (Murray et al. 1998; Lavigne 2003; Bracha et al. 2005). Bruxism could thus be considered a behavior that is important for human survival over the course of evolution (Raphael et al. 2016).

Partly because of the use of different definitions, study methods, criteria and population samples, the prevalence figures of bruxism vary between studies. Regardless of the type of bruxism activity, it is estimated that bruxism occurrence varies from about 8% to 30%, and that the prevalence declines with increasing age (Manfredini et al. 2013a; Manfredini et al. 2013b). Recently, a grading system for the diagnosis of awake bruxism and sleep bruxism was developed by consensus among an international group of experts (Lobbezoo et al. 2013). This system provides a tool for clinical and research purposes, making use of the terms ‘possible’, ‘probable’, and ‘definite’ awake bruxism or sleep bruxism (**Table 1**).

Table 1. Diagnostic grading system for awake bruxism and sleep bruxism developed by consensus among an international group of experts (Lobbezoo et al. 2013).

Possible anamnestic	Based on self-report using a questionnaire and/or the part of the clinical examination.
Probable	Based on self-report plus the inspection part of the clinical examination.
Definite	Based on self-report plus the inspection part of the clinical examination, and on a polysomnographic recording, preferably containing audio/visual recordings.

### **TMDs and bruxism in the young population**

Health studies on children are important, because in many cases a health condition that occurs during childhood sets the stage for adult health (National Academy of Sciences 2004). For instance, children who experience pain in early life often show long-term changes in terms of pain perception and related behaviors (McGrath and Frager 1996). Studies on pain and related disorders conducted in the young population are thus deemed necessary to learn more about the course of such disorders. In addition, an early detection and possible intervention might reduce future problems (da Silva et al. 2017).

Epidemiologic studies among children and adolescents suggest that TMD pain has a prevalence of 16-68%, whereas the prevalence of TMJ sounds varies between 4% and 40% (Fernandes De Sena et al. 2013; Da Silva et al. 2016). Studies on children suggest that TMD pain reports increase with age, and that they are



related to bruxism, oral habits, bodily pain, and psychological factors (LeResche et al. 2005; Casanova-Rosado et al. 2006; Fernandes et al. 2014). As also found in studies on adults, the occurrence and intensity of TMD pain are assumed to be higher among the female population (Liljeström et al. 2008; Nilsson et al. 2009). TMJ sounds, on the other hand, are supposed to be more related to anatomical factors (Isberg et al. 1998; Huddleston Slater et al. 2007). Even though it is assumed that TMJ sounds are almost equally distributed among the sexes and ages (Manfredini and Guarda Nardini 2010), studies on children and adolescents indicated that increasing age is closely related to the occurrence of joint sounds (Keeling et al. 1994; Huddleston Slater et al. 2007).

Especially in young children, the identification of possible bruxism is often based on parental report (Cheifetz et al. 2005; Clementino et al. 2017). As the teeth grinding activity creates noise, it is believed that the parents have no difficulties in observing sleep bruxism in their children. This is, however, not the case for awake bruxism, which mainly consist of ‘soundless’ clenching activities. Epidemiologic studies have shown that bruxism activity is a common condition among children and adolescents, with a prevalence of 5% - 41% for sleep bruxism, and of 9% - 19% for awake bruxism (Carra et al. 2011; Manfredini et al. 2013a). Many factors may be involved in bruxism presence in the young population, and it is generally acknowledged that pathophysiological and psychological factors play more dominant roles than morphological factors (Carra et al. 2011; Emodi-Perlman et al. 2016; van Selms et al. 2013).

## **Thesis research questions and aims**

Though much needed, health studies performed on children are still facing several issues that will be discussed in this thesis. For example, little is known about potential

geographic differences in TMDs and bruxism prevalence rates among the young population, as they have been established in only a few populations other than those of Western nations. In addition, differences in definition, diagnostic method, and age distribution prevent drawing conclusions about their prevalence or associated factors. The general aim of the present thesis therefore was to obtain a deeper insight into the prevalence and risk indicators of TMDs and bruxism in children and adolescents in multiple countries. Specific research questions and aims will be formulated for each chapter of this thesis.

For the evaluation of disorders within the TMJ, such as ADDR, magnetic resonance imaging (MRI) is considered the gold standard. However, due to its low availability and high costs, MRI is not feasible to be used in large-scale studies. Functional diagnostic methods, such as clinical examination of joint sounds, is easier to be used in such studies. However, there are indications that there is a poor agreement between diagnoses made with MRI and those made using functional diagnostic methods (Barclay et al. 1999; Emshoff and Rudisch 2001). **Chapter 2** of this thesis will therefore investigate the validity of two functional diagnostic methods of ADDR assessment, namely clinical examination and mandibular movement recording, with MRI as the gold standard.

Several epidemiological studies have indicated that the occurrence of ADDR among the young population can be high. Unfortunately, most of the available information was derived from studies that were conducted on Caucasians (Egermark et al. 2001; Huddleston Slater et al. 2007; Kalaykova et al. 2011b). As ethnicity may have an influence on the form and shape of the TMJ complex (Fletcher 1985; Jasinevicius et al. 2005), data from other ethnic groups are highly needed. **Chapter 3** of this thesis describes a study on ADDR prevalence and risk indicators that has been conducted on the Asian population.

It is well known that TMDs prevalence rates vary widely between studies. Besides differences in methodology and sample characteristics, there are indications that socio-economic status and living area (rural versus urban) play a role in the prevalence of self-reported TMD pain in the young population (Fernandes et al. 2015; Hongxing et al. 2016). The characteristics of Indonesia as a country, with marked socioeconomic disparities and a large proportion of young population, makes this a suitable country to learn more about the nature of pain-related TMDs in children and adolescents. **Chapter 4** describes the results of a questionnaire study designed to assess the prevalence and risk indicators of pain-related TMDs in Indonesia.

It is generally believed that a variety of biological, psychological, and social factors may reduce the adaptive capacity of the masticatory system, thus resulting in TMDs (de Leeuw and Klasser 2013; Suvinen et al. 2005). However, many previous studies either focused only on one category of TMDs, namely pain-related TMDs or TMJ sounds (e.g., Fernandes et al. 2015; Kononen et al. 1996; Nilsson 2007), or merged the signs and symptoms into one overall TMD diagnosis (e.g., Karibe et al. 2015; Pereira et al. 2009; Sermet Elbay et al. 2017). As it is generally agreed that TMDs embody a nonspecific umbrella term, it is essential to differentiate pain-related TMDs from TMJ sounds. By differentiating between the two, one can also learn if there are any common risk indicators for both types of TMDs. **Chapter 5** therefore focuses on the prevalence rates of both categories of TMDs in a large group of 12-18 year-old Dutch adolescents, and explores if there are similarities in risk indicators of these two categories.

As discussed earlier, prevalence rates of bruxism vary widely between studies. There are even indications that differences in the geographic, cultural, and socioeconomic background are responsible for this variation (Manfredini et al. 2017; Manfredini et al. 2013a). For instance, several self-reported bruxism studies showed

a relation between ethnicity and bruxism prevalence (Hicks et al. 1999; van Selms et al. 2013). So far, there are no studies that directly compared the prevalence of sleep bruxism between different countries by using the same methodology. **Chapter 6** focuses on potential geographic variation in parental-reported sleep bruxism prevalence rates among children aged 7-12 years old. The study was conducted in three culturally different countries, namely The Netherlands, Indonesia, and Armenia.

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## **Chapter 2**

### **Validity of functional diagnostic examination for temporomandibular joint disc displacement with reduction**

Marpaung C, Kalaykova SI, Lobbezoo F, Naeije M

## Authors' contributions

	Study conception	Study design	Data acquisition	Data analysis	Interpretation of data	Drafting the manuscript	Critically revising the manuscript
Marpaung C	√	√	√	√	√	√	√
Kalaykova SI	√	√	√	√	√	√	√
Lobbezoo F	√	√	√		√		√
Naeije M	√	√	√	√	√		√

## **Abstract**

**Objective:** The choice of approach for diagnosing temporomandibular joint (TMJ) anterior disc displacement with reduction (ADDR), viz. functional examination or TMJ imaging, is debatable and complicated by findings of low agreement between these approaches. Our aim was to investigate the validity of functional ADDR diagnostics using clinical examination and opto-electronic mandibular movement recordings versus magnetic resonance imaging (MRI).

**Materials and methods:** 53 participants (32 women and 21 men, mean age  $\pm$  s.d. of  $28.7 \pm 10.1$  years) underwent a clinical examination, mandibular movement recording and MRI of their TMJs within 1 month. All were performed and analysed in a single-blind design by different experienced examiners for each technique. The sensitivity and specificity of each functional diagnostic method was calculated, with MRI as the gold standard.

**Results:** Anterior disc displacement with reduction was diagnosed in 27.6% of the TMJs clinically, in 15.2% using the movement recordings and in 44.8% on MRI. The specificity of the clinical examination for diagnosing ADDR was 81.0%, and of the movement recordings, 96.6%. The sensitivity was 38.3% and 29.8%, respectively.

**Conclusions:** The chance of having a false-positive functional diagnosis of ADDR compared with MRI is low, and disagreement between the functional methods and MRI is mainly due to the high number of MRI diagnoses in asymptomatic subjects. In view of the fact that ADDR becomes clinically relevant only when it interferes with TMJ function, the functional diagnostic approach can be considered benchmark in ADDR recognition.

## **Introduction**

Internal derangements (IDs) of the temporomandibular joint (TMJ) are described as deviations in the anatomical position or form of the tissues within the capsule of the joint (The glossary of prosthodontic terms 2005). The clinical manifestations of IDs are interferences with smooth TMJ movements and TMJ sounds, with or without the presence of pain (McNeill 1993).

The most common type of ID among adults, as well as among children, is anterior disc displacement with reduction (ADDR) (Huddleston Slater et al. 2007). This type of ID is manifested by clicking joint sounds at the moments of disc reduction and dislocation (Farrar and MacCarty 1982) and is considered to cause only little discomfort (Kalaykova et al. 2010). At times, however, ADDR can develop into a more serious condition, as disc reduction may become hampered (i.e. anterior disc displacement without reduction, ADDwoR), which could lead to a limited mouth opening and pain (so called closed lock) (Farrar and MacCarty 1982). Anterior disc displacement with reduction diagnostics is important for clinical care and for research related to this type of derangement, among others, because its recognition contributes to gaining further insight into its pathophysiology and prognosis.

There are two approaches in diagnosing TMJ ADDR: anatomical and functional. The first approach aims at joint visualisation, for example using magnetic resonance imaging (MRI) (Larheim 2005). The second, functional approach utilises methods to observe functional interferences during mandibular movement and includes methods such as clinical examination and mandibular movement recordings (Dworkin and LeResche 1992; Huddleston Slater et al. 2004). The choice of diagnostic approach to recognise disc displacement is still a debatable matter (Limchaichana et al. 2006; Truelove et al. 2010). Although not undisputed (Palla 2009), MRI is currently considered as the gold diagnostic standard (Larheim 2005). However, MRI use is limited because of its low availability and high costs. The

choice of diagnostic approach for TMJ ADDR is further complicated by findings of a generally poor agreement between MRI diagnoses and those made using functional diagnostic methods (Barclay et al. 1999; Emshoff et al. 2002a; Emshoff et al. 2002b; Huddleston Slater et al. 2004). Insight into the reason for disagreement between the diagnostic methods (e.g. chance of false-positive or false-negative outcomes using a certain method) would assist decision-making regarding the use of the diagnostic approaches.

The aim of this study was to investigate the validity (i.e. specificity and sensitivity) of clinical examination and opto-electronic movement recordings versus MRI in the diagnostics of ADDR of the TMJ.

## **Materials and methods**

### *Participants*

In this study, 53 participants (32 women and 21 men; mean age  $\pm$  s.d. = 28.7 years  $\pm$  10.1 years) were recruited from the patients of the Clinic for Oral Kinesiology (among others, specialised in temporomandibular disorders, TMDs) and from dental students, all within the Academic Centre for Dentistry Amsterdam (ACTA). The subjects were screened clinically prior to participation. Subjects with ADDwoR, accompanied by signs and symptoms of a closed lock, as well as with other painful temporomandibular disorder conditions were excluded during the screening, as the presence of these conditions could influence mandibular function and joint movement, and bias ADDR diagnostics. All subjects gave informed consent to participate in the study. The study was approved by the institutional review board ACTA & Vrije Universiteit Medisch Centrum (VUMC) and conducted in accordance with the ethical principles of the World Medical Association Declaration of Helsinki.

### *Protocol*

Each participant underwent three examinations within a period of 1 month: clinical examination, opto-electronic mandibular movement recording and MRI of the temporomandibular joints. All were performed in a single-blind design by a different experienced examiner for each technique. The presence of ADDR was distinguished using criteria for each examination technique, which are described below. Internal derangements that did not fulfil ADDR criteria were categorised as ‘other IDs’.

### *Clinical examination*

One calibrated examiner (FL) examined the presence of internal derangements using palpation and auscultation with a stethoscope. Palpation was performed with the index and middle fingers placed over the participant’s lateral poles of the TMJ, around 1.5 cm anterior to the tragus. The finger pressure on the skin was about 5 N and was applied simultaneously to both joints while the participant was performing jaw movements. For the auscultation technique, the bell of an infant stethoscope (3M Littmann, St. Paul, MN, USA) was used on the same place as where the examiner did the palpation technique. During auscultation, the contralateral side was manually palpated. The participants performed a minimum of 3 maximal opening and closing movements and three protrusive opening movements that started from and ended in an interincisal end-to-end position (elimination test). Also three closing movements with 30 N of counter pressure were performed; the load of 30 N was manually applied to the chin to amplify possible closing clicks (Huddleston Slater et al. 2004).

The clinical criteria used to distinguish ADDR were as follows (Huddleston Slater et al. 2004; Naeije et al. 2009):

1. Reciprocal TMJ clicking, reproducible on at least two of three opening and (loaded) closing movements and
2. Elimination of the TMJ clicking on protrusive opening and closing movements.

### *Opto-electronic movement recording*

Functional signs of internal derangement were recorded by means of the Oral Kinesiology Analysis System (OKAS-3D) (Naeije et al. 1995). It is an opto-electronic device that is capable of accurately recording mandibular motion with 6 degrees of freedom at a sampling frequency of 300 Hz per coordinate. Small condenser microphones were placed over the lateral poles of both TMJs to simultaneously record joint sounds. All recordings were later interpreted offline in a single-blind manner by a single investigator (MN), using specialised software which can graphically capture the movement traces of the incisal point and of the kinematic centers of both condyles in a sagittal, horizontal and frontal plane. The occurrences of joint sound were also depicted in the movement traces. The participants performed the same types of movements as during the clinical examination; they performed each type of movement on average 6-9 times during recordings of 20 s each.

The presence of an ADDR was noted based on the following criteria (Figure 1) (Huddleston Slater et al. 2004):

1. Reproducible deflections in the sagittal kinematic condylar movement traces during opening and (loaded) closing, which coincided with clicking joint sounds; and
2. Elimination of the deflections and clicks during protrusive opening and closing movements.

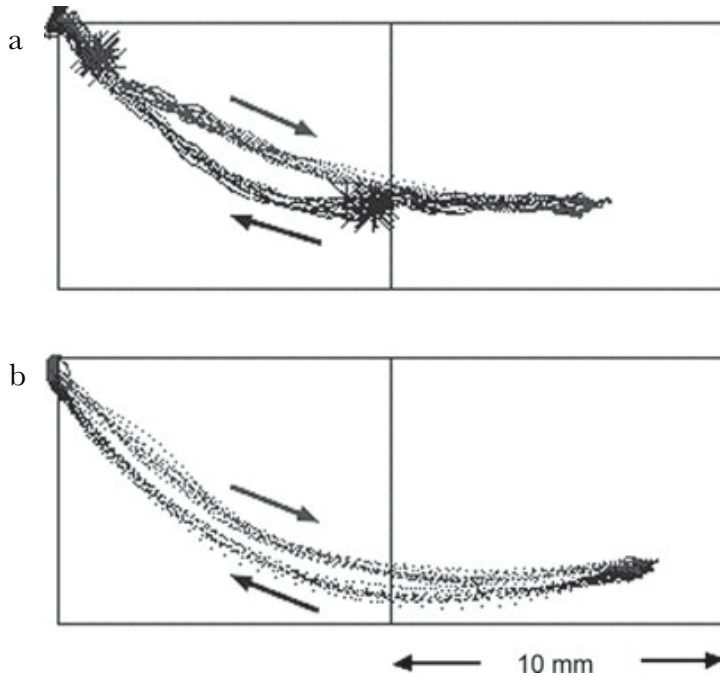


Figure 1. Example of the superimposed multiple sagittal condylar movement traces of the kinematic centre of a temporomandibular joint (TMJ) with anterior disc displacement with reduction (ADDR) (a) or without an internal derangement (b). On average 6 condylar movements are performed within a 20-s period of repeated maximal open–close jaw movements, starting from the intercuspal position. The movements of the condyle in the TMJ with ADDR (a) showed interferences coinciding with the opening clicks (i.e. disc reduction) and closing clicks (i.e. disc dislocation). The condyle in (b) performed smooth movements without interferences and without clicking sounds. The superimposed movement traces also illustrate the reproducibility of the movement recordings. Arrows denote the direction of condylar movement during mandibular opening ( $\rightarrow$ ) and closing ( $\leftarrow$ ). Opening and closing clicking sounds are indicated with asterisks (\*).



*Magnetic resonance imaging.*

For MR imaging of the TMJ, T1-weighted images were made at the Faculty of Medicine, Utrecht University, The Netherlands, using 1.5 T MRI system (Gyroscan NT Intera, Philips Medical Systems, Eindhoven, The Netherlands). A surface coil was used as receiver. The repetition time was 530 ms; the echo time was 18 ms. Imaging was performed with the patient's head placed in a headrest in the MR imager, in a closed-mouth position with nine interleaved 3-mm sagittal planes (perpendicular to the mediolateral axis of the condyle) obtained from lateral to medial, followed by nine interleaved 3-mm coronal planes. Imaging was also performed in the maximally opened-mouth position, controlled with a resin bite block, with nine interleaved 3-mm sagittal planes obtained lateral to medial. For all images made, the data matrix was 205 9 256 pixels, and the imaging time 4 min and 21 s.

All MRI's were interpreted blindly by the same examiner (Dr. Y.J. Chen, expert in TMJ imaging). The following criteria, modified from Katzberg and Westesson (1993) (Katzberg and Westesson 1993), were used to diagnose the ADDRs (see also Figure 2):

1. The inferior surface of the intermediate zone was anterior to the anterior prominence of the condyle, and not in contact with the condyle when the mouth was closed and
2. The condyle was underneath the intermediate zone of the disc when the mouth was fully open.

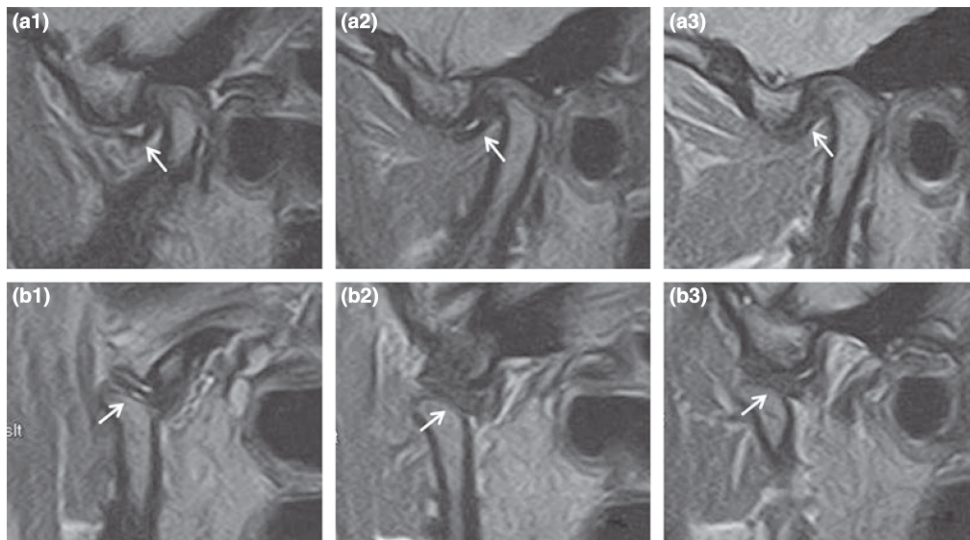


Figure 2. Example of magnetic resonance images of a temporomandibular joint (TMJ) with anterior disc displacement with reduction (ADDR). Multiple consequent oblique sagittal images of the TMJ are shown with the mouth closed (a-1 to 3) and fully opened (b-1 to 3). On all images with the mouth closed, the inferior surface of the intermediate zone is anterior to the anterior prominence of the condyle, and not in contact with the condyle. On the images with the mouth opened, the condyle is underneath the intermediate zone of the disc. Arrows ( $\rightarrow$ ) denote the intermediate zone of the articular disc.

