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# JAW EXERCISES IN THE TREATMENT OF MASTICATORY MYOFASCIAL PAIN – EFFICACY, PATIENTS' VIEWS AND DENTISTS' EXPERIENCES

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# Jaw Exercises in the Treatment of Masticatory Myofascial Pain - Efficacy, Patients' Views and Dentists' Experiences

## THESIS FOR DOCTORAL DEGREE (Ph.D.)

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*To my family.*

*Especially to my wife Lisa for her love, never-ending support and for reminding me of what's really important in life*

*and to my wonderful daughters Sophia, Frida and Maya for making everything worthwhile.*



## ABSTRACT

The main aim of this thesis was to gain better knowledge concerning different aspects of jaw exercises in the treatment of masticatory myofascial pain. Special emphasis was directed towards efficacy and cost-effectiveness of jaw exercises as well as patients' views and dentists' experiences.

In *study I* the general practicing dentists (GPDs) self-perceived level of knowledge, attitudes and clinical experience in treatment of temporomandibular disorders (TMD) was investigated through a web-based questionnaire. The questionnaire was based on an earlier postal questionnaire study from 2001 and it was sent to all GPDs in the Public Dental Health service, Uppsala in 2010 and 2014. The cross-sectional follow-up study design and high response rate allows for comparison of data over time. In *study II and III* the patients' experiences of jaw exercises in the treatment of masticatory myofascial pain were investigated both in a qualitative and a quantitative way. To gain a deeper understanding of the patients' experiences, 10 patients were interviewed in a semi-structured manner according to an interview guide with 10 domains. Both open-ended and follow up questions were used to encourage the patient to reflect and freely comment on the different themes. The interviews were then transcribed, and the text material was arranged and analysed through systematic text condensation (STC). The data from the qualitative interviews were then used to construct a quantitative postal questionnaire that was sent to 150 consecutive patients with masticatory myofascial pain in order to check if the data could be generalized to a larger population. In *study IV* the opinions of an international group of 14 TMD experts concerning jaw exercises in the management of TMD were investigated. A Delphi method was used where the experts, anonymous to each other, were asked to respond to statements in a web-based questionnaire according to a five-item verbal Likert scale that ranged from "Strongly agree" to "Strongly disagree". The questionnaire was answered in different rounds and the experts received a compilation of the other experts' responses in relation to their own answer after each round. The process was repeated until consensus was reached or a stability in answers between rounds was seen. Finally, in *study V* the efficacy and cost-effectiveness of jaw exercises in the treatment of masticatory myofascial pain were studied in a randomized controlled trial (RCT) that included 97 patients. Jaw exercises were compared to occlusal appliance and no treatment (waiting-list patients). The primary outcome variable was reduction of pain intensity on a visual analogue scale (VAS 0-100 mm).

The results from 2010 and 2014 showed that the GPDs (n=91 and n= 82, respectively) felt more insecure concerning TMD diagnostics, therapy decisions and treatment of TMD in children/adolescents compared to adults. The GPDs reported a high need for orofacial pain/TMD specialists and a majority of the respondents wanted the specialists to offer continuing education in TMD. The reported frequency of taking a case history of facial pain and headache increased between 2010 and 2014. In 2014, the GPDs were more secure and reported higher frequency of good clinical routines in treatment of children/adolescents with jaw exercises and pharmacological intervention compared to 2001. Interocclusal appliance

was the treatment with which most dentists felt confidence and reported good clinical routines. In the process of analysing the qualitative data of the interview study (*study II*), four main themes were identified: *Patient adherence, Symptoms, Treatment and Participation*. Some informants suspected serious disease behind their symptoms. Jaw exercises were reported to reduce pain and physical impairment. To do the jaw exercises in conjunction with an already established routine was reported to be important to enhance adherence. Some of the patients also emphasized that they wanted to continue with the jaw exercises and that they felt safe and secure that they had tools to tackle the problems themselves if the symptoms should return. The follow-up postal questionnaire study (*study III*) showed that all these results could be generalized to a larger population of patients with masticatory myofascial pain. The international expert panel (*study IV*) came to consensus that jaw exercises are effective in the treatment of myalgia in the jaw muscles and in increasing mouth opening capacity due to hyperactivity in jaw-closing muscles and disc displacement without reduction. The patients should always be instructed in an individualized jaw exercise program and also receive both verbal advice and written information about the treatment modality. Even though jaw exercises might aggravate TMD-pain in some cases, the experts considered the treatment to be without any major adverse effects. The RCT (*study V*) showed that jaw exercises, compared to no treatment, significantly reduced pain intensity, headache and consumption of analgesics in patients with masticatory myofascial pain. Jaw exercises were also shown to be more cost-effective than occlusal appliance with lower over-head cost, fewer appointments and a lower mean treatment time.

In conclusion, this thesis has shown that general practicing dentists seem to be more insecure concerning treatment with jaw exercises in children/adolescents compared to adults, but their confidence with the treatment increased over time. Patients with jaw myalgia experience that jaw exercises is an effective treatment for TMD and international orofacial pain/TMD experts recommend jaw exercises for treatment of jaw myalgia, reduced mouth opening and disc displacement without reduction. Finally, jaw exercises reduce jaw myalgia, headache and the need for analgesic medication (in the short term) and is a cost-effective treatment compared to occlusal appliance therapy.