### *Statistical analysis*

The prevalence of ADDR and of other TMJ internal derangements was calculated for the study sample. Percentage agreement and Cohen's kappa ( $\kappa$ ) between the three diagnostic methods (i.e. MRI, OKAS-3D and clinical examination) regarding the presence or absence of ADDR were calculated. Kappa values were interpreted according to Landis and Koch (1977): 0–0.20 as slight agreement, 0.21–0.40 as fair agreement, 0.41–0.60 as moderate agreement, 0.61–0.80 as substantial agreement and 0.81–1 as nearly perfect agreement. Furthermore, data were arranged in cross-tables, with the MRI diagnoses as the gold

standard and each of the functional diagnoses (i.e. made by clinical examination or by OKAS-3D) as the predictor variables. The sensitivity and specificity of each method for predicting ADDR diagnoses were then calculated.

## Results

One hundred and five TMJs from the 53 participants were analysed. One joint was excluded from analysis as its MR images were not interpretable. Clinical examination diagnosed 29 joints as having ADDR (27.6%), and 76 joints as having no ADDR (72.4%) (Table 1). Functional examination using OKAS-3D diagnosed 16 joints with ADDR (15.2%), and 89 joints as having no ADDR (84.8%) (Table 2). Magnetic resonance imaging diagnosed 47 joints with ADDR (44.8%), and 58 joints as having no ADDR (55.2%) (Tables 1 and 2).

Table 1. Number of temporomandibular joints (TMJs) with and without anterior disc displacement with reduction (ADDR) recognized by the diagnostic methods: clinical examination and magnetic resonance imaging (MRI)

	<b>MRI</b>		
	No ADDR	ADDR	Total
<b>CLINICAL EXAMINATION</b>			
No ADDR	47	29	76
ADDR	11	18	29
Total	58	47	105

Table 2. Number of temporomandibular joints (TMJs) with and without anterior disc displacement with reduction (ADDR) recognized by the diagnostic methods: condylar movement recordings with Oral Kinesiology Analysis System 3-D (OKAS 3D) and magnetic resonance imaging (MRI)

	<b>MRI</b>		
	No ADDR	ADDR	Total
<b>OKAS 3-D</b>			
No ADDR	56	33	89
ADDR	2	14	16
Total	58	47	105

The percentage agreement between the two functional diagnostic methods (i.e. clinical examination and OKAS-3D) regarding the presence or absence of ADDR was 87.71%, and the Cohen’s  $\kappa$  indicated a substantial between-method agreement (0.64). The percentage agreement between the clinical examination, or OKAS-3D, and MRI was 61.91% and 66.67%, respectively, and the Cohen’s  $\kappa$  indicated slight to fair agreement (0.2 and 0.28, respectively) between the functional diagnostic methods and MRI.

With MRI as the gold standard, the clinical examination and OKAS-3D showed specificities for diagnosing ADDR of 81.0% and 96.6%, respectively. The sensitivity was 38.3% and 29.8%, respectively.

## **Discussion**

In the present study, the validity of functional diagnostics (viz. clinical examination and opto-electronic movement recordings) versus imaging diagnostics (viz. MRI) of TMJ ADDR was investigated. For both functional approaches, a high specificity and

low sensitivity were found when MRI was used as the gold standard. This indicates that the chance of having a false-positive functional ADDR diagnosis is low and that disagreements between the functional and imaging diagnostic approaches are mainly due to the high number of MRI diagnoses in asymptomatic subjects.

To our knowledge, so far, two previous studies have reported the sensitivity and specificity of clinical examination compared with MRI regarding ADDR. Truelove et al. (Truelove et al. 2010) found, similarly to our results, a low sensitivity (38%) and high specificity (88%) for clinical ADDR diagnostics. Emshoff et al. (Emshoff et al. 2002a) found just the opposite results, namely a high sensitivity (85%) and low specificity (25%). These different results are probably explained by differences in clinical tests and in diagnostic criteria used. Emshoff et al. (Emshoff et al. 2002a) used the Clinical Diagnostic Criteria for TMD (CDC/TMD) (Truelove et al. 1992); Truelove et al. (Truelove et al. 2010) used the Research Diagnostic Criteria for TMD (RDC/TMD) (Dworkin and LeResche 1992), and we used the criteria suggested by Huddleston Slater et al. (Huddleston Slater et al. 2004) and Naeije et al. (Naeije et al. 2009). In contrast to the RDC/TMD and the criteria used in our study, ADDR diagnosis by CDC/TMD is based solely upon the presence of reproducible TMJ clicking during mandibular movements. Reciprocity and elimination of TMJ clicking on open–close movements from protruded jaw position (i.e. positive elimination test) are not required by the CDC/TMD. As TMJ clicking is a symptom of several types of ID, not only of ADDR (Huddleston Slater et al. 2004; Huddleston Slater et al. 2007), it is likely that in the study by Emshoff et al. (Emshoff et al. 2002a), the clinical ADDR diagnoses were overrepresented and this could partly explain the high sensitivity and low specificity found in that study.

Comparisons between clinical and MRI diagnostics for ADDR, however, without reporting sensitivity and specificity, have been made in several other studies as well. Barclay et al. (Barclay et al. 1999), Emshoff et al. (Emshoff et al. 2002b) and Limchaichana et al. (Limchaichana et al. 2007) reported low agreement between

MRI and clinical examination using the RDC/TMD or CDC/TMD, which was primarily explained by MRI findings of a disc displacement in clinically asymptomatic subjects. Manfredini and Guarda-Nardini (2008) (Manfredini and Guarda-Nardini 2008) did find that cases of ADDR predicted by RDC/ TMD showed good agreement with MRI. From their presented data, it was visible that cases of disagreement were mainly due to findings of a TMJ disc displacement (with or without reduction) in clinically asymptomatic subjects, which is in line with our findings.

Previous studies on the sensitivity and specificity of condylar movement recordings regarding the diagnostics of ADDR have resulted in findings that are similar to ours, namely low sensitivities and high specificities (Ozawa and Tanne 1997; Parlett et al. 1993). In these studies, ADDR was diagnosed using axiography, without TMJ sound recordings, but with an analysis of specific movement deflections in the condylar traces. These deflections are caused by reductions and dislocations of the disc (Isberg-Holm and Westesson 1982) and are similar to those recorded by OKAS-3D [Figure 1; see also Huddleston Slater et al. (Huddleston Slater et al. 2004)]. The simultaneous registration of TMJ sounds and condylar movement interferences, however, remains important to eliminate possible false-positive movement traces, for example due to interference from cross-talking from the contralateral TMJ. Moreover, in our study, not only the presence of condylar movement interferences coinciding with joint clicking was necessary to diagnose ADDR, but additionally a positive elimination test was required. This requirement helped to further decrease the chance of ADDR false positivity.

From the above, it can be gathered that the sensitivity of both functional methods for diagnosing ADDR, when compared to MRI, is low and lower than the minimal value of 70% recommended by others (Dworkin and LeResche 1992; Truelove et al. 2010). However, previously it has been shown that ADDR is in most cases a stable, benign condition that causes no, or only minor discomfort to the

patients (Kalaykova et al. 2010). For low morbid conditions like ADDRs, specificity is more important than sensitivity to prevent false positives and overtreatment.

The techniques of mandibular movement recordings and clinical examination showed substantial agreement ( $\kappa = 0.64$ ) and resulted in similar specificities (i.e. 96.6% and 81.0%) and sensitivities (29.8% and 38.3%) in recognising ADDR compared with MRI. This indicates that these methods can interchangeably be used in ADDR diagnostics. Mandibular movement recording devices are not largely available, which makes clinical diagnostics the method of choice in daily practice. The movement recordings, however, have the advantage of registering in detail the condylar movements, the joint sounds, and the moments of disc reduction and dislocation, all of which can be analysed multiple times later on offline (Huddleston Slater et al. 2004; Kalaykova et al. 2010; Naeije et al. 1995). Thus, movement recordings are especially valuable for research purposes and have been used in studies to the long-term ADDR time course (Kalaykova et al. 2010) and in simulation studies of TMJ condylar and disc movements and TMJ loading (Palla et al. 2003).

## **Conclusions**

The chance of having a false-positive functional ADDR diagnosis compared with MRI is low, and disagreement between the functional diagnostic and imaging methods is mainly due to MRI findings in asymptomatic subjects. In view of the fact that ADDR becomes clinically relevant only when it interferes with TMJ function, we recommend that ADDR recognition relies primarily on the functional diagnostic approach.

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## **Chapter 3**

# **Temporomandibular Joint Anterior Disc Displacement with Reduction in a Young Population: Prevalence and Risk Indicators**

Marpaung C, van Selms MKA, Lobbezoo F

## Authors' contributions

	Study conception	Study design	Data acquisition	Data analysis	Interpretation of data	Drafting the manuscript	Critically revising the manuscript
Marpaung C	√	√	√	√	√	√	√
van Selms MKA	√	√		√	√	√	√
Lobbezoo F	√	√		√	√		√

## **Abstract**

**Objective:** Temporomandibular joint (TMJ) anterior disc displacements with reduction (ADDR) are commonly found in the young population, and often found to be associated with biomechanical and anatomical factors. Until now, most knowledge on ADDR among children and adolescents comes from studies performed on Caucasian subjects. The aim of this study was to assess the clinically determined prevalence rates of ADDR among the young Indonesian population, and to evaluate its risk indicators.

**Materials and methods:** In this cross-sectional study, 1,562 pupils and students of 7-21 years old completed a questionnaire and underwent a clinical examination.

**Results:** The prevalence rates of ADDR were 7.0% among children (7-12 years), 14.4% among adolescents (13-18 years), and 12.3% among young adults (19-21 years). Logistic regression analyses revealed that increasing age and lip biting were associated with ADDR in children, while pen biting was associated with ADDR in the adolescent population. None of the included factors were found to be associated with ADDR in the young adult population.

**Conclusions:** The present findings indicate that prevalence of ADDR increases with age, with a peak during the years of adolescence. Biomechanical factors seem to play a significant role in ADDR development.

## **Introduction**

Internal derangements (IDs) of the temporomandibular joint (TMJ) are defined as deviations in the position or form of the tissues within TMJ capsule (Prosthodontics 2017). Among the several IDs that are mentioned in the literature, Anterior Disc Displacement with Reduction (ADDR) is the most common ID encountered in adults, with prevalence rates up to 35% (Naeije et al. 2013). It is assumed that ADDRs are almost equally distributed among the sexes, and that prevalence rates of ADDR increase during the period of adolescence (Huddleston Slater et al. 2007).

An ADDR occurs when the articular disc is anteriorly displaced relative to the condylar head in the closed mouth position and restores (reduces) its normal physiological relation with the affected condyle during mouth opening. The disc reduction is usually accompanied by a clicking, popping, or snapping sound (de Leeuw and Klasser 2013). In general, ADDR is considered a stable and painless condition, causing little or no discomfort to the patient (de Leeuw and Klasser 2013). In some patients, however, it may develop into a more serious condition, causing TMJ pain or severe jaw movement limitation (Kalaykova et al. 2011a).

Several factors, such as biomechanical and anatomical factors, have been suggested to play a role in the etiology of ADDR. Biomechanically, heavy compressive forces within the TMJ during oral habit activities, such as gum chewing and pen biting, might contribute to a tendency of the articular disc to be dislodged off the condyle (Kalaykova et al. 2011a). Anatomically, space insufficiency within the joint to accommodate both the condyle and disc is thought to force the disc anteriorly (Naeije et al. 2013). This assumption is supported by the fact that the closing click occurs just before the condyle re-enters the glenoid fossa (Huddleston Slater et al. 2007). Unfortunately, little is known about if and why this space insufficiency within the TMJ occurs. Besides the two mentioned factors, there are also indications that psychological factors play a role in the occurrence of TMJ sounds (Spruijt and

Wabeke 1995). In a recent study, it appeared that TMJ sounds in adolescents were associated with a propensity to somatisation and concerns with body image (Ukra et al. 2017). However, as both studies made use of self-reported information regarding the presence of TMJ sounds, it remains unknown whether the association between psychological factors and the presence of TMJ sounds also applies for studies that make use of objective assessments. Therefore, this study aimed to evaluate the clinically determined prevalence of ADDR among children and adolescents, and to evaluate potential risk indicators related to this. So far, most knowledge of risk indicators of ADDR among children and adolescents comes from studies performed on Caucasian subjects (e.g., Egermark et al. 2001; Huddleston Slater et al. 2007; Kalaykova et al. 2011b). As ethnicity may be of influence on the anatomy of the TMJ complex (Fletcher 1985; Jasinevicius et al. 2005), this might be reflected in differences in occurrence and risk indicators of ADDR when other ethnic groups are investigated. Data from this study will, therefore, be collected among young Asian subjects.

## **Materials and methods**

This study was approved by the ethics committee of Trisakti University-School of Dentistry, Jakarta, Indonesia (No.142/KE/FKG/10/2014). Three groups of participants were recruited for this study. The first group, henceforth referred to as children, comprised of children between 7 and 12 years of age. The second group comprised of adolescents aged 13-18 years old, whereas the last group consisted of young adults aged 19-21 years old. Participants were recruited from national elementary schools, high schools, and a dental school in the greater area of Jakarta. To represent the general population in Indonesia, we selected schools in rural and urban areas, and schools representing high and low socioeconomic status (SES). Regarding the latter, socioeconomic data were derived from local government

offices. The participants' inclusion criteria were that elementary school pupils had to be aged 7-12 years, whereas the high school pupils and university students were aged 13-21 years at the time of data collection. In addition, all participants had to speak the Indonesian language and had to be in good health as indicated in their routine health report.

Prior to the data collection, elementary school class teachers distributed information leaflets about TMDs and bruxism, and an informed consent letter to pupils' parents or legal representatives. After consent was provided, parents or legal representatives were asked to fill in the questionnaire together with their child/children. Questionnaires had to be returned within one week after they were distributed. For high school pupils, notifications from the school regarding this study were provided in pupils' daily report books, which were to be read and signed by their parents. As for university students, notifications on the content of the study and data collection schedules were distributed to their email address. On the day of the data collection, two researchers who collected the data at high schools and the dental school gave a brief explanation of the study goals. After this, the pupils and students had to complete the questionnaire in the class under the supervision of two researchers.

### *Questionnaire*

To investigate the role of potential risk indicators for the presence of ADDR, the Indonesian version of a Dutch questionnaire was employed. This questionnaire has been used in a previous study among adolescents (van Selms et al. 2013), and includes items on demographic status, sleep bruxism, and oral habits (Table 1). Two versions of the questionnaires were created: one for children in elementary schools, that made use of the parental report, and a self-report questionnaire for adolescents and young adults.



### *Clinical examination*

The clinical examination was performed according to guidelines implemented in the research diagnostic criteria for temporomandibular disorders (RDC/TMD) (Dworkin and LeResche 1992). The RDC/TMD suggest the following criteria for the diagnosis of an ADDR on clinical examination: 1. Reciprocal TMJ clicking, reproducible on at least two of three opening and (loaded) closing movements; AND 2. Elimination of the TMJ clicking on protrusive opening and closing movements (Dworkin and LeResche 1992; Huddleston Slater et al. 1999). Each TMJ was examined separately by having the examiner place one fingertip on the skin overlying one TMJ while the other hand stabilised the head.

### *Reliability study*

Prior to the study, a pilot study was conducted to assess the test-retest reliability of the questionnaire, and the intra-examiner reliability of the examiners in detecting ADDR. For the questionnaire, reliability was estimated for each item by calculating intra-class correlation coefficients (ICC) using absolute agreement. The test-retest reliability of the parental-report questionnaire, to be completed by parents or caretakers of children aged 7-12, was assessed by distributing the questionnaire to 50 parents or legal representatives. An explanation brochure with pictures of grinding and clenching activities was attached to the questionnaire. This procedure was essential because the two terms are not commonly used in the Indonesian language. The adults were instructed to read the explanation page and fill in the questionnaire accompanied by their child. Regarding the self-report questionnaire, the questionnaire was distributed among 75 high school pupils. Just before the pupils filled in the questionnaires, the definition of teeth grinding and clenching was explained. Ten days later, the distribution of the questionnaire was repeated. The ICC scores were interpreted according to Fleiss (Fleiss et al. 2003): ICCs < 0.4 were

considered poor; 0.4-0.75 as fair-to-good; and  $>0.75$  as excellent. The questionnaire items, both derived from the parental report and self-report questionnaires, were found to have above fair-to-good interclass ICC scores (0.45 – 0.96). The inter-rater reliability of three calibrated examiners was assessed by examining 50 high school pupils. The ICC for the presence of ADDR sounds was 0.82, which is qualified as excellent.

### *Data analysis*

Descriptive statistics were used to describe the data characteristics, such as the frequency and distribution of demographic characteristics, along with means and standard deviations of participants' age. For both parental-report and self-report, regression models were used to assess risk indicators. First, single logistic regression analysis was performed to establish the unadjusted associations between ADDR and each of the independent variables. Each time, the linearity of the ordinal independent variables to the outcome variable was checked by analysis of dummy variables. When the regression coefficients of the dummy variables consistently increased or decreased, linearity was considered present. Otherwise, dichotomisation of the variables was conducted. When the relation or dependency between the outcome variable and the independent variable was strong enough (P-value  $<0.10$ ), this variable was incorporated into a multiple regression model. Independent variables with the weakest association with ADDR were removed using the backward stepwise approach, and the P-to-exit was reported. All variables in the final model had a P-value  $<0.05$ . Analyses were conducted using IBM SPSS statistics for windows version 24 (Armonk, NY: IBM Corp.).

## Results

During a six-month period, data of 546 children (mean (SD) age 9.5 ( $\pm$ 1.7) years), 812 adolescents (mean (SD) age 15.0 ( $\pm$ 1.5) years), and 204 young adults (mean (SD) age 20.0 ( $\pm$ 0.8) years) was collected. The prevalence rate of ADDR was 7.0% in the child population, 14.4% in the adolescent population, and 12.3% in the young adult group. As can be seen in figure 1, there appears to be an increase in prevalence with age, with a peak found in the adolescent group. To test whether the group of adolescents differed from the children and young adults with respect to the presence of ADDR, the database was split into three groups of equal age distribution (group A: 7-11 years; group B: 12-16 years, and group C:17-21 years). Analysis using Chi-square test showed a statistically significant difference in the occurrence of ADDR between these three groups ( $\chi^2(2) = 21.969$ ,  $p < 0.001$ ). Comparison between groups showed that significant difference existed between groups A and B ( $\chi^2(1) = 22.936$ ,  $p < 0.001$ ), and between groups A and C ( $\chi^2(1) = 12.044$ ,  $p < 0.05$ ). However, there was no significant difference between groups B and C ( $\chi^2(1) = 0.886$ ,  $p > 0.1$ ), as indicated in Figure 1.

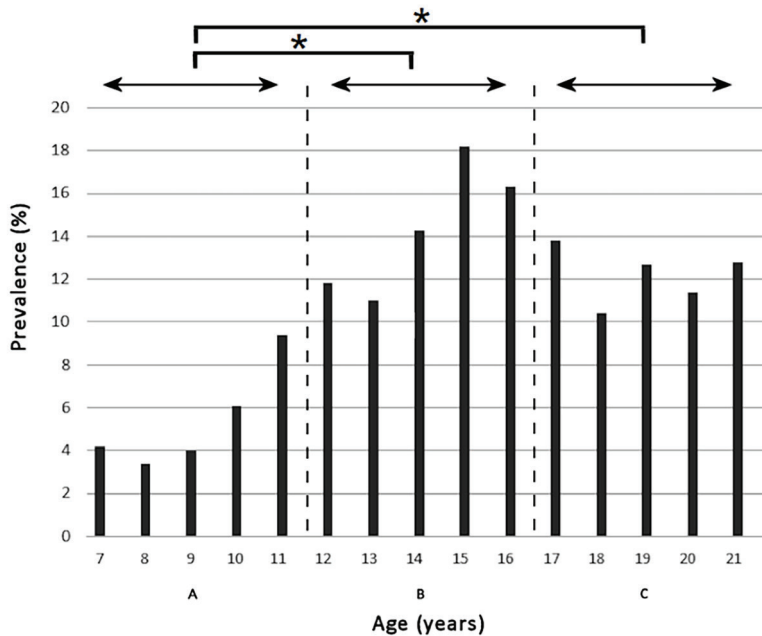


Figure 1. Prevalence rates (%) of temporomandibular joint anterior disc displacement with reduction (ADDR) in Indonesian young population.  
 \*Significant difference was detected between prevalence rates in 7-11 years old (group A) and 12-16 years old (group B), and between group A and 17-21 years old (group C).

The multiple regression analysis indicated that increasing age (OR:1.28, CI:1.02-1.60) and lip biting (OR:2.47, CI:1.16-5.26) were the strongest predictors of ADDR in the child population (Table 2). Regarding the adolescent population, the multiple regression analysis indicated that pen biting (OR:1.70; CI:1.16-2.49) was associated with ADDR, as shown in Table 3. The psychological factor ‘worrying’ just did not reach significance (P-value of 0.052). Regarding the young adults, no factors were found to be associated with ADDR (table 4).

Table 1. Items in the questionnaire. Except for demographical data and orthodontic treatment, all questions referred to the last 30 days.