## LIST OF SCIENTIFIC PAPERS

- I. **Lindfors E**, Tegelberg Å, Magnusson T, Ernberg M.  
Treatment of temporomandibular disorders - knowledge, attitudes and clinical experience among general practising dentists in Sweden.  
*Acta Odontol Scand.* 2016; 74: 460-5.
- II. **Lindfors E**, Hedman E, Magnusson T, Ernberg M, Gabre P.  
Patient experiences of therapeutic jaw exercises in the treatment of masticatory myofascial pain: A qualitative study.  
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- III. **Lindfors E**, Magnusson T, Ernberg M.  
Patients' experiences of therapeutic jaw exercises in the treatment of masticatory myofascial pain - a postal questionnaire study.  
*Submitted.*
- IV. **Lindfors E**, Arima T, Baad-Hansen L, Bakke M, De Laat A, Giannakopoulos N, Glaros A, Guimaraes A, Johansson A, Le Bell Y, Lobbezoo F, Michelotti A, Müller F, Orbach R, Wänman A, Magnusson T, Ernberg M.  
Jaw exercises in the treatment of temporomandibular disorders – an international modified Delphi study.  
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- V. **Lindfors E**, Magnusson T, Ernberg M.  
Efficacy and cost-effectiveness of therapeutic jaw exercises in the treatment of masticatory myofascial pain – a randomized controlled study.  
*Submitted.*

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# CONTENTS

|   |    |
|---|----|
| Introduction .....                                      | 1  |
| Pain .....  | 1  |
| Acute pain .....  | 1  |
| Chronic pain .....                                      | 1  |
| Temporomandibular disorders .....                       | 2  |
| Aetiology .....   | 2  |
| Prevalence and treatment need .....                     | 3  |
| Diagnosis .....   | 3  |
| Management .....  | 4  |
| Evaluating and reporting treatment outcome .....        | 6  |
| Physiotherapy .....                                     | 8  |
| Physiotherapy in the treatment of pain .....            | 8  |
| Therapeutic jaw exercises .....                         | 9  |
| Myalgia of the masticatory muscles .....                | 9  |
| Arthralgia of the TMJs .....                            | 11 |
| Arthritis of the TMJs .....                             | 11 |
| Disc displacement with reduction .....                  | 11 |
| Disc displacement without reduction (locking) .....     | 11 |
| Limitation of jaw movement .....                        | 12 |
| TMD associated with whiplash associated disorders ..... | 13 |
| Summary .....   | 13 |
| Research traditions.....                                | 13 |
| Aims .....  | 16 |
| General aim.....  | 16 |
| Specific aims.....                                      | 16 |
| Material and methods .....                              | 17 |
| <i>Study I</i> .....                                    | 17 |
| <i>Study II</i> .....                                   | 18 |
| <i>Study III</i> .....                                  | 23 |
| <i>Study IV</i> .....                                   | 25 |
| <i>Study V</i> .....                                    | 29 |
| Results and discussion .....                            | 33 |
| <i>Study I</i> .....                                    | 33 |
| <i>Study II</i> .....                                   | 36 |
| <i>Study III</i> .....                                  | 41 |
| <i>Study IV</i> .....                                   | 44 |
| <i>Study V</i> .....                                    | 48 |
| General discussion .....                                | 53 |
| Conclusions .....                                       | 58 |

|  |              |
|--|--------------|
| Populärvetenskaplig sammanfattning ..... | 59           |
| Acknowledgements.....                    | 62           |
| References .....                         | 64           |
| Scientific papers .....                  | Appendix I-V |

## LIST OF ABBREVIATIONS

|                |  |
|----------------|--|
| <b>ADDWoR</b>  | Anterior disc displacement without reduction                               |
| <b>DC/TMD</b>  | Diagnostic criteria for temporomandibular disorders                        |
| <b>GPD</b>     | General practicing dentist   |
| <b>HADS</b>    | Hospital anxiety and depression scale                                      |
| <b>IASP</b>    | International association for the study of pain                            |
| <b>IMPACT</b>  | Initiative on methods, measurement, and pain assessment in clinical trials |
| <b>JFLS</b>    | Jaw function limitation scale  |
| <b>NRS</b>     | Numerical rating scale   |
| <b>NSAID</b>   | Non-steroid anti-inflammatory drug   |
| <b>PGIC</b>    | Patient global impression of change  |
| <b>RCT</b>     | Randomized controlled trial  |
| <b>RDC/TMD</b> | Research diagnostic criteria for temporomandibular disorders               |
| <b>STC</b>     | Systematic text condensation   |
| <b>TENS</b>    | Transcutaneous electric nerve stimulation                                  |
| <b>TMD</b>     | Temporomandibular disorders  |
| <b>TMJ</b>     | Temporomandibular joint  |
| <b>VAS</b>     | Visual analogue scale  |
| <b>WAD</b>     | Whiplash associated disorder   |

# INTRODUCTION

## *Pain*

The international association for the study of pain (IASP) define pain as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage or described in terms of such damage” (Merskey and Bogduk, 1994). According to IASP terminology, pain can be divided into the three categories nociceptive pain, neuropathic pain and nociplastic pain (Treede, 2018). Pain is also divided into acute and chronic pain.

## *Acute pain*

Pain perception is one of the most important mechanisms for survival. Acute pain warns us about potential injury, danger and severe illness (Flor and Turk, 2015). Lack of pain perception, for example in patients with congenital insensitivity to pain, results in an increased risk of severe illness, for example orthopedic complications due to untreated fractures (Peddareddygari et al, 2014). Acute pain is most often associated with nociception, which includes activation of nociceptors in peripheral tissue and transmission of the pain signal through A $\delta$  - and c-fibers into the central nervous system (Marchand, 2008).

## *Chronic pain*

Pain is considered to be chronic when there is “persistent or recurrent pain lasting longer than 3 months” (Treede et al, 2015). Chronic pain does not have any protective value or obvious function concerning survival (Manchikanti et al, 2009). Central sensitization is defined by IASP as “increased responsiveness of nociceptive neurons in the central nervous system to their normal or subthreshold afferent input“ (Loeser and Treede, 2008) and is an important factor in the development of chronic pain (Fornasari, 2012). In economically developed countries the prevalence of chronic pain is roughly 20% (Breivik et al, 2006, Johannes et al, 2010, Leadley et al, 2012, Reid et al, 2011, Van Hecke et al, 2013). The yearly incidence of musculoskeletal pain in the population is high, where for example neck pain occur in 30 to 50% of the adult population every year (Hogg-Johnson et al, 2008). One of the most common reasons for visits to physicians in primary care is pain (Enthoven et al, 2004 and Gerdle et al, 2004). Chronic pain results in several consequences for both the individual patient and the society (Leadley et al 2012, Reid et al 2011). Patient related consequences include sleep disturbances, difficulties in managing work and basic household activities, isolation and fear of social activities (Breivik, 2006 and Breivik et al, 2013). Chronic pain can also affect the patients emotional state of mind and fear of pain and catastrophizing thoughts have been described (Edwards et al, 2016). Emotions such as anxiety and depression are common in

patients with chronic pain (Sagheer et al, 2013) and these feelings may both worsen the pain condition and increase the pain-related disability (Lerman et al, 2015). There are also economic consequences for both patient and society. Chronic pain often involves a reduced ability to work and also an increase in social insurance and health care utilization (Breivik et al, 2013 and Phillips et al, 2008). The most common treatment modalities in the management of chronic pain, such as pharmacological, psychological and physical treatment, are often not effective enough to eliminate pain completely (Turk et al, 2011). Research concerning new treatments and different treatment combinations are therefore important.

### *Temporomandibular disorders*

TMD is defined as a group of conditions affecting the masticatory system which includes the temporomandibular joints (TMJs) and masticatory muscles. Pain is one of the most common symptoms of TMD (Okeson, 1996). Other frequent symptoms of TMD include TMJ sounds (popping sound and crepitus), tiredness/stiffness of jaws and limitation of mandibular movement (Okeson, 1996). Pain due to TMD is also the most common non-dental pain condition in the orofacial area (Lipton et al, 1993). Myalgia of the masticatory muscles, arthralgia of the TMJs and headache associated with TMD are the three most frequent conditions of TMD pain (Schiffman et al, 2014). Other TMD conditions such as TMJ disc displacement with and without reduction and degenerative TMJ disease are also common (Shiffman et al, 2014).

### *Aetiology*

The aetiology of TMD is multifactorial and several factors have been suggested to predispose, initiate and prolong/maintain TMD. Etiological factors that have been discussed are trauma and parafunction, occlusal factors, psychosocial factors, genetics, hormonal factors, pain comorbidity and some general diseases, e.g. rheumatic disease (List and Jensen, 2017). Trauma can be divided into macrotrauma (due to an injury) and microtrauma (e.g. due to bruxism) and these entities might cause TMD (Macfarlane et al, 2001). Self-reported bruxism has been shown to be associated with, and also a predictor for, onset of TMD (Manfredini and Lobbezoo, 2010 and Slade et al, 2016). Clenching and grinding have also been shown to increase the risk for myofascial pain (Huang et al, 2002, Velly et al 2003). Earlier, occlusal factors were believed to be a major etiologic factor in the development of TMD (McNeill, 1997). Several epidemiological and population-based studies have not been able to show that occlusal factors have a strong association with TMD (Seligman and Pullinger, 1991). Only weak associations have been shown between malocclusions and TMD (Gesch et al, 2004 and 2005). In line with these findings, it has been concluded that orthodontic treatment neither prevents nor causes TMD on a group level (Macfarlane et al, 2009). Psychosocial factors such as stress, anxiety, depression, and

catastrophizing are more common in TMD cases compared to healthy controls (Carlson et al, 1993, Manfredini et al, 2003, De Leeuw et al, 2005, Macfarlane et al, 2009, Quartana et al, 2010, Visscher et al, 2010). In the Orofacial Pain Prospective Evaluation and Risk Assessment study (OPPERA) it was concluded that several psychological factors are associated with the onset of TMD (Fillingim et al, 2013). The OPPERA study has also explored the possible association between genetics and TMD and has found indications that at least two genes are associated with TMD (Smith et al, 2011). Hormonal factors, especially estrogen, have also been discussed since TMD is more prevalent in women than in men (Progiante et al, 2015). Some studies have shown an association between estrogen levels and TMD pain (LeResche et al, 2003), but a systematic review concluded that the evidence for this correlation is weak (Berger et al 2015). Pain comorbidity has been shown to be an important factor in the onset of TMD (Slade et al, 2016.) TMD patients often report other comorbid pain conditions (Plesh et al, 2011) where headache, neck- and back pain are common conditions (Dominick et al, 2012). TMD patients are more likely to have headache compared to healthy controls (Ohrbach et al, 2011) and headache sufferers seem to have an increased risk to develop orofacial pain (Macfarlane et al, 2009). It is well known that some general diseases, e.g. rheumatic disease and some neurological conditions can give dysfunction and pain in the orofacial region (Okeson, 1996).

### *Prevalence and treatment need*

There is a great variation of prevalence figures of TMD in the literature. It has been reported that 40-75% of the adult population have signs and symptoms of TMD (Okeson, 1996). In a meta-analysis it was reported that the prevalence of clinical signs of TMD range from 0–93% and TMD symptoms from 6–93% in different studies (De Kanter et al, 1993). It is probable that these variations are mainly explained by different diagnostic methods used by research groups over the years. TMD pain has been reported to affect approximately 5-12% of the adult population (National Institute of Dental and Craniofacial Research, 2018). The prevalence of TMD increases during adolescence (Nilsson et al, 2005) and often peak in midlife (Progiante et al, 2015). The treatment need has earlier been estimated to be 7-9% in the adult population (Kuttila et al, 1996). However, in a meta-analysis the estimated treatment need was reported to be approximately 16% in the adult population (Al-Jundi et al, 2008).

### *Diagnosis*

TMD classification systems have been developed over the years both in the form of index systems (Helkimo et al 1974, Fricton et al, 1987) and diagnostic systems (Eversole and Machado, 1985, Dworkin and LeResche, 1992). The Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) was developed for research purpose and was presented in 1992 (Dworkin and LeResche, 1992). RDC/TMD consist of a functional (axis

I) and a psychological part (axis II) and the system has been accepted and extensively used, not only in research, but also in clinical situations. The diagnoses of axis I consist of three groups; 1) muscle diagnoses, 2) disc displacements and 3) arthralgia, arthritis and arthrosis. The system provides clear and standardized examination methods which enables comparison of findings from different research groups. The importance of calibration of clinicians have been stressed (Dworkin et al, 1990, Schmitter et al, 2005). A majority of the axis I diagnoses have been shown to have good reliability (Look et al, 2010). In a study concerning the validity of the axis I it was concluded that only the myofascial pain diagnosis had an acceptable validity and therefore a revision of the system was suggested (Truelove et al, 2010). A revision of RDC/TMD was made and resulted in an updated version, the Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) that was published in 2014 (Schiffman et al, 2014). DC/TMD was, unlike RDC/TMD, developed specifically to be able to cover both clinical and research settings.