	Parental report (children 7-12 years)	Self-report (Adolescents – young adults 13-21 years)
Demographic data	<ul style="list-style-type: none"> <li>- Gender (Male/Female)</li> <li>- Age (Years)</li> </ul>	
Sleep Bruxism	Does your child grind or clench his/her teeth while sleeping? (No/Yes/Don't know)	Have you been told, or did you notice yourself that you grind or clench your teeth when you are sleep? (No/Yes/Don't know)
Awake bruxism		Have you been told, or did you notice yourself that you grind or clench your teeth during the day? (No/Yes/Don't know)
Oral habits	<ul style="list-style-type: none"> <li>- Does your child chew gum? (No/Yes)</li> <li>- Does your child bite her/his nails? (No/Yes)</li> <li>- Does your child bite on pens/pencils? (No/Yes)</li> <li>- Does your child bite his/her lips and/or cheeks? (No/Yes)</li> </ul>	<ul style="list-style-type: none"> <li>- Do you chew gum? (No/Yes)</li> <li>- Do you bite your nails? (No/Yes)</li> <li>- Do you bite on pen/pencils? (No/Yes)</li> <li>- Do you bite on your lips and/or cheeks? (No/Yes)</li> </ul>
Psychological factors	<ul style="list-style-type: none"> <li>- Does your child worry about things? (No/Rarely/Sometimes/Often/Always)</li> <li>- Does your child experience pressure and/or tension from the home situation? (No/Rarely/Sometimes/Often/Always)</li> <li>- Is your child easily scared? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you think your child is in a state of mental tension when he/she gets home from school? (No/Rarely/Sometimes/Often/Always)</li> </ul>	<ul style="list-style-type: none"> <li>- Do you worry about things? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you experience pressure and/or tension from the home situation? (No/Rarely/Sometimes/Often/Always)</li> <li>- Are you easily scared? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you think you're in a state of mental tension when you get home from school? (No/Rarely/Sometimes/Often/Always)</li> </ul>

Table 2. Single regression and multiple logistic regression analysis to assess associated factors of ADDR in the child population (7-12 years old). Associations are expressed as Odds Ratio (OR) and 95% confidence interval (CI).

Predictor variable	<i>Single regression models</i>			P-to-Exit	<i>Multiple regression model; n=545</i>		
	P-value	OR	95% CI		P-value	OR	95% CI
Gender (Female)	0.501	1.27	0.63-2.57	0.060	0.030	1.28	1.02-1.60
Age	0.010	1.33	1.07-1.65				
Sleep bruxism (yes)	0.710	1.14	0.56-2.33				
Oral habits							
- Gum chewing (yes)	0.112	1.73	0.66-2.68				
- Nail biting (yes)	0.006	2.55	1.14-4.54		-	-	-
- Pen biting (yes)	0.514	0.76	0.27-1.66				
- Lip biting (yes)	0.001	3.48	1.35-5.92		0.019	2.47	1.16-5.26
Psychological factors							
- Worrying	0.670	1.16	0.54-2.30				
- Home tense	0.233	1.61	0.74-3.53				
- School tense	0.540	1.26	0.40-1.50				
- Scared	0.453	0.77	0.60-2.68				

Table 3. Single regression and multiple logistic regression analysis to assess associated factors of ADDR in adolescents (13-18 years old). Associations are expressed as Odds Ratio (OR) and 95% confidence interval (CI).

Predictor variable	<i>Single regression models</i>			P-to-Exit	<i>Multiple regression model; n=812</i>		
	P-value	OR	95% CI		P-value	OR	95% CI
Gender (Female)	0.508	1.04	0.92-1.19				
Age	0.854	1.04	0.70-1.54				
Sleep bruxism (yes)	0.469	1.16	0.78-1.72				
Awake bruxism (yes)	0.736	0.90	0.49-1.65				
Oral habits							
- Gum chewing (yes)	0.434	1.18	0.78-1.77				
- Nail biting (yes)	0.133	1.36	0.91-2.04				
- Pen biting (yes)	0.002	1.87	1.26-2.80		0.005	1.78	1.19-2.67
- Lip biting (yes)	0.029	1.58	1.05-2.38	0.187	-	-	-
Psychological factors		1					
- Worrying	0.024	1.75	1.08-2.83	0.052	-	-	-
- Home tense	0.506	1.15	0.77-1.71				
- School tense	0.150	1.34	0.90-1.98				
- Scarred	0.960	1.01	0.67-1.51				

Table 4. Single regression and multiple logistic regression analysis to assess associated factors of ADDR in young adults (19-21 years old). Associations are expressed as Odds Ratio (OR) and 95% confidence interval (CI).

Predictor variable	<i>Single regression models</i>			P-to-Exit	<i>Multiple regression model; n=204</i>		
	P-value	OR	95% CI		P-value	OR	95% CI
Gender (Female)	0.303	0.61	0.24-1.57				
Age	0.971	1.01	0.60-1.69				
Sleep bruxism (yes)	0.172	2.03	0.74-5.58				
Awake bruxism (yes)	0.268	0.31	0.04-2.44				
Oral habits							
- Gum chewing (yes)	0.378	0.51	0.11-2.29				
- Nail biting (yes)	0.861	1.10	0.38-3.14				
- Pen biting (yes)	0.708	1.28	0.35-4.74				
- Lip biting (yes)	0.866	1.10	0.44-2.66				
Psychological factors							
- Worrying	0.516	0.75	0.32-1.77				
- Home tense	0.550	0.77	0.33-1.80				
- School tense	0.560	0.79	0.33-1.90				
- Scared	0.902	0.94	0.35-2.50				

## Discussion

The first aim of this study was to evaluate the clinical prevalence of ADDR among Indonesian children and adolescents. It appeared that the prevalence rate of ADDR was 7.0% in the child population and 14.4% in the adolescent population, whereas clinical examinations performed in a group of young adults revealed that the prevalence was 12.3%. Increasing age and lip biting were positively associated with ADDR in children, while pen biting was associated with ADDR in the adolescent population.

For detection of TMJ sounds, this study made use of a clinical examination



following recommendations proposed by the RDC/TMD (Dworkin and LeResche 1992). Even though clinical examinations are to be preferred over interview or self-report, MRI examination is considered the imaging modality of choice for evaluation of the joint area for ADDR (Larheim 2005). However, besides the negative aspects of high costs and low availability, MRI has a high sensitivity for both intra- and extra-articular changes of TMJ components (Westesson 1993). However, as MRI detects ADDR in clinically asymptomatic subjects as well (Marpaung et al. 2014), the use of MRI to detect ADDR might be clinically less relevant because ADDR not only involves sound, but also movement interference.

In this study, it was decided not to include the question about awake bruxism in the parental report questionnaire. The main reason was that it could be difficult for parents to differentiate awake bruxism from concomitant normal oro-motor activities involved in wakefulness that might be present in their children. On the other hand, as the detection of awake bruxism is mostly based on awareness of tooth clenching or bracing activities during wakefulness, this question was included in the self-report questionnaire for adolescents and young adults.

The present findings demonstrate that prevalence rates of ADDR increase during the period of adolescence. The same trend was observed in studies on Caucasian children and adolescents (Egermark et al. 2001; Huddleston Slater et al. 2007; Kalaykova et al. 2011b). This increase of prevalence among children and adolescents fits with the suggestion that ADDR development is related to space insufficiency within the joint (Huddleston Slater et al. 2007). It is known that until children reach the age of six, the glenoid fossa is shallow and the articular eminence is not apparent (Bjork 1963). The articular eminence then undergoes rapid change during the mixed dentition period and slows down at around the age of ten years, which is just before the adolescent growth spurt begins. Meanwhile, the mandibular condyle grows steadily during childhood, followed by a growth spurt at around 15

years of age (Bjork 1963; Nahhas et al. 2014). As a result, growth spurt might lead to incongruence between the articular eminence and the condyle. As the articular disc serves as a space-correcting apparatus in the TMJ complex, its displacement to the anterior part would thus compensate this incongruence. This might explain the significant peak in ADDR prevalence found in this study in the group of adolescents aged 12-16 years as compared to other groups.

As with several other studies, this study seems to indicate that ADDR prevalence rates remain stable from adolescence into adulthood (Huddleston Slater et al. 2007; Isberg et al. 1998). This corroborates with the suggestion that the occurrence of ADDR is related to the growth of TMJ components, because it is known that both maxilla and mandible bone structures cease growth at approximately the age of 20 years (Nahhas et al. 2014). A further longitudinal study is, however, needed to investigate this assumption.

Oral habits were positively associated with ADDR, both in children and adolescents. This coincides with several other studies (Gavish et al. 2000; Winocur et al. 2001). Gavish et al. found an association between gum chewing habit and ADDR (Gavish et al. 2000), whereas Winocur et al. noted that jaw playing was the most significant oral habit related to the presence of joint sounds (Winocur et al. 2001). A possible explanation for the association between oral habits and ADDR is that they induce large stresses on the articular disc as depicted in finite element analysis models (Hirose et al. 2006). Excessive stresses force the disc to be dislodged off the condyle to the anterior side (Nitzan 2003). The effect of loading was also demonstrated in an experimental study by Kalaykova et al., in which intensive chewing exercises resulted in a loss of reducing capacity of the articular disc (Kalaykova et al. 2011a).

It has been suggested that ADDR prevalence is more common among girls

than boys (Dibbets and van der Weele 1992). On the other hand, in other studies on children and adolescents, no gender difference was observed (Huddlestone Slater et al. 2007; Keeling et al. 1994). In the present study, female gender was not found to be a risk indicator for ADDR. More research is needed to elucidate this point.

In this study, the psychological factor ‘worrying about things’ was positively associated with the presence of ADDR in the adolescents’ single regression analysis. This (unadjusted) association might be explained by the presumed close relationship between psychological factors and oral habits (Leme et al. 2014). Psychological factors such as stress, depression, and anxiety have shown to have strong associations with some forms of oral habits (Leme et al. 2014). Although this factor dropped out in the multiple regression analysis (P-to-exit 0.052), the purported association would be that psychological distress increases oral habits occurrence, eventually leading to the development of ADDR.

Anthropological studies on body growth and TMJ anatomical shape have indicated that variation can exist in bone morphology between certain ethnic groups. For example, a study using cephalometric radiographs showed a significant difference between Caucasian and Asian adolescents with respect to the mandible and cranium base relation (Wu et al. 2007). Other studies showed that Caucasians seem to have a steeper articular eminence than Asians (Fletcher 1985; Wu et al. 2007). As a steep eminence inclination has been indicated as a predisposing factor for ADDR (Sato et al. 1996), it might be hypothesised that ADDR prevalence rates of Asian children are lower than those among Caucasians. Direct comparison between results obtained in this study and those of other studies with different ethnicities, however, could not be performed because of differences in ADDR assessment protocols. Future observational studies with standardised diagnostic criteria might shed light on potential ethnic differences concerning the occurrence of ADDR.

## **Conclusions**

In conclusion, within the limitations of the present study, our findings indicate that there appears to be an increase in ADDR prevalence with age, with a peak found during the years of adolescence. The observed association of ADDR with oral habits suggests that biomechanical factors play a significant role in ADDR development. Longitudinal studies to confirm the current findings are highly needed.

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## **Chapter 4**

### **Prevalence and risk indicators of pain-related temporomandibular disorders among Indonesian children and adolescents**

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## Authors' contributions

	Study conception	Study design	Data acquisition	Data analysis	Interpretation of data	Drafting the manuscript	Critically revising the manuscript
Marpaung C	√	√	√	√	√	√	√
van Selms MKA	√	√		√	√	√	√
Lobbezoo F	√	√		√	√		√



## **Abstract**

**Objectives:** To assess the prevalence of pain-related temporomandibular disorders (TMDs) among Indonesian children and adolescents, and to investigate which risk indicators are associated with it.

**Materials and Methods:** In this cross-sectional study, 1,800 questionnaires were distributed among pupils of schools in the greater Jakarta area. This was done for two samples: children with ages ranging from 7 to 12 years (parental report) and adolescents aged 13-18 years old (self-report).

**Results:** The prevalence rates for pain-related TMDs in Indonesian children and adolescents were 23.4% (95% CI=20-27) and 36.9% (95% CI=33-41), respectively. Regression models revealed that psychological factors and the presence of bodily pain were strongly associated with pain-related TMDs in both children and adolescents, next to oral habits (in children), and sleep and awake bruxism (in adolescents). On the other hand, the socioeconomic status of parents was not associated with pain-related TMDs in either sample.

**Conclusions:** Pain-related TMDs are common among the young Indonesian population. These findings corroborate those from earlier studies of young populations, namely that bruxism and oral habits, bodily pain complaints, and psychological factors are risk indicators for pain-related TMDs.

## **Introduction**

Temporomandibular disorders (TMDs) is a collective term embracing a variety of temporomandibular joint (TMJ) disorders, masticatory muscle disorders, headache disorders, and disorders affecting the associated structures (de Leeuw et al. 2013; Peck et al. 2014). Among the various clinical problems that fall under this term, most research focuses on pain-related TMDs (LeResche 1997). The rationale for this is that, in most cases, pain has a higher impact in terms of individual suffering than signs related to TMDs, such as TMJ sounds or deviated mandibular movements (LeResche 1997). Pain-related TMDs may interfere with daily activities such as eating and talking (de Leeuw et al. 2013; Dworkin et al. 1990; LeResche 1997). The pain typically has a mild character and is usually transient (de Leeuw et al. 2013; Dworkin and Massoth 1994). However, for some individuals, the pain can become a chronic and persistent condition (Karibe et al. 2015; Scrivani et al. 2008). It is commonly reported that bruxism and oral habits, bodily pain complaints, female gender, and a variety of psychological factors are risk indicators for pain-related TMDs (Fillingim et al. 2011; Maixner et al. 2011a; Michelotti et al. 2010).

It is generally acknowledged that pain-related TMDs are most frequently observed in the adult population between 18 and 45 years old, with prevalence rates up to 25% (de Leeuw et al. 2013; LeResche 1997). On the other hand, TMD pain is also commonly reported among children and adolescents, with prevalence rates ranging between 4% and 30% (Barbosa Tde et al. 2008; Sena et al. 2013). This wide range is most likely due to differences in methodology, diagnostic instruments, and sample characteristics among the various studies. Regarding the latter, as pointed out by Fernandes et al. (2015), there are indications that adolescents in underdeveloped and developing countries are more likely to report pain-related TMDs than those raised in developed countries (Fernandes et al. 2015). For example, in several studies conducted with adolescents in Brazil and Iran (Bonjardim et al. 2005b; Ebrahimi et al. 2011; Fernandes et al. 2015), both considered as developing

countries, higher prevalence rates for pain-related TMDs were found than in northern European countries (Kohler et al. 2009; List et al. 1999). So far, only a few studies on TMD pain have been conducted on children and adolescents living in developing countries located in Asia. The authors of one of these studies, conducted in China, concluded that TMD pain was relatively common among Chinese adolescents, and that its occurrence was associated with socioeconomic status (SES) and type of living area (rural vs urban) (Hongxing et al. 2016). Similar to China, Indonesia is considered a developing country with marked socioeconomic disparities (OECD). The majority of the Indonesian population is 19 years old or younger (Nuraini; Wahyuni 2015). Indonesia is therefore a suitable country to learn more about the nature of pain-related TMDs in young people, and to investigate their associations. Therefore, the aim of this questionnaire study was to assess the prevalence and associations of pain-related TMDs among Indonesian children and adolescents.

## **Materials and Methods**

### *Study Design*

This study was approved by the ethics committee of Trisakti University School of Dentistry, Jakarta, Indonesia (No.142/KE/FKG/10/2014). The sample size was calculated using  $\alpha=0.05$ , power 95%, and effect size of 0.143 based on 25% of TMD pain prevalence in adolescents from a previous study, (Fernandes et al. 2015) which yielded a minimum size of 1,267 participants. Participants were recruited from national elementary schools and high schools located in rural and city areas in the greater area of Jakarta. Indonesian elementary school children are typically 7 to 12 years old, whereas adolescents attending high schools are 13 to 18 years old. In Indonesia, the level of SES can be inferred from the type of school. Accordingly, only schools of the highest and lowest SES from each area were chosen to participate.

This was done using socio-economic data obtained from local government agencies. The inclusion criteria for this study were that: the pupils had to speak the Indonesian language as their mother language; they should be aged 7-12 years for elementary school and 13-18 years for high school at the time of data collection; and they should be in good general health as indicated in their school health report.

For pupils attending the elementary schools, an information leaflet about pain-related TMDs and bruxism, and an informed consent letter were distributed to all parents or legal representatives by the class teachers. Parents or legal representatives were asked to fill in the questionnaire together with their child/children. Questionnaires had to be returned within one week after they were distributed. For high school pupils, notifications from school regarding this study were provided in pupils' daily report books, which were to be read and signed by their parents. On the day of data collection in the high schools, two researchers who collected the data gave a brief explanation about the study goals. After this, the pupils completed the questionnaire in the class under supervision of the teacher and the two researchers.

The questionnaire in this study was adapted from a Dutch questionnaire that was used earlier to investigate TMDs and bruxism among Dutch adolescents (Table 1). (van Selms et al. 2013) The items in the questionnaire were slightly modified in order to be used for parental report in the child sample. The original questionnaire was translated into the Indonesian language using a forward/backward translation protocol (Ohrbach 2013). Subsequently, the reliability of each item in the questionnaire was established by calculating the intraclass correlation coefficient (ICC) using absolute agreement. To that end, the questionnaire was distributed among 50 parents or legal representatives of elementary school children (aged 7-12 years) and 75 high school adolescents (aged 13-18 years). The questionnaire was again distributed after 10 days. All items in the questionnaire, both for children's

parents and for adolescents, appeared to have fair-to-good (0.4 – 0.75) or excellent (>0.75) test–retest reliability scores according to Fleiss (Fleiss et al. 2003).

Table 1. Items included in the questionnaire directed to the children (parental report) and adolescents (self-report). Except for the demographic data, all questions referred to the last 30 days.

	Parental report (children 7-12 years)	Self-report (Adolescents 13-18 years)
Outcome Variable		
Pain-related TMDs	- Does your child have pain at the location of his/her temples, face, in front of the ear, or in the ear? (No/Yes)	- Does your child have pain at the location of his/her temples, face, in front of the ear, or in the ear? (No/Yes)
Independent Variable		
Demographic data	<ul style="list-style-type: none"> <li>- Gender (Male/Female)</li> <li>- Age (Years)</li> <li>- School socioeconomic status (Low/High)</li> <li>- Living area (Rural/Urban)</li> </ul>	
Sleep bruxism	Does your child grind or clench his/her teeth while sleeping? (No/Yes/Don't know)	Have you been told, or did you notice yourself that you grind or clench your teeth while sleeping? (No/Yes/Don't know)
Awake bruxism		Have you been told, or did you notice yourself that you grind or clench your teeth during the day? (No/Yes/Don't know)
Oral habits	<ul style="list-style-type: none"> <li>- Does your child chew gum? (No/Yes)</li> <li>- Does your child bite on her/his nails? (No/Yes)</li> <li>- Does your child bite on pens/pencils? (No/Yes)</li> <li>- Does your child bite on his/her lips and/or cheeks? (No/Yes)</li> </ul>	<ul style="list-style-type: none"> <li>- Do you chew gum? (No/Yes)</li> <li>- Do you bite on your nails? (No/Yes)</li> <li>- Do you bite on pen/pencils? (No/Yes)</li> <li>- Do you bite on your lips and/or cheeks? (No/Yes)</li> </ul>
Psychological factors	<ul style="list-style-type: none"> <li>- Does your child worry about things? (No/Rarely/Sometimes/Often/Always)</li> <li>- Does your child experience pressure and/or tension from the home situation? (No/Rarely/Sometimes/Often/Always)</li> <li>- Is your child easily scared? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you think your child is in a state of mental tension when he/she gets home from school? (No/Rarely/Sometimes/Often/Always)</li> </ul>	<ul style="list-style-type: none"> <li>- Do you worry about things? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you experience pressure and/or tension from the home situation? (No/Rarely/Sometimes/Often/Always)</li> <li>- Are you easily scared? (No/Rarely/Sometimes/Often/Always)</li> <li>- Do you think you're in a state of mental tension when you get home from school? (No/Rarely/Sometimes/Often/Always)</li> </ul>
Bodily Pain	Does your child have other pain complaints? (for example: abdominal pain, back pain) (No/Yes)	Does your child have other pain complaints? (for example: abdominal pain, back pain) (No/Yes)

### *Data analysis*

Descriptive statistics were used to summarise the features of the collected data. To analyse the roles of psychological factors and oral habits, responses to the questions were grouped into three categories: no positive items, 1-2 positive items, and 3-4 positive items. Single logistic regression analyses were performed to establish the associations between the outcome variable and each of the independent variables. Each time, linearity of the ordinal independent variables to the outcome variable was checked by analysis of dummy variables. When the regression coefficients of the dummy variables consistently increased or decreased, then linearity was considered present. Otherwise, dichotomisation of the variables was conducted. When the relation or dependency between outcome variable and independent variable was strong enough (P value <0.10), this predictor was incorporated into a multiple regression model. Predictors with the weakest association with pain-related TMDs were removed using the backward stepwise approach, and the P-to-exit was reported. All predictors in the final model had a P value <0.05. All analyses were conducted using IBM SPSS statistics for windows version 23 (Armonk, NY: IBM Corp.).