### *Management*

Treatment of TMD has been described since the ancient civilization of Egypt, where jaw dislocation was treated. Hippocrates also described a manual technique for treating jaw dislocation in the fifth century BC (McNeill, 1997). For more than a century occlusal appliance therapy has been a treatment in cases of e.g. bruxism and TMD (Goodwillie, 1881 and Karolyi, 1901). Costen (1934) put TMD problems on the map when he, in his famous article, claimed that jaw pain and related ear symptoms could be improved by alteration of the occlusion. As mentioned above, occlusion was for a long period of time considered to be the primary cause of TMD (McNeill, 1997) even though criticism occurred already in the late 1950s (Schwartz, 1959). The treatment was often focused on irreversible occlusal equilibration and several gnathological concepts emerged (McCollum, 1927, Stallard 1930, Stuart 1955 and Mann and Pankey, 1963). During the 1960s and 1970s several authors emphasized the role of neurophysiology and physical treatment approaches in the management of TMD and a multifactorial etiology began to be acknowledged (McNeill, 1997). Today, reversible treatments such as occlusal appliances, pharmacological treatment, cognitive behavioral therapies and therapeutic jaw exercises are therefore more common in the management of TMD than irreversible ones (Carlsson and Magnusson, 1999 and Shedden et al, 2013). Occlusal appliances are very common and popular treatments for TMD (Klasser and Greene, 2009) and the stabilization appliance is the appliance which has the best scientific support for efficacy and is therefore often recommended (List and Jensen, 2017). Ekberg et al have in a number of randomized, controlled short- and long-term trials shown that stabilization appliances have a favourable effect on tension-type headache and TMD of both myogenous and arthrogeous origin (Ekberg et al, 1999, 2002, 2004). It has been concluded that there is good evidence that occlusal appliances have a modest treatment effect on TMD pain (Fricton et al, 2010). The mechanisms of action of occlusal appliances have not been fully understood (List and



Jensen, 2017). Mechanisms such as reversible elimination of occlusal interferences, increased vertical dimension and non-specific effects linked to patient-doctor relationship have been discussed (Carlsson and Magnusson, 1999, Michelotti et al, 2012). The positive end result is most likely a result of a combination of different mechanisms (Carlsson and Magnusson, 1999, List et al, 1999). Therapeutic jaw exercises are also a common treatment in the management of TMD. The treatment aims to attain relaxation in tender/sore jaw muscles, strengthen jaw muscles, improve the lower jaw's range of motion and reduce pain (Magnusson and Syrén, 1999). Jaw exercises might also help patients overcome fear of motion in cases of jaw kinesiophobia (List and Jensen, 2017). The mechanisms behind the treatment effect are considered to be a result of proprioceptive neuromuscular facilitation, reciprocal muscle inhibition, increased awareness and stretching (Magnusson and Syrén, 1999). Studies have reported that jaw exercises can be effective in the treatment of TMD myalgia (Magnusson and Syrén, 1999, Michelotti et al, 2004). Due to limited numbers of RCTs and small sample sizes in published studies, there is still not enough evidence to fully support the effectiveness of jaw exercises in the management of TMD (Armijo-Olivo et al, 2016). Jaw exercises will be discussed in detail below. Behavioural treatments such as cognitive behavioural treatment, education, counselling, relaxation, biofeedback, self-treatment regimes and stress management (List and Axelsson, 2010, Story et al, 2016) have been shown to be effective in the treatment of TMD (Aggarwal et al, 2011). Pharmacologic treatment with e.g. NSAIDs, diazepam, glucocorticoids and tricyclic antidepressants, might be effective in the treatment of TMD pain according to some reviews, even though definite conclusions cannot be made (Mujakperuo et al, 2010, List and Axelsson, 2010). Pharmacological management of other chronic pain conditions are well studied and seems to give effective pain relief. It is therefore plausible that these drugs will be effective also for the management of TMD pain (List and Jensen, 2017). Sensory stimulation of afferent nerves for pain relief has been practiced for a long time. In China, archaeologists have found inscriptions in bones and turtle shells from 2100 BC with descriptions of treatment with acupuncture. Acupuncture came to Europe in the 16<sup>th</sup> century and in 1829 the Swedish physician Gustav Landgren presented his thesis on Acupuncture at Uppsala University. Landgren wrote in his summary that acupuncture was effective in e.g. neuralgias, rheumatic diseases, local spasms, toothache and headaches (Carlsson, 1992). Studies have shown that acupuncture is superior to no treatment and as effective as other treatments, e.g. occlusal appliance, in the management of TMD myalgia (Cho and Wang, 2010). In 1965, the Americans Melzack and Wall presented their Gate Control Theory. This theory is based on the assumption that activation of afferent nerve fibers (A $\beta$ -fibers, that convey pressure and touch) inhibits the transmission of pain impulses (mediated by, e.g. C-fibers) in the spinal dorsal horn. Transcutaneous electric nerve stimulation (TENS) uses this mechanism and a number of studies have concluded that TENS has a pain-relieving effect. For example, an RCT showed that TENS has a pain-relieving potential in patient with different types of dental pain (Hansson and Ekblom, 1983). Some studies have also reported that TENS can be effective in treatment of TMD (Wessberg et al, 1981, Møystad et al, 1990, Linde et al, 1995). Still, there is not enough evidence to support TENS in the treatment of TMD pain

(List and Axelsson, 2010). Some authors have reported that occlusal adjustments reduce the incidence of TMD (Kirveskari et al 1998, Kirveskari and Jämsä, 2009). In contrast, several systematic reviews have not been able to show that occlusal adjustments are better than placebo in the treatment of TMD pain (Koh and Robinsson, 2004, Forssell and Kalsö, 2004, List and Axelsson, 2010). Due to the irreversible nature of occlusal adjustments, most researchers are restrictive in the recommendation of this treatment modality in the management of TMD (List and Jensen, 2010). However, occlusal adjustment is an important treatment to achieve occlusal comfort and to alleviate traumatized teeth in patients with e.g. open bite due to earlier degenerative process in the TMJs (The National Board of Health and Welfare, 2011). Surgical treatment with arthroscopy and discectomy has been found to be effective in the treatment of disc displacement without reduction (Rigon et al, 2011, Wänman, 2016). Arthroscopy is suggested to be the first choice of surgical treatment in patients with disabling disc displacement since it is less invasive, requires less time and results in fewer days of sick-leave compared to discectomy (Wänman, 2016). Since conservative non-surgical treatment of TMD is effective in most cases, surgical intervention should only be considered in patients with disabling symptoms from the TMJs where non-surgical treatments, for at least six months, have shown no or only minor effect (Wänman, 2016, List and Jensen, 2017).

### *Evaluating and reporting treatment outcome*

Evaluation of treatment outcome through standardized and well-defined outcome measures is very important to make it possible to compare results from different studies. In order to provide recommendations for core outcome domains in pain research, the Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT) was founded (Dworkin et al 2005, 2008). The following five core domains for evaluation of treatment effect are recommended by IMMPACT; pain intensity, global improvement, emotional functioning, physical functioning and adverse events (Dworkin et al, 2005). Pain is a very subjective feeling (Treede, 2018) and patient self-report is therefore a “golden standard” in measuring pain intensity (Dworkin, 2005). Pain intensity is often measured through the use of different scales. The following scales are frequently used; the Visual Analogue Scale (VAS), the Numerical Rating Scale (NRS), the Faces Pain Rating Scale and the Verbal Rating Scale (Hjermstad et al, 2008, Jensen and Karoly, 2011). In a study evaluating the validity of these scales it was concluded that there is a very strong association between VAS and NRS indicating that they measure the same thing. VAS and NRS also have “more pure” measures of pain intensity with less influence by non-pain intensity factors, e.g. pain unpleasantness, compared to the other scales (Thong et al, 2018). Still, VAS has a number of limitations compared to NRS (Von Korff et al, 2000, Holdgate et al, 2003, Breivik et al, 2008), such as need for abstract thinking, normal vision and adequate motor skills in the respondents. VAS also requires physical equipment e.g. pen/paper or a technical interactive device. Due to these limitations, IMMPACT recommend a 0-10 (i.e. 11-point) NRS scale

for the measurement of pain intensity (Dworkin et al, 2005). A 30% reduction of self-reported pain intensity on NRS (or VAS) is considered to be clinically relevant (Farrar et al, 2001). As mentioned above, TMD has a multifactorial aetiology and therefore participant rating through global improvement, physical functioning and emotional functioning are also important measurements in the evaluation process. Global improvement defined as an experience of overall change of status, following a certain treatment can be measured by the Patient Global Impression of Change (PGIC) (Farrar et al, 2001), which is a scale that has been widely adopted in clinical pain trials (Geisser et al, 2010). PGIC consists of the following alternatives; Pain free, Much improved, Improved, Unchanged, Worse, Much worse, Very much worse. Improvement of the ratings on PGIC has a high association to clinically significant reduction of pain intensity (Farrar et al, 2001, Geisser et al, 2010, Gagnon et al, 2018) and also improvement of other outcomes such as sleep, physical functioning and depression (Geisser et al, 2010). In DC/TMD, the Jaw Functional Limitation Scale (JFLS) has been suggested for the measurement of physical functioning of the masticatory system in TMD patients (Ohrbach et al, 2008). Initially, JFLS was developed as a short global scale with eight questions (JFLS-8) with the purpose to measure the overall functional limitation of the masticatory system. In order to expand the instrument “to also include masticatory limitation, vertical mobility limitation and verbal and non-verbal communication limitation” a 20-item questionnaire (JFLS-20) was developed (Ohrbach and Knibbe, 2018). The full instrument (JFLS-20) contains the following subscales; Mastication, Mobility and Verbal and non-verbal communication. Data are presented either as the mean of the separate subscale scores or as a global score which can be obtained by calculating the mean of the three subscale scores. When interpreting the data, it is important to remember that norms have not been established for JFLS-20 (Ohrbach and Knibbe, 2018). In a systematic review (De La Torre Canales et al, 2018) it was concluded that moderate to severe somatization and depression are highly prevalent in TMD patients. Therefore, evaluation of emotional functioning as a treatment outcome is very important. Anxiety often precedes depression and it is therefore important to evaluate both these conditions (Stern, 2014). The Hospital Anxiety and Depression Scale (HADS) was developed in the early 1980s as an instrument for the measurement of anxiety and depression (Zigmond and Snaith, 1983). Due to its simplicity and ease of use it has been widely adopted in both clinical and research settings (Stern, 2014). The scale consists of fourteen questions, seven for anxiety and seven for depression and it takes approximately five minutes to complete the form. It is vital that the scores for anxiety and depression are handled separately. Scores of 0-7 represent non-cases, 8-10 doubtful cases and 11-21 definite cases (Zigmond and Snaith, 1983). The scale has been validated in several languages and in different settings (Herrmann, 1997, Bjelland et al, 2002, Snaith 2003). In treatment evaluation, both in clinical and research settings, it is important to register and report adverse events or harms of treatment to secure the quality of the treatment and also to provide transparency for critical evaluation of studies (Ioannidis et al, 2004). Without thorough and transparent reporting in a clinical study, the reader will have great difficulties in judging the reliability and validity of the trial findings and thereby the

risk of biased estimates of treatment effects is increased (Moher et al, 2012). The Consolidated Standards of Reporting Trials (CONSORT) is a set of guidelines created by a group of scientists and editors in order to remedy the general lack of quality in reporting data of randomized controlled trials (RCTs). The majority of the scientific community have endorsed the CONSORT statement (Moher et al, 2012).