### **Results**

From May to October 2014, 1,800 questionnaires were distributed among elementary and high school pupils aged 7-18 years old. In total, 1,378 questionnaires were returned (23.0% loss to non-response). Of the data collected at the elementary schools, 21 pupils were excluded because they were 13 years or older. Based on the reports of parents or legal guardians of the 545 children aged 7-12 (mean 9.5 + 1.7), the prevalence of pain-related TMDs was 23.4% (95% CI=20-27). The prevalence of self-reported pain-related TMDs among the 812 adolescents aged 13-18 years (mean 15.1 + 1.5) was 36.9% (95% CI=33-41). As can be seen in Figure 1, the presence of TMD pain among adolescents demonstrates

that the prevalence was consistently higher in the older age groups. The distribution of participants with pain-related TMDs is shown in Table 2, stratified by the predictive variables. Gender, SES, living area, sleep bruxism, awake bruxism, and bodily pain are depicted as dichotomous variables.

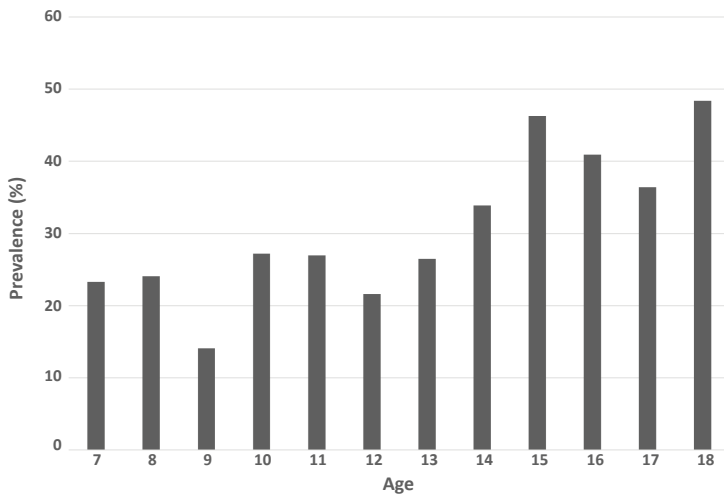


Figure 1. Prevalence of pain-related TMDs by different age group.

Based on the parental-report information, the single regression analyses indicated that sleep bruxism, awake bruxism, oral habits, 3-4 psychological factors and bodily pain were associated with pain-related TMDs among children (Table 3). The multiple regression model identified the following factors to be associated with pain-related TMDs in children: the reports of 1-2 oral habits and 3-4 oral habits, the presence of bodily pain, and the report of 3-4 psychological factors.

In the adolescent sample, single regression analyses indicated that age, sleep bruxism, awake bruxism, oral habits, psychological factors, and bodily pain were associated with pain-related TMDs, as shown in Table 4. The multiple regression model indicated that the following factors were associated with pain-related TMDs in adolescents: the reports of awake bruxism and sleep bruxism, the presence of bodily pain, and the reports of 1-2 psychological factors and 3-4 psychological factors.

Table 2. Pain related TMDs prevalence stratified by the predictive variables.

Variable	Children (age 7-12; n=545)			Adolescents (age 13-18; n=812)		
	No TMD n (%)	TMD pain n (%)	Combined	No TMD n (%)	TMD pain n (%)	Combined
Gender						
Boys	166 (75.5)	54 (24.5)	220	207 (65.9)	107 (34.1)	314
Girls	239 (77.1)	71 (22.9)	310	204 (60.5)	133 (39.5)	337
Socioeconomic status						
Low	246 (75.7)	79 (24.3)	325	162 (62.1)	99 (37.9)	261
High	159 (77.6)	46 (22.4)	205	249 (63.8)	141 (36.2)	390
Living Area						
Rural	149 (76.8)	45 (23.2)	194	150 (61.5)	94 (38.5)	244
Urban	256 (76.2)	80 (23.8)	336	261 (64.1)	146 (35.9)	407
Sleep bruxism						
Absent	304 (79.6)	78 (20.4)	382	287 (68.7)	131 (31.3)	418
Present	97 (68.3)	45 (31.7)	142	68 (52.3)	62 (47.7)	130
Awake bruxism						
Absent	-	-	-	360 (66.7)	180 (33.3)	540
Present	-	-	-	37 (46.8)	42 (53.2)	79
Oral habits:						
- None	146 (85.9)	24 (14.1)	170	79 (72.5)	30 (27.5)	109
- 1-2 oral habits	220 (74.8)	74 (25.2)	294	228 (66.5)	115 (33.5)	343
- 3-4 oral habits	39 (59.1)	27 (40.9)	66	104 (52.3)	95 (47.7)	194
Psychological factors						
- None	108 (83.1)	22 (16.9)	130	74 (84.1)	14 (15.9)	88
- 1-2 factors	224 (79.7)	57 (20.3)	281	193 (68.9)	87 (31.1)	280
- 3-4 factors	73 (61.3)	46 (38.7)	119	144 (50.9)	139 (49.1)	283
Bodily pain						
Absent	318 (82.0)	70 (18.0)	388	205 (74.5)	70 (25.5)	275
Present	87 (61.3)	55 (38.7)	142	206 (54.8)	170 (45.2)	376



Table 3. Single and multiple logistic regression model for predicting pain related TMDs in the child population.

Predictor variable	Single regression models			P-to-Exit	Multiple regression model; n=545		
	OR	95% CI	P value		OR	95% CI	P value
Gender							
Male	1						
Female	0.93	0.62-1.40	0.729				
Age	1.03	0.91-1.16	0.652				
Socioeconomic status							
Low	1						
High	0.91	0.60-1.38	0.665				
Living area							
Rural	1						
Urban	0.91	0.70-1.61	0.791				
Sleep bruxism							
No	1						
Yes	2.00	1.32-3.07	0.001	0.088			
Oral habits							
None	1						
1-2 oral habits	2.02	1.22-3.35	0.007		1.71	1.01-2.90	0.046
3-4 oral habits	4.21	2.19-8.10	< 0.001		2.35	1.15-4.80	0.019
Psychological factors							
None	1						
1-2 factors	1.23	0.71-2.11	0.461		0.99	0.56-1.74	0.963
3-4 factors	3.09	1.72-5.57	<0.001		1.96	1.04-3.69	0.038
Bodily pain							
No	1						
Yes	2.82	1.84-4.32	<0.001		2.19	1.39-3.47	0.001

Table 4. Single and multiple logistic regression model for predicting pain related TMDs in the adolescent population.

Predictor variable	Single regression models			P-to-Exit	Multiple regression model; n=812		
	OR	95% CI	P value		OR	95% CI	P value
Gender				0.325			
Male	1						
Female	1.26	0.92-1.74	0.155				
Age	1.15	1.04-1.28	0.009				
Socioeconomic status							
Low	1						
High	0.93	0.67-1.28	0.645				
Living area							
Rural	1						
Urban	0.89	0.64-1.24	0.497				
Sleep bruxism							
No	1						
Yes	1.74	1.26-2.40	0.001		1.67	1.17-2.39	0.005
Awake bruxism							
No	1						
Yes	2.27	1.41-3.66	0.001		1.86	1.12-3.08	0.016
Oral habits				0.171			
None	1						
1-2 oral habits	1.33	0.83-2.14	0.243				
3-4 oral habits	2.41	1.45-3.98	0.001				
Psychological factors							
None	1						
1-2 factors	2.38	1.28-4.45	0.006		2.37	1.20-4.69	0.013
3-4 factors	5.10	2.75-9.46	<0.001		4.81	2.42-9.57	<0.001
Bodily pain							
No	1						
Yes	2.42	1.72-3.39	<0.001		1.55	1.07-2.25	0.020

## **Discussion**

The aims of this questionnaire study were to assess the prevalence rates of pain-related TMDs and its associated factors among children (aged 7-12) and adolescents (aged 13-18) living in Indonesia. The overall prevalence of pain-related TMDs in the child population was 23.4%, whereas it was 36.9% in the adolescent population. In both samples, psychological factors and the presence of bodily pain were among the strongest predictors of pain-related TMDs, next to the reports of oral habits (children), and awake and sleep bruxism (adolescents).

Completion of a questionnaire is often the method of choice in large-scale epidemiology studies, because large numbers can be collected efficiently that in turn can provide good statistical power. The subjective nature of this method, however, may give rise to questions on its validity due to inherent risks of over or underscoring the condition. Nevertheless, as pain is a subjective entity, it is what the individual who suffers from pain says it is. Therefore, a single question as implemented in the Axis I instrument of the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) (Schiffman et al. 2010) was used in order to detect orofacial pain indicative of TMD pain. On the other hand, since no clinical examinations were performed, it was not possible to set a definite diagnosis of pain-related TMDs, nor to distinguish this pain from other pain conditions that might be present in the orofacial area, such as headache and toothache.

This study made use of two different versions of a questionnaire originally constructed by Van Selms et al. (van Selms et al. 2013). Since children aged 7 to 12 years old may encounter difficulties in comprehending the questions, parental report was used to acquire information in the child population. Potential risks of parental reports, however, include inaccurate memory of events, desire for acquiescence, and inadequate interpretation of behaviour (Damon et al. 2010). It was therefore decided that these questionnaires had to be filled in by parents or legal representatives accompanied by the child/children. Nevertheless, we decided not to include the

question about awake bruxism in the parental report questionnaire for it can be difficult for parents to differentiate awake bruxism from concomitant normal oromotor activities involved in wakefulness that might be present in their children. Self-report was used to acquire information from the adolescents. As the interrelationship between these two data acquisition methods with respect to the gold standard (a diagnosis of TMD pain based on clinical examinations) is unknown, it was decided not to statistically compare both samples.

Responses to the questions on psychological factors and oral habits were categorised in three categories. A consequence of this is that the roles of individual items could not be observed. However, as the questions used in the study were not designed as diagnostic tools, but rather as a tool for screening on potential associated factors, this approach gives an overall view of the relations of psychological factors and oral habits with TMD pain.

The obtained findings indicate that pain-related TMDs are common among Indonesian children and adolescents. Even though the observed prevalence rates for children (23.4%) and adolescents (36.9%) are more or less in line with those of other studies, (Feteih 2006; Sonmez et al. 2001) comparisons with earlier studies are always hampered by methodological issues (such as differences in the age range or collection method). On the other hand, our findings support the general assumption that the prevalence rate of TMD pain is lowest among young children, after which it increases during adolescence (Hongxing et al. 2016; LeResche et al. 2005b; List et al. 1999).

Based on the multiple regression models, it appeared that the number of psychological problems was one of the strongest predictors for pain-related TMDs, especially in the adolescent population. The interrelation between TMD pain and psychological factors has also been observed in several other studies on children and adolescents (List et al. 2001; Pereira et al. 2009). The common rationale is that the

same neurotransmitters, especially serotonin and norepinephrine, are involved in both pain and mood regulation (Linton and Bergbom 2011; Trivedi 2004).

In this study, oral habits (viz., chewing gum, and/or biting on nails, pens/pencils, and/or lips/cheeks), bruxism, and bodily pain were found to be associated with pain-related TMDs. This is in line with the findings of several other studies performed on child and adolescent populations (Fernandes et al. 2015; LeResche et al. 2005b; Winocur et al. 2006). The presumed mechanism behind this is that pain-related TMDs might arise in the form of delayed onset muscle soreness (DOMS), induced by excessive loading of the masticatory system from oral habits and bruxism activity (Koutris et al. 2013). As for bodily pain, it is generally acknowledged that other bodily pain conditions, like neck, back, and joint pain, are considered comorbidities in pain-related TMDs (LeResche et al. 2007; LeResche et al. 2005b).

In the present study, we did not find any relation between female gender and the presence of pain-related TMD complaints, neither among children nor adolescents. Although it is often reported that the prevalence of pain, in general, is higher among women, conflicting results are common in pain-gender studies (Fillingim et al. 2009) as well as in TMD pain studies (Bonjardim et al. 2005b; LeResche et al. 2007; Nilner and Kopp 1983; Nilsson et al. 2007).

Finally, low SES has been mentioned as a potential risk factor for orofacial pain in adults (Aggarwal et al. 2003; Riley et al. 2003). With this premise in mind, the relation between SES and pain-related TMDs was analysed in this study. The purported association, however, was not found. The majority of studies on children's health highlight the psychological impacts related to the SES. Psychological factors were found to be more important in explaining the difference in prevalence of pain in socio-economically contrast areas (Brekke et al. 2002; Davies et al. 2009), which might also be the case in this study. Therefore, we suggest that the focus of future

studies on TMD pain should be more on the children's psychological status than on their socioeconomic status.

## **Conclusions**

This study showed that pain-related TMDs are common among the young Indonesian population. In both children and adolescents, psychological factors and the presence of bodily pain were among the strongest predictors of pain-related TMDs, next to oral habits (children), and sleep and awake bruxism (adolescents). On the other hand, the socioeconomic status of parents appeared to be of no influence on pain-related TMDs prevalence in both children and adolescent population.

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## **Chapter 5**

### **Temporomandibular disorders among Dutch adolescents: prevalence and biological, psychological, and social risks indicators**

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## Authors' contributions

	Study conception	Study design	Data acquisition	Data analysis	Interpretation of data	Drafting the manuscript	Critically revising the manuscript
Marpaung C	√	√	√	√	√	√	√
Lobbezoo F	√	√			√		√
van Selms MKA	√	√	√	√	√	√	√

## **Abstract**

**Objective:** To assess the prevalence rates of pain-related temporomandibular disorders (TMDs) and temporomandibular joint (TMJ) sounds in a large group of Dutch adolescents, aged between 12 and 18 years old, and to determine if the same biological, psychological, and social risk indicators are related to both TMD pain and TMJ sounds.

**Materials and methods:** In this cross-sectional questionnaire survey, 4,235 questionnaires were analysed, with an about equal gender distribution.

**Results:** The overall prevalence of pain-related TMDs was 21.6% (26.1% for girls, 17.6% for boys); that for TMJ sounds was 15.5% (19.3% for girls, 11.7% for boys). Logistic regression analyses revealed that the following variables appeared to be the strongest predictors of TMD pain: female gender, increasing age, sleep bruxism, biting on lips and/or cheeks, stress, and feeling sad. Regarding self-reported TMJ sounds, the multiple regression model revealed that female gender, increasing age, awake bruxism, and biting on lips and/or cheeks were the strongest predictors.

**Conclusions:** TMDs are a common finding among Dutch adolescents. Except for the psychological factors that appeared to be associated with TMD pain only, pain-related TMDs and TMJ sound shared similar biological risk indicators.

## **Introduction**

Temporomandibular disorders (TMDs) is a collective term that embraces a variety of temporomandibular joint (TMJ) disorders, masticatory muscle disorders, headache disorders, and disorders affecting the associated structures (Peck et al. 2014; Schiffman et al. 2014). One way of classifying the different types of TMDs is by dividing them into two broad categories: 1. pain-related TMDs; and 2. intra-articular TMDs (Schiffman et al. 2014). Regarding the first category, pain can originate from the TMJs, but more frequently the masticatory muscles are involved (Okeson and de Leeuw 2011; Zakrzewska 2013). Pain-related TMDs are usually transient over time and resolve without serious long-term effects (de Leeuw and Klasser 2013; Dworkin and Massoth 1994). Intra-articular TMDs are expressed by biomechanical signs like TMJ sounds (clicking and crepitation), jaw locking, and limited mouth opening (Schiffman et al. 2014). TMJ sounds are the most common expression of intra-articular TMDs (Farrar and MacCarty 1982b), and usually occur without pain or jaw movement limitation (Greene and Laskin 1988; Kononen et al. 1996). Even though both categories of TMDs are primarily present among young and middle-aged adults (Kohler et al. 2009; LeResche 1997), studies performed on children and adolescents seem to indicate that the prevalence of pain-related forms of TMDs increases with increasing age in this age group (Hirsch et al. 2012; Kohler et al. 2009; Nilsson 2007). Likewise, several studies on intra-articular TMDs report an increase of TMJ sounds in the young population (Huddleston Slater et al. 2007; Kononen et al. 1996; Wänman and Agerberg 1990).

It is generally believed that a variety of biological, psychological, and social factors may reduce the adaptive capacity of the masticatory system, thus resulting in TMDs (de Leeuw and Klasser 2013; Suvinen et al. 2005). Since pain-related TMDs and TMJ sounds represent clusters of related disorders in the masticatory system (de Leeuw and Klasser 2013), this would imply that overlap exists among the risks indicators for both categories of TMDs. For instance, it is commonly believed that teeth grinding or jaw clenching (i.e., bruxism) causes TMD pain due to overloading of

the musculoskeletal structures (Svensson and Graven-Nielsen 2001). At the same time, bruxism-induced overloading of the TMJs that exceeds the normal adaptive capacity might result in more TMJ sounds due to degenerative changes of the anatomical structures, or a tendency of the disc to be dislodged off the condyle (Naeije et al. 2013; Tanaka et al. 2008). Surprisingly, many risk assessment studies on TMDs in the young population focused on one category of TMDs only (Fernandes et al. 2015; Kononen et al. 1996; Nilsson 2007), whereas in others the various signs and symptoms of TMDs were merged into one overall TMD diagnosis (Karibe et al. 2015; Pereira et al. 2009; Sermet Elbay et al. 2017). As it is, however, generally agreed that TMDs represent a nonspecific umbrella term, it is essential to differentiate pain-related TMDs from intra-articular TMDs. The aims of the present study, therefore, were (1) to assess the prevalence rates of self-reported pain-related TMDs and TMJ sounds in a large group of adolescents aged between 12 and 18 years old, (2) to determine their associations with biological, psychological, and social risks indicators, and (3) to determine if the same risk indicators are related to both categories of TMDs.

## **Materials and methods**

### *Data collection*

This investigation was designed as a cross-sectional, population-based study. During three subsequent semesters, participants were drawn from among adolescents attending nine Dutch secondary schools that were willing to participate in this investigation. Because of time demand or other priorities at that time, 23 schools declined participation. All approached schools were dispersed over the southern and western parts of The Netherlands, and were situated in urban areas. Prior to the data collection, the parents/legal representatives received an information letter about the study. The children and/or the parents/legal representatives had the right to refuse participation.

On the day of data collection, a questionnaire was handed over to the schools'

pupils and collected several minutes later, before the lessons started. This questionnaire contained 17 items that covered demographic items, sleep and awake bruxism, signs and symptoms of TMDs, and psychosocial and behavioural factors (van Selms et al. 2013). Most questions were derived from already existing questionnaires, like the Dutch translation of the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) (Lobbezoo et al. 2005), and an oral habits questionnaire (van der Meulen et al. 2006). During the time the questionnaires were completed, the pupils were supervised by the class teacher and the investigators as to ensure that the questionnaires were completed individually. Due to this approach, the participation rate was 100%. The institutional review board of the Academic Centre for Dentistry Amsterdam (ACTA) and the school boards of the participating schools approved the data collection procedures. Prior to the investigation, the feasibility of the research process was field-tested in a pilot study. In addition, the test-retest reliability of the employed questionnaire was assessed, yielding fair-to-good to excellent reliability scores. For detailed information about the data collection methods, see van Selms et al. (van Selms et al. 2013).

#### *Outcome variables*

- Orofacial pain, indicative of TMD pain, was assessed by means of the following question: ‘Have you had pain in the face, jaw, temple, in front of the ear or in the ear?’ (no, yes). The question referred to the presence of pain within the last month.
- The presence of TMJ sounds was assessed using the question ‘Does your jaw make a clicking or popping sound when you open or close your mouth, or while chewing?’ (no, yes). The question referred to the presence of TMJ sounds within the last month.

Since no clinical diagnoses were established in this study, the term “pain-related TMDs” has to be interpreted as “pains indicative of TMD pain”, and “TMJ sounds” as “self-perceived TMJ sounds”.



### *Independent variables*

#### a. Biological items

- Age (years) and sex (0, 'male'; 1, 'female').
- The presence of sleep bruxism was assessed using the question 'Have you been told, or did you notice yourself, that you grind your teeth or clench your jaws when you are asleep?'. The presence of awake bruxism was assessed using the question 'Do you grind your teeth or clench your jaws during the day?'. These questions referred to the last month, and the pupils could choose between no, yes, or unknown. Other oral activities that may be stressful to the masticatory system were asked by the following four questions: Do you chew on chewing gum? Do you bite your nails? Do you bite on pens/ pencils? Do you bite your lips/ cheeks? Again, these questions referred to the last month, and the answer possibilities were: no, occasionally, regularly, often, very often.
- The following exogenous aspects were assessed: 'Do you smoke cigarettes?', and 'Do you drink alcohol?' (both questions: no, occasionally, regularly, often, very often).

#### b. Psychological items

- An impression of the psychological status was assessed by means of the following two questions 'Are you stressed?' and 'Are you feeling sad?' (both questions referred to the last month: no, occasionally, regularly, often, very often).