## *Physiotherapy*

The Royal Gymnastic Central Institute (today the Swedish School of Sport and Health Science) was founded in 1813 by Per Henrik Ling (1776-1839) for the education of gymnastic instructors (Peterson, 2011). Ling (1834) developed a system for medical gymnastics to promote better health, which was the foundation of the first education for physiotherapists in the world (Broberg and Tyni-Lenné, 2009, Peterson, 2011). Since then the field has grown and today the Swedish Association of Physiotherapists issues specialist qualifications in 16 different areas (Fysioterapeuterna, 2014). In physiotherapy the key concepts are the human body, movement, function and interaction in relation to health from a biopsychosocial perspective (Broberg and Tyni-Lenné, 2009). The biopsychosocial model was presented in the late 1970s (Engel, 1977) and describes how biological, psychological and social factors interact in the development of illness and disease (Gustavsson, 2011).

### *Physiotherapy in the treatment of pain*

Exercises have been shown to give short term decrease of pain perception in both chronic pain patients and healthy subjects (Naugle et al, 2012, Nijs et al 2012). This hypoalgesia can probably be explained by activation of the endogenous opioid system and descending inhibitory pathways in the central nervous system (Nijs et al 2012). Less is known concerning the effect of exercise on long term central regulated pain inhibitory control. Still, a reduction of pain sensitivity has been reported in healthy individuals after frequent aerobic exercise (Naugle and Riley, 2014, Sajedi and Bas, 2016). Several reviews have concluded that exercise reduces pain severity in chronic pain patients and improve physical function (Geneen et al, 2017). Exercise has also been shown to have a positive influence on depression and anxiety (Dinas et al, 2011, Asmundson et al, 2013). However, a recent review concluded that even though some studies show that exercise have a positive effect on psychological function, other studies show no effect (Geneen et al, 2017) and therefore these results must be considered to be inconclusive (Karlsson, 2017). Exercise combined with education is more effective in reducing pain intensity and functional impairment than single treatment modalities (Hurwitz et al, 2008). The positive treatment effect in interventions that combine body and mind is probably in part mediated by an increase in patient self-efficacy (Hadhazy et al, 2000). The concept of self-efficacy was developed in the late 1970s (Bandura, 1977) and it is defined as a person's belief in one's own ability to

accomplish something, for example a treatment regime, in a successful way. In general, people are often afraid of failing and therefore only try things they believe they can accomplish (Bandura, 1997). Self-efficacy is an important part of self-management strategies and aims to make the patient an active part of the treatment modality (Lorig and Holman, 2003, Smith and Elliott, 2005). Almost all chronic pain patients adopt some kind of self-management. The process of active self-management in chronic pain patients has been associated with decrease in pain related disability, medication, use of medical services and also improvement in general health (Smith and Elliott, 2005). The importance of self-management has also been stressed in the treatment of TMD. Therapeutic jaw exercises are an important part of these self-management strategies (Durham et al, 2016).

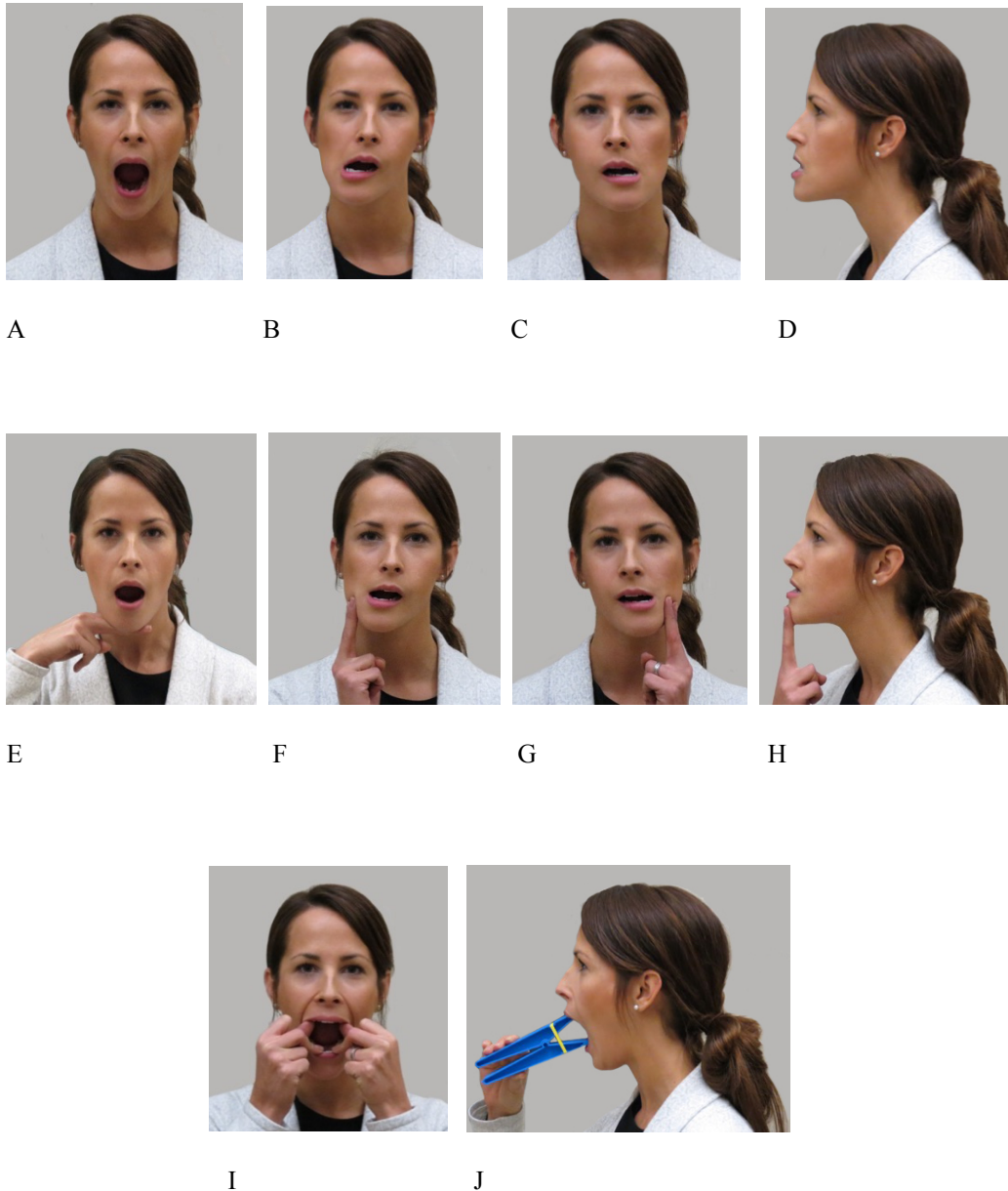
### *Therapeutic jaw exercises*

As mentioned above, reversible conservative treatments are often preferred in the management of TMD. Since jaw exercises are simple to do and has a low cost compared to other treatments, it is often regarded as a first line of therapy in combination with education, in selected TMD patients (Michelotti et al, 2005, Clark, 2008). Several studies, but also clinical experience, imply that individually prescribed jaw exercises are promising and may be effective in the management of TMD (Michelotti et al, 2005, Medlicott and Harris, 2006). In a study investigating the perceived effectiveness of jaw exercises among specialists in oral and maxillofacial surgery it was shown that 79% considered the treatment to be effective and 69% of the specialists prescribed jaw exercises to their TMD patients (Rashid et al, 2012). A jaw exercise program can be executed in a number of different ways. It often contains relaxation exercises, free movements of the mandible, movements of the mandible with a small resistance and finally stretching of the jaw muscles, Fig 1 (Carlsson and Magnusson, 1999). Jaw exercises have been suggested in a number of different TMD conditions.

### *Myalgia of the masticatory muscles*

Several studies have reported that different kinds of jaw exercises seem to be effective in reducing pain due to TMD myalgia (Burgess et al, 1988, Magnusson and Syrén, 1999, Carlson et al, 2001, Grace et al, 2002, Nicolakis et al, 2002, De Laat et al, 2003, Michelotti et al, 2004, Gavish et al 2006, De Felício et al, 2010, Kalamir et al, 2013, Tuncer et al, 2013, Kraaijenga et al, 2014, Häggman-Henrikson et al 2018). Nevertheless, there are studies that have not been able to show that jaw exercises are more effective in reducing TMD myalgia compared to control groups that only received education (Craane et al, 2012) and simple self-care (Mulet et al, 2007).

Fig. 1. Jaw exercise programme: 1. *Free movements of the lower jaw*. Maximal jaw opening (A), laterotrusion (B-C) and protrusion (D) without resistance. 2. *Movement of the lower jaw with a small resistance*, for example with a couple of fingers. Jaw opening (E), laterotrusion (F-G) and protrusion (H) toward a small resistance. 3. *Stretching* with fingers (I) and a jawtrainer (J).



Jaw exercises have also been studied in combination with other treatment modalities. In a study comparing jaw exercises alone or in combination with an occlusal appliance in the treatment of myofascial pain according to RDC/TMD, it was shown that the occlusal appliance did not provide an additional benefit compared to jaw exercises as a standalone treatment. All groups showed reduction of TMD pain over time (Truelove et al, 2006). In another study, jaw exercises in combination with psychological intervention was shown to be more effective in reducing pain intensity among patients with TMD pain compared to jaw exercises alone (Makino et al, 2013).

### *Arthralgia of the TMJs*

In an RCT concerning patients with “articular TMD” according to RDC/TMD it was shown that jaw exercises were significantly more effective in reducing TMJ pain compared to a waiting-list control group (De Felicio et al, 2008). In an uncontrolled study it was concluded that jaw exercises significantly reduced the pain intensity in patients with disc displacement with reduction and jaw pain (Nicolakis et al, 2000). It has also been shown that jaw exercises reduce jaw pain and disability significantly in patients with radiologically proven osteoarthritis and pain (Nicolakis et al, 2001). These positive results remained stable over time and a majority of the successfully treated patients stated, at long-term follow-up, that they had not needed any additional treatment for their original complaints (Nicolakis et al, 2002).

### *Arthritis of the TMJs*

In an RCT investigating the effect of jaw exercises in patients with rheumatoid arthritis and ankylosing spondylitis it was shown that the treatment increased the maximum mouth opening capacity and reduced clinical dysfunction according to Helkimos index (Tegelberg and Kopp, 1988). At long-term follow-up after three years, a majority of the patients reported a stable treatment result and a decrease of severity of signs and symptoms from the masticatory system compared to baseline (Tegelberg and Kopp, 1996). Jaw exercises should not be introduced in patients with acute arthritis. In those cases, the first treatment of choice is pharmacological treatment (Häggman-Henrikson et al, 2017).

### *Disc displacement with reduction*

Clicking sound from the TMJs due to disc displacement is very common (Wänman, 1987, Könönen and Nyström, 1993, Magnusson et al, 1994, Onizawa and Yoshida, 1996) with a prevalence of 15-16% and an incidence of 4% (Marklund and Wänman, 2007). Disc displacement with reduction seldom develops into more serious conditions such as TMJ locking (Lundh et al, 1987, Sato et al, 2003). In some studies jaw exercises have been shown to effectively eliminate or decrease the frequency of clicking (Au and Klineberg, 1993, Yoda et al, 2003). When compared to a soft repositioning appliance, jaw exercises were reported to significantly decrease jaw pain and increase mouth opening capacity in patients with anterior disc displacement in the TMJs (Carmeli et al, 2001).