#### c. Social items

- Ethnic background was classified following the method of Statistics Netherlands (CBS), using the country of birth from both parents. This procedure resulted in a classification into two subgroups, viz., native Dutch (i.e., both parents were born in the Netherlands, regardless of the country of birth of the subject; coded '0') and non-native Dutch (i.e., all other subjects; coded '1').

- Educational level was characterised by the type of the secondary educational system that was followed. Depending on their abilities, Dutch children around the age of 12 can choose for either vmbo, vmbo/havo, havo, havo/vwo, or vwo. The vmbo diploma gives access to advanced vocational education, the havo diploma to polytechnic education, and the vwo diploma gives access to university education. The 5-point Likert scale item educational level was recoded into a dichotomous variable (vwo [1] versus the other levels [0]).

### *Data analysis*

Descriptive statistics included frequency distributions of each of the independent variables. In order to determine the prevalence rates of TMD pain and TMJ sounds, the prevalence data were stratified by gender and age, and ratios were calculated. The Chi-square test was performed to test the association between TMD pain and TMJ sounds as depicted in a 2x2 contingency table. To determine the association between the outcome variables and each of the independent variables, hierarchical logistic regression analyses were performed. First, single regression analyses were executed to determine the associations between each of the various predictors and the outcome variable. Regarding the ordinal variables, initial analyses were based on the full range of the 5-point Likert response options, and linearity of their effect on the presence of TMD pain was checked by analysis of dummy variables. When the regression coefficients of the dummy variables consistently increased or decreased, linearity was considered present. In case of a non-linear association, the variable was dichotomised. Second, independent variables that showed at least a moderate association with the outcome measure were entered in a multiple regression model. Due to the fact that the large sample size may impact the corresponding P-values, a more conservative level of significance was chosen (i.e., P-value < 0.05 instead of P-value < 0.1). Subsequently, the variables with the weakest association with the outcome variable were removed from the multiple regression model. This was repeated in a backward stepwise manner

until all variables that were retained in the model showed a P-value < 0.01; for each removed independent variable, the P-to-Exit is reported. Of the independent variables included in the final models, the odds ratios and their confidence intervals are reported. All analyses were conducted using the IBM SPSS Statistics 24 software package (Armonk, NY: IBM Corp.). The data in the multiple regression model were checked for multicollinearity, using a tolerance value <0.10 and a variance inflation factor >10.

## **Results**

Initially, a total of 4,285 pupils, with ages ranging from 10 to 22 years old, completed the questionnaire. Since the present study focuses on TMD pain during adolescence, the data of pupils under twelve years (children) and above eighteen years (adults) old were excluded (n = 42; <1% of the total number). An additional eyeball verification of the paper questionnaires was performed in order to check the face validity of the data. In case a pupil deliberately had noted only extremes on all single items, this questionnaire was removed from further analysis (n=8). Therefore, the final sample consisted of 4,235 adolescents with a mean age of 14.5 ( $\pm$  1.6) years old (Table 1). Of the 3,940 adolescents who completed the question about gender, 1,966 (49.9%) were girls. In addition, 82.0% of the adolescents were classified as native Dutch, and 43.7% of the pupils followed the highest educational level (vwo).

Of the 3,935 adolescents who completed the questions about gender and TMDs, the overall prevalence of pain-related TMDs was 21.6% (26.1% for girls, 17.6% for boys). The overall prevalence of TMJ sounds was 15.5% (n=3,920; 19.3% for girls, 11.7% for boys). The prevalence rates of both TMD pain and TMJ sounds, stratified by age and gender, revealed that girls had higher rates at all ages studied, and that the prevalence tended to increase with age for both genders (Figure 1). TMD pain and TMJ sounds appeared to be highly associated ( $\chi^2(1) = 176.6$ ; P <0.001).

In order to find out which biological, psychological, or social factors had the strongest

association with the presence of pain-related TMDs, logistic regression analyses were performed. In the first step, all variables were entered consecutively in a single regression model in order to determine their unadjusted association with the TMD pain. Regarding the included 5-point ordinal variables, inspection of the regression coefficients of the dummy variables revealed that perfect linearity of their effect on the presence of TMD pain was present only for the predictor 'biting lips and/or cheeks'. All ordinal variables were therefore dichotomised (no = 0; all other categories = 1). Table 2 shows the results of the single and multiple regression models. Except for the biological items gum chewing and nail biting, and the social items ethnic background and educational level, all variables had a significant association with TMD pain in the single regression model. According to the multiple regression model, the following variables appeared to be the strongest predictors of TMD pain: female gender, increasing age, sleep bruxism, biting on lips and/or cheeks, stress, and feeling blue. There were no signs of multicollinearity among the predictor variables in the final model.

Table 3 shows the results of the regression analyses with the presence of self-reported TMJ sounds as outcome variable. Again, most biological items were associated with joint sounds in the single regression model. In addition, feeling stressed and feeling sad had a significant association with TMJ sounds. The multiple regression model revealed that female gender, increasing age, awake bruxism, and biting on lips or cheeks were the strongest predictors of TMJ sounds.

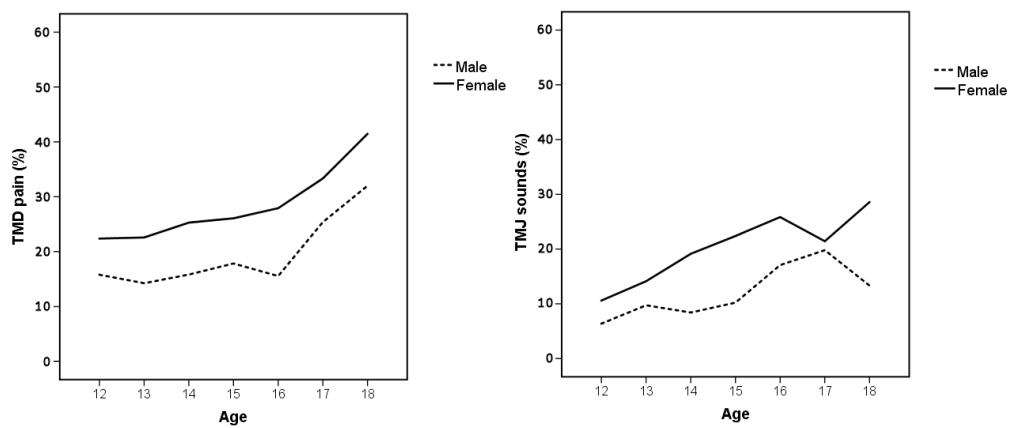


Figure 1. Age- and gender-specific prevalence of TMD pain (left) and TMJ sounds (right) among Dutch adolescents.

Table 1. Descriptive statistics of the predictor variables. The dichotomised categorical variables are presented as absolute numbers [ratio]; age is presented as mean value ( $\pm$  standard deviation).

<b>Independent variables</b>		
Age (years)		14.5 ( $\pm$ 1.6)
Gender	Male	1,974 [50.1%]
	Female	1,966 [49.9%]
Sleep bruxism	No	2,874 [82.0%]
	Yes	633 [18.0%]
Awake bruxism	No	3,334 [90.0%]
	Yes	372 [10.0%]
Chewing gum	No	261 [6.2%]
	Yes	3,943 [93.8%]
Biting nails	No	2,105 [50.0%]
	Yes	2,104 [50.0%]
Biting pens and pencils	No	2,397 [56.9%]
	Yes	1,819 [43.1%]
Biting lips and cheeks	No	1,793 [42.6%]
	Yes	2,414 [57.4%]
Smoking of cigarettes	No	3,658 [86.7%]
	Yes	559 [13.3%]
Alcohol consumption	No	2,166 [51.4%]
	Yes	2,046 [48.6%]
Being stressed	No	1,680 [39.9%]
	Yes	2,534 [60.1%]
Feeling sad	No	2,183 [51.8%]
	Yes	2,030 [48.2%]
School type	Lower levels	2,386 [56.3%]
	Highest level	1,849 [43.7%]
Ethnic background	Dutch native	3,368 [82.0%]
	Non-native Dutch	740 [18.0%]

Table 2. Single and multiple logistic regression models for the prediction of TMD pain among Dutch adolescents. Associations are expressed as Odds Ratio (OR), and 95% confidence interval (CI). For each removed predictor variable, the P-to-Exit is reported; n.s. = not significant. Significance levels are 0.05 and 0.01, respectively.

	n	Single regression			Multiple regression (n=3,131)			
		P-value	OR	95% CI	P-to-exit	P-value	OR	95% CI
<b>Biological items</b>								
Female gender	1,964	<0.001	1.66	1.42 – 1.94		0.008	1.29	1.07 – 1.55
Age (years)	4,106	<0.001	1.12	1.06 – 1.17		<0.001	1.11	1.05 – 1.17
Smoking cigarettes (positive)	559	<0.001	1.60	1.31 – 1.95	0.467	-	-	-
Drinking alcohol (positive)	2,044	<0.001	1.49	1.29 – 1.73	0.097	-	-	-
Sleep bruxism (positive)	631	<0.001	1.76	1.45 – 2.14		<0.001	1.60	1.29 – 1.98
Awake Bruxism (positive)	372	<0.001	1.93	1.53 – 2.44	0.262	-	-	-
Chewing gum (positive)	3,938	n.s.	1.00	0.74 – 1.36				
Biting nails (positive)	2,100	n.s.	0.95	0.82 – 1.10				
Biting pencils (positive)	1,816	<0.001	1.34	1.16 – 1.55	0.435	-	-	-
Biting lips and/or cheeks (positive)	2,409	<0.001	1.69	1.45 – 1.97		0.003	1.33	1.10 – 1.61
<b>Psychological items</b>								
Being stressed (positive)	1,679	<0.001	2.33	1.97 – 2.74		<0.001	1.60	1.28 – 1.99
Feeling sad (positive)	2,025	<0.001	2.14	1.84 – 2.48		<0.001	1.55	1.27 – 1.88
<b>Social items</b>								
Non-Dutch ethnicity	738	n.s.	0.97	0.80 – 1.18				
Highest educational level	1,848	n.s.	1.03	0.88 – 1.19				

Table 3. Single and multiple logistic regression models for the prediction of TMJ sounds among Dutch adolescents. Associations are expressed as Odds Ratio (OR), and 95% confidence interval (CI). For each removed predictor variable, the P-to-Exit is reported; n.s. = not significant. Significance levels are 0.05 and 0.01, respectively.

	Single regression					Multiple regression (n=3,337)		
	n	P-value	OR	95% CI	P-to-exit	P-value	OR	95% CI
<b>Biological items</b>								
Female gender	1,959	<0.001	1.81	1.51 – 2.16		<0.001	1.77	1.45 – 2.16
Age (years)	4,090	<0.001	1.19	1.13 – 1.26		<0.001	1.21	1.14 – 1.29
Smoking cigarettes (positive)	557	<0.001	1.55	1.23 – 1.94	0.156	-	-	-
Drinking alcohol (positive)	2,040	<0.001	1.53	1.29 – 1.82	0.406	-	-	-
Sleep bruxism (positive)	633	<0.001	1.62	1.30 – 2.02	0.045	-	-	-
Awake Bruxism (positive)	369	<0.001	1.98	1.53 – 2.56	0.262	<0.001	1.79	1.36 – 2.36
Chewing gum (positive)	3,922	0.046	1.50	1.01 – 2.22	0.011	-	-	-
Biting nails (positive)	2,093	n.s.	1.14	0.96 – 1.34		-	-	-
Biting pencils (positive)	1,811	n.s.	1.34	0.96 – 1.35	0.435	-	-	-
Biting lips and/or cheeks (positive)	2,406	<0.001	1.66	1.39 – 1.98		<0.001	1.46	1.19 – 1.80
<b>Psychological items</b>								
Being stressed (positive)	1,668	<0.001	1.81	1.50 – 2.17	0.042	-	-	-
Feeling sad (positive)	2,019	0.001	1.31	1.12 – 1.56	0.123	-	-	-
<b>Social items</b>								
Non-Dutch ethnicity	732	n.s.	0.81	0.64 – 1.02				
Highest educational level	1,846	n.s.	0.86	0.73 – 1.02				



## **Discussion**

The present questionnaire study aimed to assess the prevalence rates of two categories of temporomandibular disorders (TMDs), namely pain-related manifestations of TMDs and TMJ sounds, in a large group of Dutch adolescents aged between 12 and 18 years old. In addition, we examined which biological, psychological, or social risk indicators were associated with them, and if both categories of TMDs yielded similar risk indicators. The results demonstrated that self-reported TMD pain is relatively common among 12 to 18-year-old Dutch adolescents, with an overall prevalence of about 20%. Besides the fact that the occurrence of TMD pain was highly associated to that of TMJ sounds, this pain was correlated to female gender, increasing age, reports of sleep bruxism, biting on lips and/or cheeks, stress, and feeling sad. The overall prevalence of TMJ sounds was about 15%; female gender, increasing age, awake bruxism, and biting on lips or cheeks were the best predictors. Except for the psychological factors that appeared to be associated with TMD pain only, pain-related TMDs and TMJ sound shared similar biological risk indicators.

### *Prevalence of TMD pain*

It is generally acknowledged that, depending on the study, the prevalence of TMD pain in children and adolescents varies widely (Toscano and Defabianis 2009). In 2007, a large-scale study was published that focused on TMD pain among adolescents aged 12-19 (Nilsson 2007). Of the 28,899 adolescents that participated, 4.2% reported TMD pain during their annual routine examination in Public Dental Service (PDS) clinics. In another Swedish study, seven percent of the 862 adolescents from a public dental clinic was diagnosed with TMD pain (List et al. 1999). This rate was also found in a recent study on Norwegian adolescents (Ostensjo et al. 2017). The most likely explanations for the fact that the present study yielded a higher prevalence rate (viz., 21.6%) are differences in diagnostic criteria and the method of data collection. In the

present study, orofacial pain had to be present within the last month, whereas in the study by Nilsson, a time span of one week was used. In the studies by List et al. and Ostensjo et al., a clinical pain diagnosis according to the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) was set, which may have resulted in a lower prevalence. On the other hand, when these clinical criteria were applied in two Brazilian studies performed on young adolescents, it was concluded that about 25% of the schoolchildren could be diagnosed with painful TMDs (Fernandes et al. 2015; Franco-Micheloni et al. 2015). As long as no uniform diagnostic criteria are available to obtain a reliable diagnosis of TMDs in the young population, studies on this topic will continue to present a multitude of different results. Future studies must therefore aim to develop a standardised assessment tool for the young population. Unfortunately, the recently published Diagnostic Criteria for TMD (DC/TMD) (Schiffman et al. 2014) have not yet been validated for usage among children and adolescents.

#### *Risk indicators for pain-related TMDs*

Regarding the role of biological risks indicators on pain-related forms of TMDs, we demonstrated that the prevalence of TMD pain increases with increasing age in the period of adolescence. This is in line with several other studies (Kohler et al. 2009; LeResche et al. 2005a; Magnusson et al. 2005; Nilsson 2007), and coincides with the suggestion that pubertal development increases the probability of self-reported TMD pain (Hirsch et al. 2012; LeResche et al. 2005a). Moreover, girls had higher rates of TMD pain at all ages studied compared to boys (viz., 26.1% and 17.6% respectively), which corroborates with most studies on this topic (Hirsch et al. 2012; List et al. 1999; Nilsson 2007; Pereira et al. 2010). Even though it is likely that sex differences exist in basic pain mechanisms and in associated psychosocial factors, the mechanisms underlying this difference are still not well understood (Leresche 2011). Another biological factor that is frequently suggested to be associated with TMD pain in adolescents is overloading of the masticatory system due to oral habits (Fernandes et

al. 2015; Winocur et al. 2006). As a result, it was not surprising that the final regression model included sleep bruxism and the adverse oral habit ‘biting on lips and/or cheeks’.

Based on the present findings, it appeared that the two included psychological factors (viz., being stressed and feeling sad) contributed significantly to the presence of TMD pain among adolescents. Again, this is not surprising as both factors are frequently mentioned in relation to this pain (Bonjardim et al. 2005a; LeResche et al. 2005a; List et al. 2001; Wahlund 2003). The same neurotransmitters, especially serotonin and norepinephrine, are involved in both pain and mood regulation (Trivedi 2004). An increase of cortisol secretion in people with high psychological load has also been shown to be related with chronic pain development (Gatchel et al. 2007). However, caution has to be paid to this assumption, as causal links have not been clearly defined. Do these factors increase the risk of TMD pain, or are they the result of this pain because such persons have become more stressed and less cheerful by their pain condition?

Finally, the social factors ethnic background and educational level were not associated with the presence of TMD pain. The negative findings in this study might show that differences in ethnicity and educational level in Dutch adolescents do not necessarily represent different social environments in relation to the report of pain. Out of a vast range of social factors that have been considered to influence an individual’s pain behaviour, parent emotions, behaviours, and health seem to play an important role in a child’s pain experience (Palermo et al. 2014). This topic might be an interesting avenue for future research.

#### *Prevalence of TMJ sounds*

The overall prevalence of self-reported TMJ sounds was 15.5%, which is in line with the approximately 14% as reported in a recent meta-analysis on the prevalence of TMJ sounds (click or crepitation) in children and adolescents (da Silva et al. 2016). Unfortunately, the authors of that systematic review did not differentiate between

boys and girls. The gender-specific prevalence rates that we found in the present study (19.3% for girls and 11.7% for boys) seem to corroborate with those presented in earlier studies (Hirsch et al. 2012; Michelotti et al. 2016). On the other hand, even though a lower overall prevalence was found in a study of Feteih et al. (8.7% of the participants reported joint sounds), they still observed a higher prevalence in girls (Feteih 2006). It is generally acknowledged that differences in methodology lead to considerable variation in prevalence of TMJ sounds (da Silva et al. 2016). However, it can still be concluded that TMJ sounds are a commonly reported sign of TMDs in the adolescent population.

#### *Risk indicators for TMJ sounds*

As for TMD pain, four biological factors appeared to be associated with TMJ sounds. Consistent with other studies on the young population, the prevalence of TMJ sounds increased considerably with age (Huddleston Slater et al. 2007; Könönen and Nystrom 1993; Wänman and Agerberg 1990), especially during adolescence. Until now, there is no explanation for this trend. It has been suggested that increasing age leads to a temporary space insufficiency within the TMJ (Huddleston Slater et al. 2007). During the period of adolescence, the articular eminence gets its more prominent anatomical shape (Dibbets and Dijkman 1997), which can cause a lack of space within the TMJ complex (Huddleston Slater et al. 2007). As a result, this insufficient space forces the disc to be pushed from its normal position on top of the condyle to the anterior or anterolateral side during the closing movement of the mouth. The disc only resumes its normal position during the opening movement, during which TMJ sounds are produced (Kalaykova et al. 2011a; Osborn 1985). As with TMD pain, female gender was found to be associated with TMJ sounds. However, conflicting evidence exists regarding this association (Huddleston Slater et al. 2007; Riolo et al. 1987; Wänman and Agerberg 1990). As the current study utilised self-report data, the finding that prevalence rates are higher among girls might also be due to the fact that female adolescents report

physical symptoms more often than their male counterparts (Eminson et al. 1996; Larsson 1991; Powers et al. 2006). Finally, the associations found in this study between daytime clenching and/or grinding and TMJ sounds, and between biting on lips and/or cheeks and TMJ sounds corroborate with other studies (Emodi-Perlman et al. 2016; Michelotti et al. 2010; Velly et al. 2002). A possible explanation for these associations is that adverse oral activities cause compression of the articular disc as was shown in a finite element model study (Hirose et al. 2006). The occurring stresses may facilitate the disc to be dislodged off the head of the condyle to the or anterior or anterolateral side, thus creating clicking sounds upon condyle translation movements (Naeije et al. 2013; Osborn 1985).

### *Methodology*

This study has several limitations. First of all, pain-related TMDs and TMJ sounds were obtained by a questionnaire with no objective confirmation of signs and symptoms, thus being at risk of recall bias. However, high validity can exist between self-reported pain questions and the outcome of a clinical examination in adolescents (Nilsson 2007). Likewise, in a longitudinal study on signs and symptoms of TMDs in Finnish adolescents by Könönen and Nystrom, reported and clinically examined TMJ clicking sounds correlated significantly with each other (Könönen and Nystrom 1993). Second, for an indication of TMJ sounds, all pupils had to note if they experienced any clicking or popping sound when opening or closing the mouth. The presence of crepitation was, however, not asked for. Even though crepitation has a much lower occurrence in the adolescent population, if present at all (da Silva et al. 2016; Wänman and Agerberg 1990), other results might have been obtained in case this type of TMJ sound was included. Third, the present study was conducted in an adolescent population composed by non-patients. However, to fulfil the objective of determining associations with biological, psychological, and social risk indicators, different results might have been obtained in case a group of symptomatic patients was

included. Therefore, further studies should be performed with representative samples of patients with TMD pain and TMJ sounds as well. The fourth aspect that should be mentioned is that with increasing age, larger cognitive capacity, and better recall, older adolescents might remember, and therefore report any physical symptoms better than younger ones (Eminson et al. 1996). This might have influenced the obtained results with respect to prevalence and associations.

## **Conclusions**

This study indicates that both pain-related manifestations of TMDs and TMJ sounds are a common finding in the adolescent population. Both categories share similar biological risk indicators, whereas psychological factors were only associated with pain-related TMDs.

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## **Chapter 6**

### **Geographic variation of parental-reported sleep bruxism among children: comparison between The Netherlands, Armenia, and Indonesia**

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## Authors' contributions

	Study conception	Study design	Data acquisition	Data analysis	Interpretation of data	Drafting the manuscript	Critically revising the manuscript
van Selms MKA	√	√	√	√	√	√	√
Marpaung C	√	√	√	√	√	√	√
Pogosian A	√	√	√				√
Lobbezoo F	√	√			√		√

## **Abstract**

**Objective:** The aim of this study was to investigate whether geographic variation exists in the prevalence rates and associated factors of parental-reported sleep bruxism (SB) among 7-12 year old children living in three culturally different countries.

**Materials and Methods:** An identical questionnaire was completed by parents or guardians of children in The Netherlands (Europe), Armenia (West Asia), and Indonesia (Southeast Asia). Pearson's Chi-square tests were used to investigate geographic variation in occurrence; logistic regression analyses were performed to study associations.

**Results:** In total, data of 2,562 questionnaires were analysed. The overall prevalence of parental-reported SB was highest in Armenia (viz., 36.5%) compared to The Netherlands and Indonesia (19.5% and 24.2%, respectively;  $P < 0.001$ ). However, differences between countries seemed to have disappeared around the age of twelve. Geographic variation in associated factors is reflected in the fact that, depending on the country, a variety of variables were positively related with parental-reported SB (viz., younger age, and/or male gender, and/or experiencing pressure or tension from the home situation, and/or being more easily scared, and/or having difficulties in falling asleep).

**Conclusions:** Considerable geographic variation can exist in the epidemiology of parental-reported SB in children. The possibility that cultural rules and standards could explain these findings is discussed.

## **Introduction**

Sleep bruxism (SB) is a repetitive jaw-muscle activity that is characterized by clenching or grinding and/or bracing or thrusting of the mandible during sleep (Lobbezoo et al. 2013). Prevalence data have indicated that SB is a relatively common behaviour in the young population (Carra et al. 2012; Restrepo 2010). It is commonly believed that the prevalence of SB is highest among children around the age of ten to twelve, after which rates tend to decline (Kato et al. 2003; Restrepo 2010). According to a systematic review of the literature, it was concluded that the prevalence of SB in children can be highly variable between studies (Manfredini et al. 2013). Even though all papers included in that review based their diagnosis of sleep bruxism on proxy reports by the parents, prevalence rates ranged from about 5% up to 40%. This great variation in prevalence estimates was presumably due to differences in definition, age distribution, or other research methodologies.

Manfredini et al. (2013) argued that much has yet to be done to improve knowledge on the epidemiological picture of SB in the different populations (Manfredini et al. 2013). Unfortunately, no studies have attempted to compare the prevalence of this condition directly between different countries. In order to investigate the existence of potential geographic differences in SB prevalence among children, study samples of different cultural origins should be examined by employing a uniform study methodology (viz., diagnostic procedure and data collection method). Therefore, the aim of the present study was to compare the prevalence rates of parental-reported SB in 7-12 year old children, living in three culturally different countries (viz., The Netherlands [Europe], Armenia [West Asia], and Indonesia [Southeast Asia]). In contrast to The Netherlands, Armenia and Indonesia are considered developing countries, with higher levels of absolute poverty and income inequality found in Indonesia (OECD 2014). On the other hand, since Armenia declared its independence in 1991, the country has faced large economic,

political and social problems (Babloyan et al. 2008). In case low socioeconomic status (SES) and its correlates, such as lower education, poverty, and poor health, were to be of influence on parental-reported SB prevalence, we hypothesized that this would be reflected in the study results. In addition, we aimed to assess which factors were associated with parental-reported SB prevalence in each country.

## **Material and Methods**

### *Populations and sampling procedure*

In 2011, this study was initiated at the Academic Centre for Dentistry Amsterdam (ACTA) in The Netherlands. To obtain more knowledge about the prevalence and potential risk indicators of parental-reported SB, primary schools located in the western part of The Netherlands were approached by email and telephone. The schools that were willing to participate received a letter with information about this study. After permission was granted by the school board, information letters were distributed among the parents or guardians of children aged between seven and twelve years. Each information letter contained an URL (Web address) to a digital questionnaire. On request, a paper version of that questionnaire was available. The parents or guardians were asked to complete the questionnaire at home in company of their child (children). One week later, a reminder letter was distributed as to encourage the non-responders. In total, four couples of third year dental students and one sixth year dental student conducted this study that ended in 2014. Since The Netherlands is a densely populated country that belongs to the most equal countries in the world with respect to economic welfare, it was not possible to stratify on that item.

In 2014, a sixth-year dental student at ACTA replicated this study in her native country Armenia. To that end, she visited three primary schools in the capital city Yerevan, which is a modern city, and seven primary schools in small villages located in the Lori region, of which the rural residents have a higher rate of unemployment, and experience more poverty. The study was approved by the Yerevan State Medical University. In contrast to the study performed in The Netherlands, only a paper version of the questionnaire was used. This was done because of limited access to internet, especially in the rural areas.

In 2014, a PhD student based at the Trisakti University school of dentistry in Jakarta conducted this study in Indonesia, using the same protocol. Data were collected from primary schools in Jakarta and its satellite areas on Java island. The sample was stratified based on living area, which was 40% in the rural area, and 60% in the urban area. Within both areas, schools of high and low socio-economic levels were chosen. The socio-economic information was obtained from a local government office. As in Armenia, data in Indonesia was collected with a paper version of the questionnaire. Ethical clearance was granted by the ethical committee of Trisakti University.

The World Medical Association Declaration of Helsinki principles for Medical Research involving human subjects were followed to maintain the ethics. This present study did not fall under the scope of the Dutch Medical Research Involving Human Subjects Act (WMO). Participation was voluntary and anonymous, and all parents or guardians were informed and gave informed consent based on an information letter. Furthermore, the Medical Ethics Review Committee (METc) of the Vrije Universiteit Medical Center (VUmc) reviewed and approved the procedures and documents (e.g. informed consent form).



### *Data collection instrument*

The original questionnaire was constructed in 2011 by using questions that were derived from existing questionnaires, such as the Dutch version of the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) (Lobbezoo et al. 2005), the Dutch version of the Strengths and Difficulties Questionnaire (SDQ) (van Widenfelt et al. 2003), and a questionnaire that had already been used in an earlier study on bruxism among Dutch adolescents (van Selms et al. 2013). Prior to the study, a pilot study was conducted to assess the test-retest reliability of the questionnaire. In total, 80 parents or legal representatives completed the questionnaire twice in a two-week interval. Cohen's kappa values ranged between 0.66 and 0.85, indicating that the test-retest agreement of the dichotomous questionnaire items was substantial to almost perfect. In March 2013, a question about the presence of TMJ sounds was added to the Dutch instrument, because it became clear that this information was lacking. In order to get Armenian and Indonesian versions of the modified Dutch questionnaire (viz., including the question about TMJ sounds), the data collection instrument was translated according to the forward-backward translation method, following guidelines that were also used by the World Health Organization (WHO/UNESCAP 2006).

### *Outcome variable*

The presence of parental-reported SB during the past month was assessed using the question: 'Does your child grind his/her teeth?' (no, yes, or unknown).

### *Independent variables*

- Demographic data: gender (boy or girl) and age (in years).

- The presence of orofacial pain and temporomandibular joint sounds (TMJ) was assessed by means of the following questions: ‘Does your child have pain in the face, jaw, temple, in front of the ear or in the ear? (no, yes), and ‘Does the jaw make a clicking or popping sound when your child opens or closes the mouth, or while chewing?’ (no, occasionally, regularly, often, or very often).
- An impression of the psychological status of the child was obtained by means of the following questions: ‘Does your child experience pressure and/or tension from the home situation?’, ‘Do you think your child is in a state of mental tension when he/she gets home from school?’, ‘Does your child worry about things?’, and ‘Is your child easily scared?’ (no, occasionally, regularly, often, or very often).
- Potential sleeping problems were assessed by asking ‘Does your child experience difficulties in falling asleep?’ (no, occasionally, regularly, often, or very often).

### *Statistical analysis*

Descriptive statistics were calculated for the three populations (viz., children living in The Netherlands, Armenia, and Indonesia). For the ease of use and to avoid zero-cell problems, all ordinal variables were entered as binary (viz., 0 = no; 1 = occasionally, regularly, often, or very often). Regarding the independent variables, one-way analysis of variance (ANOVA) was used to determine whether there were any statistically significant differences between the three countries in average score of the continuous variable ‘age’; for the dichotomous variables, Pearson’s Chi-square tests were employed. In order to determine the prevalence rates of parental-reported SB among children, the prevalence data were stratified by country and age, and plotted in a diagram. Again, comparisons were performed by Pearson’s Chi-squared tests. Due to the fact that the relatively large sample size of the aggregated dataset might impact the corresponding P-values, a more conservative level of significance was chosen ( $P < 0.01$ ).

To study which of the various independent variables were associated with the presence of parental-reported SB, logistic regression analyses were performed. First, for each population, single regression analyses were executed to determine the associations between each of the various independent variables and parental-reported SB. Second, independent variables that showed at least a moderate association with the outcome variable (i.e.,  $P < 0.10$ ) were entered in a multiple regression model. According to the backward elimination procedure, the variables with the weakest association with the outcome variable were removed until all variables that were retained in the final model showed a  $P < 0.05$ . The data in the multiple regression model were checked for multicollinearity, using a tolerance value  $< 0.10$  and a variance inflation factor  $> 10$ . All analyses were conducted using the IBM SPSS Statistics 24 software package (IBM Corp, Armonk, NY, USA).

## **Results**

In total, 1,800 parents or caretakers of children living in The Netherlands received the information letter including the link to the digital questionnaire, or the paper version of it. Of them, 1,168 completed the questionnaire (35% loss to non-response), of which 37 had to be excluded as they expressed their wish not to use the data for scientific research. Of the 1,060 questionnaires that were distributed In Armenia, 886 questionnaires were collected a week later (16% loss to non-response). Finally, of the 893 questionnaires that were distributed in Indonesia, 545 were returned (39% loss to non-response). Table 1 shows the sample characteristics of the variables representing the Dutch, Armenian, and Indonesian children. Even though this study focused on a homogeneous group of children aged between seven and twelve, the three groups differed in mean age as determined by one-way ANOVA ( $F(2, 2,482) = 85.1, P < 0.001$ ). As for the dichotomized variables, group differences existed for the presence of orofacial pain, experienced pressure and/or tension from the home

situation, mental tension upon returning from school, worrying, being easily scared, and having difficulties in falling asleep.

Table 1. Sample characteristics of Dutch, Armenian, and Indonesian children, depicted for each of the independent variables. Age is presented as mean value ( $\pm$  standard deviation); the dichotomized ordinal variables are presented as absolute numbers [+ ratio]. In order to test for differences, one-way ANOVA was used for the continuous variable ‘age’; Chi-squared tests were used for all other ordinal variables. An asterisk denotes significant difference ( $P < 0.01$ ).

Variable	The Netherlands (n=1,131)	Armenia (n=886)	Indonesia (n=545)	Difference between countries
Age (range 7 – 12)	10.0 (1.2)	9.1 (1.5)	9.5 (1.7)	F=85.1*
Boys	503 [44.6%]	430 [49.0%]	226 [41.5%]	Chi <sup>2</sup> =8.3
Orofacial pain	56 [5.0%]	25 [2.8%]	124 [23.4%]	Chi <sup>2</sup> =216.2*
TMJ sounds	20 [6.2%]	75 [8.5%]	55 [10.1%]	Chi <sup>2</sup> =4.0
Tension at home	243 [21.8%]	212 [24.0%]	90 [16.5]	Chi <sup>2</sup> =11.5*
School tension	264 [23.9%]	277 [31.4%]	122 [22.4%]	Chi <sup>2</sup> =19.6*
Worried	525 [46.7%]	435 [49.3%]	327 [60.0%]	Chi <sup>2</sup> =26.7*
Easily scared	302 [27.0%]	555 [62.7%]	318 [58.3%]	Chi <sup>2</sup> =294.5*
Difficulties falling asleep	681 [60.4%]	272 [30.8%]	117 [21.5%]	Chi <sup>2</sup> =295.9*

Footnote: Since the question about TMJ sounds was added to the Dutch questionnaire in March 2013, a lower number of Dutch parents/ caretakers completed this question (viz., 323).

The overall prevalence of parental-reported SB among children was highest in Armenia (viz., 36.5%), compared to The Netherlands and Indonesia (19.5% and 24.2%, respectively;  $n=2,562$ ;  $\text{Chi}^2=69.73$ ;  $P < 0.001$ ). As can be seen in Figure 1, the prevalence rates of parental-reported SB in all three countries decreased with

increasing age. At the age of twelve, there was no significant difference between the three countries (n=242;  $\text{Chi}^2=0.551$ ;  $P=0.759$ ).

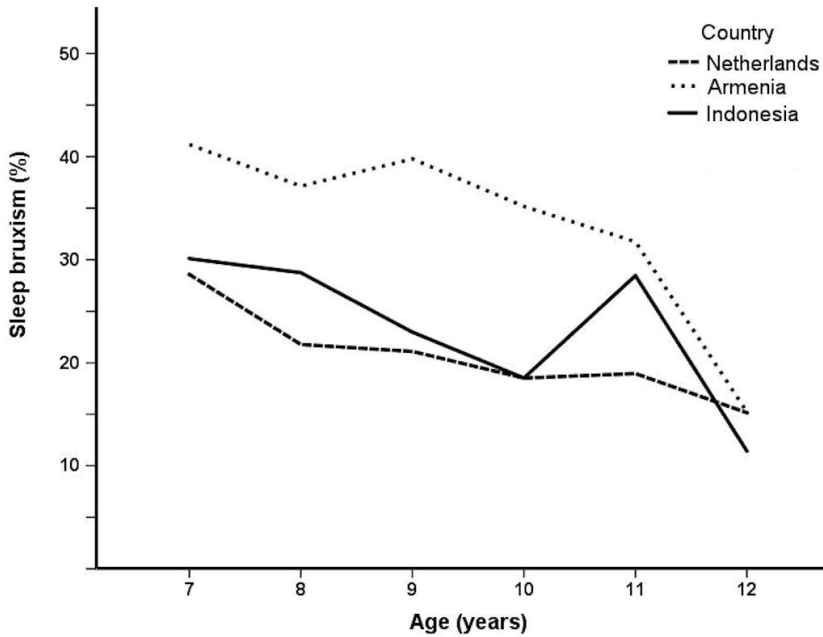


Figure 1. Parental-reported sleep bruxism according to age.

The single regression models performed among Armenian children identified the highest number of associations with parental-reported SB (viz., eight out of nine), whereas six and four associations were observed within the Dutch and Indonesian group, respectively. The multiple regression models revealed the following associations (Table 2). First, increasing age has a significant inverse relationship with parental-reported SB among Armenian and Indonesian children ( $P=0.008$  and  $P=0.041$ , respectively). Second, parental-reported SB is more prevalent among boys as compared to girls in The Netherlands and Armenia

( $P=0.041$  and  $P=0.004$ , respectively). Third, Dutch and Indonesian children, who experience pressure and/or tension from the home situation, are more likely to have parental-reported SB ( $P=0.009$  and  $P=0.013$ , respectively). Four, there is a positive association between being more easily scared and the parental report of SB in The Netherlands and Armenia ( $P=0.019$  and  $P=0.002$ , respectively). Finally, it appeared that parental-reported SB is more prevalent among Armenian children who experience difficulties in falling asleep ( $P=0.001$ ).

Table 2. Associations between parental-reported sleep bruxism and the independent variables, estimated by means of multiple logistic regression analyses (P value, Odds Ratio and 95% confidence interval). For each independent variable that was removed during the backward elimination procedure, the P-to-exit is reported.

<i>Variable</i>	<i>The Netherlands</i>			<i>Armenia</i>			<i>Indonesia</i>		
	<i>P value</i>	<i>OR (95% CI)</i>	<i>P value</i>	<i>OR (95% CI)</i>	<i>P value</i>	<i>OR (95% CI)</i>	<i>P value</i>	<i>OR (95% CI)</i>	
<i>Age</i>	n/a	1.40 (1.01-1.94)	0.008	0.87 (0.79-0.96)	0.041	0.88 (0.79-1.00)			
<i>Male gender</i>	0.041	1.40 (1.01-1.94)	0.004	1.54 (1.15-2.07)		n/a			
<i>Orofacial pain</i>	n/a	n/a		n/a	-	P-to-exit: 0.130			
<i>TMJ sounds</i>	n/a	n/a		P-to-exit: 0.062		n/a			
<i>Tension at home</i>	0.009	1.63 (1.13-2.35)	-	P-to-exit: 0.555	0.013	1.89 (1.14-3.11)			
<i>School tension</i>	-	P-to-exit: 0.366	-	P-to-exit: 0.079		n/a			
<i>Worried</i>	-	P-to-exit: 0.099	-	P-to-exit: 0.298	-	P-to-exit: 0.226			
<i>Easily scared</i>	0.019	1.52 (1.07-2.15)	0.002	1.67 (1.22-2.28)		n/a			
<i>Difficulties falling asleep</i>	-	P-to-exit: 0.455	0.001	1.65 (1.21-2.26)		n/a			

n/a = not applicable: based on the outcome of the single regression analysis, this variable was not selected for inclusion in the multiple regression.

## **Discussion**

To the best of our knowledge, this is the first study in which the prevalence of parental-reported SB is directly compared between culturally different countries. Our study shows that the parental report of SB among children living in Armenia was significantly higher as compared to that of children living in The Netherlands and Indonesia. On the other hand, the report of, for example, orofacial pain among children in Armenia was not even a tenth of that found in Indonesia. In line with this was the finding that the number of associations observed in the single regression analyses was twice as high in Armenia as compared to Indonesia (eight versus four). Explanations for the fact that prevalence rates and the number of unadjusted associations varied widely between the three countries might be found in the field of social and cultural anthropology. Regarding SB in children, the notion of tooth grinding sounds by parents is presumably shaped by experience, learning, and culture, which are all factors that depend on whether the culture values or disvalues the recognition and verbal expression of such behaviours (Peacock and Patel 2008). This might explain the current finding that the prevalence rate of parental-reported SB was much higher in Armenia as compared to that in the other two countries. From a cultural point of view, the traditional Armenian family structure and traditions play an important role in many lives (Babloyan et al. 2008). Young children frequently don't have their own bedroom. In the capital city, families often live in small apartments in large building blocks. Because of space constraints, parents have to share the master bedroom with one or more children, or children sleep together with their grandparents on beds or sofas in the living room. Also in rural parts of Armenia, family members often share a bedroom in winter time because of the costs to heat additional rooms. Since family members sleeping with their children in one room are more likely to hear a child's tooth grinding (Manfredini et al. 2017), it is more likely that these sounds are noticed in Armenia as compared to the Dutch and



Indonesian situation in which children usually have their own bedroom. In future studies on this subject, it would be wise to inquire for co-sleeping practices.

Both Armenia and Indonesia are considered developing countries, with high levels of poverty and income inequality (OECD 2014). Armenia suffers from a depressed economy resulting from economic blockades and other conflicts. Despite encouraging growth indicators, unemployment and underemployment remain substantial problems, especially for the rural parts of this country. Additional analyses performed on the Armenian database revealed that children living in the capital city Yerevan were more likely to be diagnosed with parental-reported SB than those living in small villages located in the rural parts of Armenia (data not shown; OR=0.75; 95% CI: 0.57–0.99; P=0.045). However, this significance was lost in the subsequent multiple regression. Regarding Indonesia, even though poverty numbers have shown a steady downward trend over the past decade, absolute poverty is still present on the populous island of Java (OECD 2016). Also disparities between socio-economic groups remain substantial. A comparison between schools of high and low socio-economic classes learned that there was no difference with respect to parental-reported SB report (OR=0.86; 95% CI: 0.57-1.29; P=0.458) between the two social classes (data not shown).

According to the unadjusted single regression models, the prevalence rates of parental-reported SB decreased significantly with age in Armenia and Indonesia, which corroborates with current knowledge (Carra et al. 2012; Manfredini et al. 2013). These declines led to the surprising finding that, around the age of twelve, the initial difference in occurrence of parental-reported SB between the three countries had disappeared. The conclusion by Manfredini et al. as reported in their literature review on SB in children, namely that there is no remarkable geographic pattern for SB prevalence, therefore only partly holds true (Manfredini et al. 2013). As outlined above, investigators on this subject should acknowledge the possibility that

geographic differences in parental-reported SB among children do exist. Differences that, however, seem to have disappeared around the age of twelve. Whether this decline in prevalence is caused by an actual decrease with age, or by other factors remains unknown.

The multiple regression models revealed a gender effect: the odds of parental-reported SB for boys living in The Netherlands and Armenia was 1.4 or 1.5 times larger than the odds for girls, respectively. This corroborates with the finding of a large-scale study among 6,389 primary schoolchildren in Hong Kong, namely that SB was more prevalent among boys (Lam et al. 2011). On the other hand, in the majority of included papers in the review by Manfredini et al., no gender difference was reported (Manfredini et al. 2013). Perhaps this predominance among boys living in specific countries has a cultural origin as well. Future studies should therefore acknowledge gender as a potential confounder of parental-reported SB in children.

Two psychological aspects belonged to the strongest predictors of parental-reported SB among children: pressure and/or tension experienced from the home situation (Dutch and Indonesian children), and the question about being easily scared (Dutch and Armenian children). This fits well within the observation that associations between psychological disorders and SB among children are frequently described in literature (De Luca Canto et al. 2015; Manfredini and Lobbezoo 2009). However, one should bear in mind that associations with psychological factors are more apparent in studies that adopted a diagnosis of SB based on self-report than the ones using instrumental techniques (e.g., polysomnographic or electromyographic recordings) (Manfredini and Lobbezoo 2009). Perhaps this might be due to potential bias at the diagnostic level: both self-report and parental report of tooth grinding sounds might be more prone to interfering social and cultural factors, leading to an enhanced awareness of this behaviour in specific groups.

Finally, the finding that parental-reported SB was more prevalent among Armenian children who experience difficulties in falling asleep corroborates with a recent study among Colombian children. In that study, trouble with sleeping was weakly correlated with parental-reported SB (Manfredini et al. 2017). This might be explained by the fact that both conditions fit within the concept of sleep disorders. Polysomnographic studies on sleep architecture revealed that episodes of SB are frequently accompanied by brief arousals in adult patients with SB (Macaluso et al. 1998) as well as in children with SB (Herrera et al. 2006). This, combined with the observation that SB increases in the minutes before rapid eye movement sleep (Huynh et al. 2006), has led to the suggestion that sleep stage transitions exert an influence on the motor neurons that are involved in the onset of SB (Lavigne et al. 2008).

Limitations of this study include the following. First, various tools and techniques can be applied for the assessment of SB among children. For a 'definite' diagnosis of SB, objective recordings of the sleeping subject are required (Lobbezoo et al. 2013). However, due to inherent limitations, such recordings have been used mainly for research purposes on adults rather than for the evaluation of SB among children. This study therefore made use of parental report of tooth grinding sounds, which is graded with the lowest level of diagnostic accuracy ('possible') (Lobbezoo et al. 2013). Second, even though it was attempted to achieve representative populations in each of the three countries, generalization of the findings should be made with caution. This especially accounts for Indonesia, with more than seventeen thousand islands, and inhabiting hundreds of distinct native ethnic and linguistic groups. Third, even though it was aimed to employ a uniform methodology, the mode of administration of the questionnaires differed between the countries. Since inhabitants of The Netherlands are used to complete online surveys, a Web-based questionnaire was used. Due to various reasons, this was not possible in Armenia and

Indonesia. As a recent study indicated that the mode of administration bias was limited in health interview surveys for children when paper-based questionnaires were compared with Web-based questionnaires (Mauz et al. 2018), it was expected that this difference in administration did not influence the results much.. Finally, the proportions of explained variance in the final multiple regression models were relatively low (viz., about 3 – 6%; data not shown). More research is needed to elucidate whether parental-reported SB data contain an inherent high amount of unexplainable variability, or if there are other variables not included in our models that are highly associated with parental-reported SB in 7-12 year old children.

## **Conclusions**

Despite several limitations, the following conclusions can be drawn from our research. First, geographic variation in sleep bruxism (SB) prevalence among children does exist, as the overall prevalence of parental-reported SB among 7-12 year old children was highest in Armenia (viz., 36.5%) compared to the rates observed in The Netherlands and Indonesia (viz., 19.5% and 24.2%, respectively). Differences that, however, seemed to have disappeared around the age of twelve. Second, differences in cultural rules and standards are reflected in the fact that, depending on the country, a variety of variables were positively associated with parental-reported SB (viz., younger age, and/or male gender, and/or experiencing pressure or tension from the home situation, and/or being more easily scared, and/or having difficulties in falling asleep).

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## **Chapter 7**

### **General Discussion**

Pain-related forms of temporomandibular disorders (TMDs) and temporomandibular joint (TMJ) sounds are the most common types of TMDs in the general population (de Leeuw et al. 2018). A variety of diagnostic methods and techniques can be used to assess the presence of both types of TMDs. The most widely used diagnostic protocol for researchers and clinicians to diagnose TMDs is the research diagnostic criteria for temporomandibular disorders (RDC/TMD) (Dworkin and LeResche 1992). This protocol consists of a history and physical examination, accompanied by an assessment of related psychosocial dysfunction and psychological distress. Based on new evidence, this protocol was improved and became the diagnostic criteria for temporomandibular disorders (DC/TMD) in 2014 (Schiffman et al. 2014). Both sets of criteria require self-report and clinical examination to arrive to a diagnosis. In most cases, the two methods are sufficient to identify both types of TMDs. However, especially for the detection and progression of those disorders that are accompanied by TMJ sounds, a variety of additional diagnostic devices can be applied. For example, claims have been made that jaw-tracking devices can differentiate between jaw dysfunction related to pathologic conditions of the TMJ and normal variation (Gonzalez et al. 2008). Until acceptable levels of diagnostic validity of jaw-tracking devices have been clearly established, it is uncertain if these diagnostic devices can be relied on as aids in the differential diagnosis or in clinical decision making in the field of TMDs (Baba et al. 2001; Gonzalez et al. 2008; Lund et al. 1995).

Even though TMDs are common among the adult population, with a higher incidence in women aged 20-45 years (de Leeuw and Klasser 2013; Slade et al. 2013), they can also exist in children and adolescents (LeResche 1997; Nilner and Lassing 1981). Studies on young populations of different countries yielded prevalence rates between 4% and 30% (Barbosa Tde et al. 2008; da Silva et al. 2016). The same aspect applies to the occurrence of bruxism, a jaw-muscle activity that is assumed to

be associated with TMDs (Kato et al. 2013; Svensson and Lavigne 2013). Again, a high variation of prevalence rates in children has been observed in studies on this subject, ranging from about 5% to 40% (Manfredini et al. 2013). For a large part, this wide range in prevalence rates is due to methodological differences (e.g., differences in of diagnostic criteria or data collection methods). On the other hand, regarding the occurrence of TMDs, it has been proposed that social factors and ethnic background might play a role in as well (Fernandes et al. 2015; Peacock and Patel 2008; van der Meulen et al. 2009). Likewise, sociodemographic and socioeconomic conditions have been suggested to be of influence on the occurrence of bruxism (Emodi-Perlman et al. 2016; van Selms et al. 2013). Unfortunately, the importance of social factors and ethnicity on both TMDs and bruxism has hardly been investigated.

In this chapter, the results of all studies included in this thesis directed at the above questions will be summarized, and the relations between outcomes of the different studies will be discussed.

## **Diagnostic Methods**

In all studies that are presented in this thesis, two different types of data collection methods were used in order to establish the presence of TMDs and bruxism in children and adolescents, namely clinical examination methods that assess objective data (**chapter 3**) and questionnaires for collecting subjective data (**chapters 4, 5 and 6**). Self-report can be a method of choice for large-scale epidemiology studies, as large numbers of questionnaires can be distributed efficiently. The questionnaire used in this thesis to assess TMDs and bruxism was derived from an earlier study by van Selms (van Selms et al. 2013). The original Dutch questionnaire was translated into the Armenian and Indonesian language, using a standardized forward/backward translation protocol (Ohrbach et al., 2017). We further modified

the questionnaire to be used as a parental-report questionnaire for children aged 7 to 12 years old. To minimize the risk of parents' inaccurate memory, desire of acquiescence, and inadequate interpretation of children's behavior, it was decided that the modified questionnaire had to be filled in by parents or the legal representatives together with the child/ children.

Though MRI is considered the gold standard for assessing disc position and intraarticular degenerative alterations, it is an expensive, time consuming device with low availability. Therefore, clinical examination is the preferred method to observe TMJ sounds in large-scale studies (Huddleston Slater et al. 2007). The examination of TMJ sounds caused by anterior disc displacement with reduction (ADDR) includes auscultation, palpation or both. Another means to observe TMJ sounds is by recording the condylar movement pathways using an opto-electronic tracking device such as the Oral Kinesiology Analysis System-3D (OKAS-3D)(Naeije et al. 1995). However, the validity of that specific jaw-tracking device in detecting TMJ disc displacement was not yet known. Therefore, a study was conducted to investigate the validity of two functional diagnostic methods, namely clinical examinations and OKAS-3D, in the diagnosis of ADDR (**chapter 2**). The clinical examination method showed a substantial between-method agreement with the OKAS-3D recording (Cohen's  $\kappa = 0.64$ ) regarding the presence or absence of ADDR. In other words, both functional diagnostic methods can be used interchangeably in ADDR diagnostics. However, MRI evaluation of the TMJs showed a slight to fair agreement (Cohen's  $\kappa = 0.2$ ) as compared to clinical examination with respect to the presence of ADDR. Both functional diagnostic methods appeared to have a high specificity (81.0% for clinical examination and 96.6% for OKAS-3D) when compared with MRI, which implies a low chance of having a false positive diagnosis. On the other hand, with MRI as the gold standard, the clinical examination showed a low sensitivity for diagnosing ADDR due to the

high number of positive MRI diagnoses in asymptomatic subjects (38.3% for clinical examination and 29.8% for OKAS-3D). Since ADDR is clinically relevant only when there is a function interference, a diagnosis based on functional diagnostics is recommended to be used in ADDR recognition instead of MRI. Hence, for large-scale epidemiological studies, clinical examination methods can be used for ADDR recognition, whereas OKAS-3D or other similar jaw-tracking devices are needed in evaluating the progress of ADDR.

## **Temporomandibular disorders**

Epidemiology is defined as the study of distribution, determinants, and natural history of disease in populations (Lilienfeld et al. 1994). It can uncover patterns of disease distribution in human populations, and seeks determinants of those patterns (Slade 2014). The core epidemiological concept is to determine the frequency or occurrence of an event or status (e.g., a symptom or disease). Prevalence is the standard measure of occurrence, which represents the proportion of all persons in a particular population with that disease (van der Windt et al. 2010).

In several studies on ADDR, it was noticed that prevalence rates increase between childhood and adolescence (Egermark et al. 2001; Huddleston Slater et al. 2007; Kalaykova et al. 2011b). Anatomical and biomechanical factors are among the possible contributing factors of ADDR development in this period (Atkinson and Bates 1983; Hall et al. 1985; Katzberg et al. 1996; Velly et al. 2002). Anatomical factors might involve space insufficiency during a child's growth and development, whereas biomechanical factors can involve compressive forces as a result of bruxism and adverse oral habits (Naeije et al. 2013). To learn more about ADDR prevalence and its association with a variety of contributing factors, we conducted a study among the young Indonesian population, using clinical tests based on the RDC/TMD

**(chapter 3)**. We observed that ADDR prevalence rates increase from childhood to adolescence, and tend to stabilize into adulthood. This finding partly supports the suggestion of space insufficiency as an etiological factor of ADDR. Growth spurt during adolescence may lead to a temporary incongruence between the articular eminence and the condyle. As the articular disc serves as a space-correcting structure in the TMJ complex, displacement in the anterior direction would thus compensate this incongruence (Huddleston Slater et al. 2007). Both in children and in adolescents, oral habits also appeared to be associated with ADDR occurrence. It is assumed that excessive stresses from oral habits force the disc to be dislodged off the condyle in the anterior direction (Osborn 1985). In line with this are the results of an experimental study in adults that showed that a specific type of oral habits (viz., gum chewing) could hamper the reduction capacity of the articular disc on mouth opening. Based on this, the authors of that study concluded that oral habits are of influence on the position of the disc with respect to the condyle (Kalaykova et al. 2011a). Based on our findings, we suggest that additional studies on ADDR prevalence should be conducted to learn more about the influence of biomechanical and anatomical factors associated with the period of growth and development in children and adolescents.

As with other pain conditions, TMD pain is a subjective entity that involves biopsychosocial components in the etiopathogenesis. For example, age, gender, living area, and socioeconomic status have been reported to be associated with the presence of TMD pain in the general population (Fernandes et al. 2015; Fillingim et al. 2011; Maixner et al. 2011b). To learn more about the occurrence of pain-related TMDs in the young population and its association with a variety of biopsychosocial factors, we performed a questionnaire study (**chapter 4**). The reason for conducting this study in Indonesia was because of the characteristics of that country, with marked socioeconomic disparities and a large proportion of young population.

Therefore, efforts were made to collect data from schools of different socioeconomic status and living areas. We found that TMD pain is common among Indonesia's children and adolescents. Psychological factors and the presence of bodily pain were among the strongest predictors of pain-related TMDs, next to reports of oral habits (children) and awake and sleep bruxism (adolescents). On the other hand, there was no association of TMD pain with socio-economic status.

A differentiation between pain-related TMDs and intra articular forms of TMDs (e.g., TMJ sounds) is important, because both types of TMDs are assumed to have their own pathogenesis that dictates the treatment approach. The questionnaire study in **chapter 5** was conducted to learn more about the epidemiology of TMD pain and TMJ sounds in a large group of Dutch adolescents. In addition, we tried to find out if the same risk indicators apply for both types of TMDs. This study demonstrated that self-reported TMD pain is common among 12 to 18-year-old Dutch adolescents. The occurrence of TMD pain was highly associated to that of TMJ sounds, and this pain was correlated to female gender, increasing age, reports of sleep bruxism, biting on lips and/or cheeks, stress, and feeling sad. TMJ sounds were also common among this population, and were associated with female gender, increasing age, awake bruxism, and biting on lips and/or cheeks. The observed association between oral habits or awake bruxism and the presence of TMJ sounds in children and adolescents (see **chapters 3 and 5**) indicates that biomechanical factors might contribute to the etiology of ADDR. High compressive forces from oral habits and bruxism activities might dislocate the disc in the anterior direction (Naeije et al. 2013; Osborn 1985). However, since both studies on joint sounds had a cross-sectional design, we cannot yet confirm this suggestion. Based on the study in **chapter 5**, we also concluded that, except for the psychological factors that appeared to be associated with TMD pain only, pain-related TMDs and TMJ sounds share similar biological risk indicators.

## **Bruxism**

Bruxism has often been found to be associated with both pain-related TMDs and intra-articular types of TMDs (Manfredini and Lobbezoo 2010; Poveda Roda et al. 2007; Svensson et al. 2008). The outcomes of four studies included in this thesis corroborate with this, namely that the report of bruxism was associated with both pain-related TMDs and TMJ sounds among children and adolescents (**Chapters 3 to 6**). One of the possible explanations is that excessive eccentric and concentric jaw-muscle contractions during bruxism activities can induce immediate and delayed symptoms of fatigue and/or pain in the masticatory muscles (Koutris et al. 2013; Turker et al. 2010). The immediate symptoms might be the result of an accumulation of metabolites within the muscles due to an obstruction of the muscles' blood flow during bruxism activities (Graven-Nielsen et al. 2003; Kim et al. 1999). The delayed symptoms are thought to be caused by microtrauma to the muscle fibres due to mechanical overloading and rupture of the muscle, which especially occurs during the eccentric contractions. This phenomenon is known as delayed-onset muscle soreness (DOMS) in the human masticatory muscles (Cheung et al. 2003; Turker et al. 2010).

Previous studies on bruxism among children and adolescents revealed a high variety of results regarding prevalence and its associated factors (Carra et al. 2012; Manfredini et al. 2013). This high variety might be due to differences in study methodology and variety in diagnostic criteria. In order to overcome the absence of standardized diagnostic criteria for bruxism, consensus was established among bruxism experts to resolve this matter by categorizing bruxism diagnosis into possible, probable, and definite bruxism (Lobbezoo et al. 2013).

Even though sleep bruxism among children is universal, there is still a lack of knowledge whether geographic variation exists in prevalence rates and associated factors. No studies have attempted to compare the prevalence of this condition



directly between different countries. **Chapter 6** of this thesis presents the results of a study on parental-reported sleep bruxism that was conducted among children aged 7-12 years living in three countries, namely Indonesia, The Netherlands, and Armenia, employing a uniform study methodology. We found that geographic variation in sleep bruxism prevalence among children does exist, although it seemed to level out around the age of 12 years. This geographic variation was also present with regard to the factors associated with sleep bruxism. Differences in cultural rules and standards in recognizing sleep bruxism behaviour are most likely the reason for this geographic variation.

## **Conclusions**

This thesis revealed that TMDs are common among children and adolescents in The Netherlands and Indonesia. It also showed that bruxism is a common finding among the young population of The Netherlands, Armenia, and Indonesia. We demonstrated that biopsychosocial factors are closely related to the occurrence of TMD pain and sleep bruxism in the young population, whereas anatomical and biomechanical factors seem to be associated with TMJ sounds. Since all studies in this thesis had a cross-sectional design, we were only able to indicate whether particular variables were associated with the outcome in ways that are proposed by theories. Future longitudinal research designs are essential for providing information about the temporal order of the events underlying these associations, and for observing if and how the presumed associations change over time.

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## **Chapter 8**

### **Summary**

A cross-sectional study involves the direct observation of individuals in their natural setting (often described as a “snapshot” of a group of individuals). Cross-sectional studies are not only very useful to describe certain features of a population, such as the prevalence of a disease, they may also be used to determine potential risk indicators associated with the disease. Observational research may thus be the first step for screening hypotheses that can then be studied more rigorously using a cohort study or randomised controlled study. As such, cross-sectional studies on temporomandibular disorders (TMDs) that are performed in the young population are important to learn more about the presence of these disorders. In addition, they provide information about the population burden, and they can be useful clinically by providing context for diagnostic decision-making. Unfortunately, differences in definition, diagnostic method, and age distribution of the various studies prevent drawing conclusions about the occurrence of TMDs among the young population. In addition, as the prevalence of TMDs has been established in only a few populations other than those of Western nations, the potentially important racial or ethnic prevalence patterns are poorly understood. This also accounts for bruxism, an oromotor activity that is assumed to be associated with TMDs. Even though many studies have been conducted on bruxism among children and adolescents worldwide, little is known about its occurrence among non-Caucasian subjects. And again, the variety in employed methodologies prevent drawing any firm conclusions even from the most carefully conducted review. In order to obtain a deeper insight into the occurrence and risk indicators of TMDs and bruxism among the young population, we conducted several cross-sectional studies on children and adolescents living in Indonesia and in The Netherlands. Another study focused on the potential geographic variation of bruxism. To that end, questionnaires were distributed among young populations across three socio-culturally different countries: Indonesia, The Netherlands, and Armenia.

In many cross-sectional studies, a clinical assessment of TMJ sounds is performed by means of auscultation, palpation or both. However, previous studies indicated that there can be a poor agreement when these clinical examination methods are being compared to MRI as a gold standard in recognizing articular disc displacement with reduction (ADDR). Since ADDR becomes clinically relevant only when it interferes with TMJ function, a study was conducted to investigate the validity of two functional methods in the diagnosis of ADDR, namely clinical examination (by means of auscultation and palpation) and opto-electronic movement recording with Oral Kinesiology Analysis System-3D (OKAS-3D) (**chapter 2**). We found that, for both functional diagnostic methods, the chance of having a false-positive ADDR diagnosis is low as compared with MRI. We also found that disagreement between the functional methods and the imaging method is mainly due to positive MRI findings in asymptomatic subjects. This study recommends that ADDR recognition in cross-sectional studies should rely primarily on the clinical examination approach.

ADDR is the most common internal derangement within the TMJ complex encountered in adults. It is, however, also frequently detected in younger populations, with higher prevalence rates observed among adolescents as compared to children. Anatomical and biomechanical factors are among the possible contributing factors of ADDR development. In order to learn more about ADDR prevalence and its association to a variety of potential contributing factors, a clinical study was conducted in a population consisting of children, adolescents, and young adults (**chapter 3**). This study was performed in Asia, because most knowledge on this topic comes from studies performed on Caucasian subjects. The findings of this study indicated that prevalence of ADDR increases with age, with a peak during the years of adolescence. Biomechanical factors in the form of oral habits seem to play a significant role in ADDR development.

TMD pain can have a great impact on a person's physical ability and psychosocial functioning. At the same time, the longer the pain persists, the greater the potential for amplification by means of psychosocial risk factors. Therefore, we conducted a study in order to assess the prevalence of TMD pain among Indonesian children and adolescents, and to investigate which physical and psychosocial risk indicators are associated with it (**chapter 4**). A questionnaire that was used in an earlier study on a young Dutch population was translated so that it could be used in Indonesia. Questionnaires were distributed among pupils of schools in the greater Jakarta area. This was done for two samples: children with ages ranging from 7 to 12 years (parental report) and adolescents aged 13-18 years old (self-report). This study showed that pain-related TMDs are common among the young Indonesian population. In both children and adolescents, psychological factors and the presence of bodily pain were among the strongest predictors of pain-related TMDs, followed by oral habits (children) and sleep and awake bruxism (adolescents). The socioeconomic status of parents appeared to be of no influence on pain-related TMDs' prevalence in both children and adolescent population.

It is important to differentiate between the two most common types of TMDs (viz., pain-related TMDs and intra-articular TMDs), because both types have different pathogeneses that will dictate their treatment approaches. On the other hand, one can also learn about prevention and treatment strategies if there are common risk indicators for both types of TMDs. The study in **chapter 5** was conducted on a large group of Dutch adolescents to assess the prevalence rates of TMD pain and TMJ sounds, and to determine if the same biological, psychological, and social risks indicators are related to both types of TMDs. This study indicated that both TMD pain and TMJ sounds are a common finding in the adolescent population. Both categories share similar biological risk indicators, whereas psychological factors were only associated with TMD pain.



Bruxism has often been associated to the occurrence of both TMD pain and TMJ sounds. Available studies on bruxism showed variable results regarding the prevalence and its associated factors, which is partly caused by differences in study methodology. Therefore, we conducted a study in three socio-culturally different countries, employing a uniform study methodology (**Chapter 6**). The study aimed to compare the prevalence rates of parental-reported sleep bruxism among children aged 7-12 years in the Netherlands, Armenia, and Indonesia. We found that geographic variation in sleep bruxism prevalence among children does exist. The variation also applies for the factors associated with sleep bruxism. We assume that this variation may be caused by differences in socio-cultural rules and standards.



## **Samenvatting**

Cross-sectioneel onderzoek richt zich op het observeren van individuen in hun natuurlijke leefomgeving (ook wel omschreven als een "momentopname" van een groep individuen). Cross-sectionele studies zijn niet alleen erg handig om bepaalde kenmerken van een populatie te beschrijven, zoals de prevalentie van een ziekte, ze kunnen ook worden gebruikt om potentiële risico-indicatoren te bepalen die verband houden met deze ziekte. Dergelijk observationeel onderzoek kan dus de eerste stap zijn voor het screenen van hypothesen die vervolgens op degelijke wijze kunnen worden bestudeerd met behulp van een cohortonderzoek of gerandomiseerde gecontroleerde studie. Cross-sectionele studies gericht op temporomandibulaire disfuncties (TMD's) onder jongeren zijn dus belangrijk om meer te leren over de aanwezigheid van dergelijke stoornissen binnen deze groep. Daarnaast geven ze informatie over de ziektelast en kunnen ze klinische context bieden voor diagnostische besluitvorming. Helaas zorgen verschillen in definitie, diagnostische methode en leeftijdsverdeling van de verschillende studies ervoor dat er geen conclusies kunnen worden genomen over het vóórkomen van TMD's onder jongeren. Aangezien de prevalentie van TMD's voornamelijk binnen westerse populaties is vastgesteld, zijn de mogelijk belangrijke raciale of etnische prevalentiepatronen nauwelijks bekend. Dit geldt ook voor bruxisme, een oromotorische activiteit waarvan algemeen wordt aangenomen dat deze verband houdt met TMD's. Hoewel er wereldwijd veel studies zijn uitgevoerd naar bruxisme bij kinderen en adolescenten, is er weinig bekend over het vóórkomen ervan binnen niet-blanke bevolkingsgroepen. Opnieuw verhindert de variëteit in gehanteerde methodologieën het trekken van harde conclusies. Om meer inzicht te krijgen in de prevalentie en risico-indicatoren van TMD's en bruxisme onder jongeren hebben wij diverse cross-sectionele studies uitgevoerd bij kinderen en adolescenten in Indonesië en Nederland. Een andere studie richtte zich weer op de mogelijke geografische variatie van bruxisme. Daartoe werden vragenlijsten verspreid onder

jongerenpopulaties in drie sociaal-cultureel verschillende landen: Indonesië, Nederland en Armenië.

In veel cross-sectionele studies wordt een klinische beoordeling van kaakgewrichtsgeluiden uitgevoerd door middel van auscultatie, palpatie of beide. Eerdere studies wezen echter uit dat, wanneer deze klinische onderzoeksmethoden worden vergeleken met MRI (als een gouden standaard), er een slechte overeenkomst kan zijn tussen hen in het herkennen van de zogenoemde anterieure discusverplaatsing met reductie (ADDR). Aangezien ADDR pas klinisch relevant wordt zodra het de gewrichtsfunctie verstoort, is een onderzoek uitgevoerd om de validiteit van twee functionele methoden gericht op de diagnose van ADDR te onderzoeken, namelijk klinisch onderzoek (door middel van auscultatie en palpatie) en opto-elektronische bewegingsregistratie met Oral Kinesiology Analysis System-3D (OKAS-3D) (**hoofdstuk 2**). We vonden dat, voor beide functionele methoden, de kans op een vals-positieve ADDR-diagnose laag is in vergelijking met MRI. We vonden ook dat discrepantie tussen de functionele diagnostische methoden en de beeldvormingsmethode voornamelijk te wijten is aan positieve MRI-bevindingen bij asymptomatische individuen. Deze studie beveelt de klinische onderzoeksbenadering aan voor ADDR-herkenning in cross-sectionele studies.

ADDR is de meest voorkomende *internal derangement* van het kaakgewricht (verandering van de positie of vorm van de gewrichtsweefsels) bij volwassenen. Het wordt echter ook vaak waargenomen onder jongeren, waarbij hogere prevalentiecijfers worden gevonden bij adolescenten in vergelijking met kinderen. Anatomische en biomechanische factoren zijn mogelijke factoren die bijdragen aan de ontwikkeling van ADDR. Om meer te weten te komen over de prevalentie van ADDR en de associatie ervan met verschillende potentiële risicofactoren, werd een klinisch onderzoek uitgevoerd binnen een populatie bestaande uit kinderen, adolescenten en jongvolwassenen (**hoofdstuk 3**). Deze studie werd uitgevoerd in

Azië, aangezien de meeste kennis over dit onderwerp afkomstig is van studies die zijn uitgevoerd bij Kaukasische individuen. De bevindingen van deze studie wezen erop dat de prevalentie van ADDR toeneemt met de leeftijd, met een piek tijdens de adolescentiejaren. Biomechanische factoren als gevolg van mondgewoonten lijken een belangrijke rol te spelen bij de ontwikkeling van ADDR.

TMD-pijn kan een grote invloed hebben op iemands fysieke vermogen en psychosociale functioneren. Tegelijkertijd, hoe langer de pijn aanhoudt, hoe groter de verzwarende rol van psychosociale risicofactoren kan zijn. Daarom hebben we een studie uitgevoerd om de prevalentie van TMD-pijn onder Indonesische kinderen en adolescenten te beoordelen, en om te onderzoeken welke fysieke en psychosociale risico-indicatoren hiermee zijn geassocieerd (**hoofdstuk 4**). Een vragenlijst die werd gebruikt in een eerdere studie binnen een jonge Nederlandse populatie werd vertaald zodat deze in Indonesië kon worden gebruikt. Vervolgens werden vragenlijsten verspreid onder leerlingen van scholen in het gebied in en rondom Jakarta. Dit werd gedaan voor twee groepen: kinderen in de leeftijd van 7 tot en met 12 jaar (rapportage door de ouders) en adolescenten in de leeftijd van 13 tot en met 18 jaar (zelfrapportage). Uit deze studie bleek dat pijngerelateerde TMD's geregeld voorkomen binnen de jonge Indonesische bevolking. Bij zowel kinderen als adolescenten behoorden psychologische factoren en de aanwezigheid van lichamelijke pijnen tot de sterkste voorspellers van pijngerelateerde TMD's, naast mondgewoonten (kinderen) en slaap- en waakbruxisme (adolescenten). Aan de andere kant leek de sociaal-economische status van de ouders geen invloed te hebben op de aanwezigheid van pijngerelateerde TMD's bij zowel kinderen als adolescenten.

Het is belangrijk om onderscheid te maken tussen de twee meest voorkomende typen TMD's (namelijk pijngerelateerde TMD's en intra-articulaire TMD's). Dit omdat beide typen verschillende ontstaanswijzen hebben die aldus van invloed zijn op hun behandelmethoden. Aan de andere kant kan men ook leren over

preventie- en behandelstrategieën indien er gemeenschappelijke risico-indicatoren zijn voor beide soorten TMD's. De studie in **hoofdstuk 5** is uitgevoerd bij een grote groep Nederlandse adolescenten om de prevalentie van TMD-pijn en kaakgewrichtsgeluiden te bepalen, en om te bepalen of dezelfde biologische, psychologische en sociale risico-indicatoren gerelateerd zijn aan beide typen TMD's. Deze studie toonde aan dat zowel TMD-pijn als kaakgewrichtsgeluiden een veelvoorkomende bevinding zijn in de adolescente populatie. Beide categorieën delen vergelijkbare biologische risico-indicatoren, terwijl psychologische factoren enkel geassocieerd waren met TMD-pijn.

Bruxisme wordt vaak geassocieerd met de aanwezigheid van zowel TMD-pijn als kaakgewrichtsgeluiden. Voorgaande studies naar bruxisme toonden wisselende resultaten met betrekking tot de prevalentie en de geassocieerde factoren, hetgeen deels wordt veroorzaakt door verschillen in studiemethodologie. Daarom voerden we een studie uit in drie sociaal-cultureel verschillende landen, waarbij we gebruik maakten van een uniforme studiemethodologie (**hoofdstuk 6**). De studie had als doel de prevalenties van slaapbruxisme bij kinderen van 7-12 jaar oud in Indonesië, Nederland en Armenië te vergelijken. We ontdekten dat er geografische verschillen bestaan in de prevalentie van slaapbruxisme bij kinderen. De variatie is ook van toepassing op de factoren die verband houden met slaapbruxisme. We gaan ervan uit dat deze variatie wordt veroorzaakt door verschillen in sociaal-culturele normen en waarden.





## **Kesimpulan**

Studi potong lintang adalah studi yang melibatkan pengamatan langsung pada individu dalam natur alaminya (sering digambarkan sebagai "*snapshot*" dari sekelompok individu). Studi ini tidak hanya berguna untuk menggambarkan fitur-fitur tertentu dari suatu populasi, misalnya prevalensi penyakit, tapi juga dapat digunakan untuk menentukan indikator risiko yang potensial terkait dengan penyakit. Dengan demikian, studi observasional dapat menjadi langkah pertama untuk skrining hipotesis yang kemudian dapat dilakukan dengan lebih teliti menggunakan studi kohort atau studi terkontrol secara acak. Studi potong lintang tentang gangguan temporomandibula (GTM) yang dilakukan pada populasi muda penting untuk mempelajari lebih lanjut tentang kehadiran gangguan ini. Lebih lanjut, jenis studi ini dapat memberikan informasi tentang beban populasi, dan dapat berguna secara klinis dengan memberikan konteks dalam pengambilan keputusan diagnostik. Sayangnya, perbedaan dalam definisi, metode diagnostik, dan distribusi usia dari berbagai studi menghambat penarikan kesimpulan tentang adanya GTM pada populasi muda. Sebagai tambahan, karena data prevalensi GTM pada populasi selain dari negara-negara Barat hanya terbatas, pola prevalensi rasial atau etnis menjadi kurang dipahami. Ini juga terjadi pada bruxism, suatu aktivitas oromotor yang diasumsikan terkait dengan GTM. Meskipun banyak studi bruxism telah dilakukan pada anak-anak dan remaja di seluruh dunia, hanya sedikit yang diketahui tentang bruxism pada subyek non-Kaukasia. Dan sekali lagi, variasi dalam metodologi yang digunakan menghambat penarikan kesimpulan yang pasti. Hal ini terjadi juga pada studi literatur yang dilakukan secara teliti. Untuk mendapatkan wawasan yang lebih dalam tentang kejadian dan indikator risiko GTM dan bruxism pada kalangan populasi muda, kami melakukan beberapa studi potong lintang pada anak-anak dan remaja yang tinggal di Indonesia dan Belanda. Studi lain berfokus pada variasi geografis potensial dari bruxism. Untuk itu, kuesioner dibagikan di antara populasi muda di tiga negara yang berbeda secara sosio-kultural: Indonesia, Belanda, dan Armenia.

Dalam banyak studi potong lintang, penilaian klinis bunyi TMJ dilakukan dengan cara auskultasi, palpasi atau keduanya. Namun, studi sebelumnya menunjukkan adanya kesepakatan yang buruk ketika metode pemeriksaan klinis ini dibandingkan dengan MRI sebagai standar emas dalam mendeteksi perpindahan diskus artikularis dengan reduksi (ADDR). Oleh karena ADDR menjadi relevan secara klinis hanya ketika mengganggu fungsi TMJ, sebuah studi dilakukan untuk menyelidiki validitas dua metode fungsional dalam diagnosis ADDR, yaitu pemeriksaan klinis (auskultasi dan palpasi) dan perekaman gerakan opto-elektronik dengan Oral Kinesiology Analysis System-3D (OKAS-3D) (Bab 2). Kami menemukan bahwa, untuk kedua metode diagnostik fungsional, kemungkinan memiliki diagnosis ADDR positif palsu adalah rendah dibandingkan dengan MRI. Kami juga menemukan bahwa ketidaksepakatan antara metode fungsional dan MRI terutama disebabkan oleh temuan MRI yang positif pada subjek asimtomatik. Studi ini merekomendasikan bahwa pengakuan ADDR dalam studi potong lintang harus berdasarkan pada pendekatan pemeriksaan klinis.

ADDR adalah gangguan internal yang paling umum dalam kompleks TMJ yang dihadapi orang dewasa. Gangguan ini juga telah terdeteksi pada populasi yang lebih muda, dengan tingkat prevalensi yang lebih tinggi di kalangan remaja dibandingkan dengan anak-anak. Faktor anatomis dan biomekanis adalah faktor yang berkontribusi dalam perkembangan ADDR. Untuk mempelajari lebih lanjut tentang prevalensi ADDR dan hubungannya dengan berbagai faktor kontribusi, studi klinis dilakukan pada populasi yang terdiri dari anak-anak, remaja dan dewasa muda (**Bab 3**). Studi ini dilakukan di Asia karena sejauh ini sebagian besar pengetahuan berasal dari studi yang dilakukan pada subyek Kaukasia. Temuan studi ini menunjukkan bahwa prevalensi ADDR meningkat seiring bertambahnya usia, dengan puncaknya selama masa remaja. Faktor biomekanis dalam bentuk kebiasaan oral tampaknya memainkan peran penting dalam perkembangan ADDR.

Nyeri GTM dapat mempengaruhi kemampuan fisik seseorang dan fungsi psikososialnya. Di sisi lain, semakin lama rasa sakit berlanjut, semakin besar potensi untuk amplifikasi faktor risiko psikososial. Untuk itu, kami melakukan studi untuk menilai prevalensi nyeri GTM pada anak-anak dan remaja Indonesia, dan menyelidiki indikator risiko fisik dan psikososial yang terkait dengannya (**Bab 4**). Sebuah kuesioner yang digunakan dalam studi sebelumnya pada populasi muda Belanda diterjemahkan sehingga dapat digunakan di Indonesia. Kuesioner dibagikan di antara murid-murid sekolah di wilayah Jakarta dan sekitarnya untuk dua sampel: anak-anak dengan usia mulai dari 7 hingga 12 tahun (*parental-report*) dan remaja berusia 13-18 tahun (*self-report*). Studi ini menunjukkan bahwa GTM terkait nyeri adalah umum di kalangan penduduk muda Indonesia. Pada anak-anak dan remaja, faktor psikologis dan adanya nyeri tubuh adalah salah satu prediktor terkuat dari GTM yang berkaitan dengan nyeri, di samping kebiasaan oral (anak-anak), dan bruxism saat tidur dan terjaga (remaja). Di sisi lain, status sosial ekonomi orang tua tampaknya tidak berpengaruh pada prevalensi GTM yang terkait nyeri pada anak-anak dan populasi remaja.

Diferensiasi antara dua tipe GTM yang paling umum (yaitu, GTM terkait rasa sakit dan GTM intra-artikular) adalah penting, karena kedua tipe memiliki patogen yang berbeda yang akan menentukan pendekatan penanganan masing-masing. Di sisi lain, kita juga bisa belajar tentang strategi pencegahan dan penanganan jika mengetahui indikator risiko umum untuk kedua jenis GTM. Studi dalam **bab 5** dilakukan pada sekelompok besar remaja Belanda untuk menilai tingkat prevalensi bunyi TMJ dan GTM yang terkait dengan rasa sakit, dan untuk menentukan apakah indikator risiko biologis, psikologis, dan sosial yang sama terkait dengan kedua jenis GTM. Studi ini menunjukkan bahwa kedua manifestasi terkait nyeri GTM dan bunyi TMJ adalah temuan umum pada populasi remaja. Kedua kategori berbagi indikator risiko biologis yang serupa, sedangkan faktor psikologis hanya terkait dengan nyeri GTM.

Bruxism sering dikaitkan dengan terjadinya nyeri GTM dan bunyi TMJ. Studi tentang bruxism selama ini menunjukkan hasil yang bervariasi berkenaan dengan prevalensi dan faktor terkait, yang mungkin disebabkan oleh perbedaan dalam metodologi penelitian. Oleh karena itu, kami melakukan studi di tiga negara dengan sosial budaya yang berbeda, menggunakan metodologi penelitian yang seragam (**Bab 6**). Penelitian ini bertujuan untuk membandingkan tingkat prevalensi dari bruxism tidur yang dilaporkan orang tua di antara anak-anak berusia 7-12 tahun di Belanda, Armenia, dan Indonesia. Kami menemukan bahwa variasi geografis dalam prevalensi bruxism tidur di antara anak-anak memang ada. Variasi juga berlaku untuk faktor-faktor yang terkait dengan bruxism tidur. Kami berasumsi bahwa variasi ini mungkin disebabkan oleh perbedaan dalam aturan dan standar sosio-kultural.



## **List of publications and presentations**

## List of publications

### ***Included in this thesis:***

- Validity of functional diagnostic examination for temporomandibular disc displacement with reduction  
*C Marpaung, S Kalaykova, F Lobbezoo, M Naeije*  
*Journal of Oral Rehabilitation* 2014;41:243-249
- Prevalence and risk indicators of pain-related temporomandibular disorders among Indonesian children and adolescents.  
*C Marpaung, MKA van Selms, F Lobbezoo*  
*Community Dentistry and Oral Epidemiology* 2018;46:400-406
- Temporomandibular Disorders among Dutch Adolescents: Prevalence and Biological, Psychological, and Social Risk Indicators  
*C Marpaung, F Lobbezoo, MKA van Selms*  
*Pain Research and Management*, vol. 2018, Article ID 5053709, 9 pages, 2018. <https://doi.org/10.1155/2018/5053709>.
- Temporomandibular Joint Anterior Disc Displacement with Reduction in a Young Population: Prevalence and Risk Indicators  
*C Marpaung, MKA van Selms, F Lobbezoo*  
*International Journal of Paediatric Dentistry* 2018 September 14 [Epub ahead of print]
- Geographic variation of parental-reported sleep bruxism among children: comparison between The Netherlands, Armenia, and Indonesia  
*MKA van Selms, C Marpaung, A Pogonian, F Lobbezoo*  
*Accepted in International Dental Journal*

### ***Other publications:***

- The Addition of Pharmacological Treatment to Physiotherapy in Pain Reduction of TMD-Myalgia Patients  
*C Marpaung, H Parvaneh*  
*Scientific Dental Journal* 2018;2:27-30
- Clinical considerations of dental implant systems in immediate loading cases  
*C Marpaung, RN Susilo, L Nelwan*  
*Journal of Dentistry Indonesia* 2008;13:338-42



- Partial denture rehabilitation in cleft-lip and palate patient: a case report (  
*C Marpaung, C Masulili*  
*Journal of Dentistry Indonesia 2003;10:79-85*)

### **List of presentations**

- Unravel sleep bruxism in children and adolescents  
*3rd Indonesian Academy of Craniomandibular Disorders (IACMD) meeting, Jakarta, Indonesia, 2018*
- Children with TMDs  
*18th Asian Academy of Craniomandibular Disorders (AACMD) meeting, Taipei, Taiwan, 2018*
- Predictable treatments in TMDs  
*Bali International Dental Symposium, Bali, Indonesia 2018*
- TMD treatments in daily dental practice  
*West Jakarta Dentistry, Jakarta, Indonesia 2018*
- Temporomandibular disorders in children and adolescents: Indonesia report  
*17th meeting of Asian Academy of Craniomandibular Disorders (AACMD), Jakarta, Indonesia, 2017*
- TMD treatments: What you can and cannot do  
*Dentistry courses forum, Jakarta, Indonesia, 2017*
- What to do and what not to do in TMD treatments: Serial of case reports  
*International Indonesian Prosthodontic Meeting, Solo, Indonesia 2016*
- Introduction to Obstructive Sleep Apnea (OSAS)  
*Indonesian Dental Society Meeting, Banten, Indonesia, 2016*
- TMD: A challenge in your daily practice?  
*Scientific Forum International Seminar, Jakarta, Indonesia, 2015*
- Integrated lecture in Temporomandibular Disorders (Prosthodontic-Orthodontic-Ortho-Physiotherapist)  
*Makassar Scientific Meeting VI, Makassar, Indonesia, 2015*



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