### *Disc displacement without reduction (locking)*

Patients with anterior disc displacement without reduction (ADDWoR) often experience limitation of jaw movements and pain from the affected TMJ, especially during function (Al-

Baghdadi et al, 2014). In an uncontrolled short-term study, jaw exercises were shown to be effective in improving mandibular movement and reducing jaw pain in patients with ADDWoR (Sakuma et al, 2017). In another study, patients with ADDWoR served as their own waiting-list controls before receiving treatment and in this study jaw exercises resulted in a significant increase of mouth opening and a reduction of jaw pain compared to the waiting list control period (Nicolakis et al, 2001). The combination of NSAIDs and jaw exercises has been shown to significantly improve mouth opening capacity compared to a no treatment control group in patients with ADDWoR (Yuasa and Kurita, 2001). In an RCT comparing the treatment effect of NSAIDs in combination with either jaw exercises or a maxillary stabilization appliance in patients with ADDWoR, there were significant improvements in maximum mouth opening and pain intensity in both treatment groups. However, the results of the RCT suggest that jaw exercises lead to a faster improvement of jaw function compared to occlusal appliance therapy (Haketa et al, 2010). Not all studies show favorable effects of jaw exercises. In a study comparing physical therapy and information in an RCT setting, it was shown that pain intensity decreased and jaw function increased in both groups. Physical therapy did not have any significant additional effect compared to information (Craane et al, 2012). In an RCT comparing information (no treatment) to different non-surgical treatments of ADDWoR, including NSAIDs, jaw exercises, occlusal appliance therapy and jaw mobilization, it was concluded that there were no significant differences between the treatment groups and the no treatment group in treatment outcome. All groups improved in clinical signs and symptoms over time (Minakuchi et al, 2001). When comparing surgical and reversible treatments, several studies conclude that the first treatment of choice in patients with ADDWoR should be reversible and the least invasive treatment, for example pharmacological treatment and jaw exercises (Schiffman et al, 2007, Al-Baghdadi et al, 2014).

### *Limitation of jaw movement*

A decrease in maximum mouth opening is a common sign in TMD of both muscular and TMJ origin (Okeson, 1996). It has been shown that a “passive jaw motion device” (Therabite®) can increase mouth opening and decrease pain levels in treatment resistant patients with TMD of both muscular and TMJ origin according to RDC/TMD (Maloney et al, 2002). A common side effect of TMJ surgery is limitation of mouth opening. In a randomized controlled study evaluating patients after TMJ surgery, it was shown that jaw exercises increased maximum mouth opening significantly compared to a no treatment control group, during a period of eight weeks (Austin and Shupe, 1993). On the basis of these results the authors conclude that jaw exercises are very important in TMJ post-surgical rehabilitation. In radiated head and neck cancer patients, trismus and reduced mouth opening capacity are common features (Pauli et al, 2016). It has been shown that jaw-mobilizing devices are effective in increasing maximum mouth opening and reducing trismus associated symptoms in head and neck cancer patients in both short and long term (Buchbinder et al, 1993, Pauli et



al, 2016). In a systematic review it was concluded that jaw exercises and jaw-mobilizing devices are better than no treatment in increasing maximum mouth opening in radiated head and neck cancer patients (Scherpenhuizen et al, 2015).

### *TMD associated with whiplash associated disorders*

Several studies have shown a close functional association between the neck and the masticatory system (Eriksson et al, 2000, Kohno et al, 2001) and it has been concluded that whiplash associated disorders (WAD) are more prevalent in TMD patients compared to healthy controls (Häggman-Henrikson et al, 2014). A large proportion of patients with WAD develop TMD pain (Sale and Isberg, 2007) which results in an increased prevalence of TMD pain in WAD populations (Häggman-Henrikson et al, 2013). Patients with WAD who develop TMD often have more severe jaw pain and dysfunction and a poorer treatment prognosis compared to TMD patients without WAD (Häggman-Henrikson et al, 2014). In an RCT, jaw exercises were shown to have no additional effect on TMD symptoms in WAD patients compared to a regular rehabilitation program (Klobas et al, 2006). In a systematic review, it was also concluded that common treatments in TMD cases, such as jaw exercises, occlusal appliances and counseling have limited effect in patients with a combination of TMD and WAD (Häggman-Henrikson et al, 2013).

### *Summary*

Even though both clinical experience and some research evidence suggest that jaw exercises might be effective in the treatment of TMD (Michelotti et al, 2005, List and Axelsson, 2010) there is still a great uncertainty concerning this issue (Armijo-Olivo et al, 2016). A number of systematic reviews and meta-analyses have concluded that there is not enough evidence to definitely support the effectiveness of jaw exercises in the management of TMD (Moraes et al, 2013, Armijo-Olivo et al, 2016, Paço et al, 2016). The main reason for this uncertainty is the lack of RCTs with sufficient rigid treatment protocols and number of patients to produce enough power to make definite conclusions (Moraes et al, 2013, Armijo-Olivo et al, 2016, Paço et al, 2016). The lack of evidence concerning jaw exercises does not mean that the treatment is ineffective. Considering that jaw exercises is potentially effective, safe, reversible and easy to use, it can still be recommended in the management of TMD (Armijo-Olivo et al, 2016).

### *Research traditions*

Quantitative research methods are the most commonly used methods in pain research. These methods aim to objectively collect data that are broken down to units and numbers in order to analyse data statistically and to generalize the findings. The experience of pain is, as

mentioned above, one of the most subjective and complex feeling there is, and chronic masticatory myofascial pain has components of both a sensory, affective and cognitive nature (Turk et al, 2003, Dworkin et al, 2005). It is difficult to investigate subjective phenomena such as emotions and experiences with a quantitative method. A qualitative research method that takes emotional, psychological, social and existential aspects into account is more suitable to investigate these phenomena (Britten et al, 1995, Malterud, 2001). Qualitative research tradition manages many different methods with its own distinctive character and the researcher should choose a method that gives the data optimal relevance and validity (Creswell, 2012). Systematic text condensation (STC) is a qualitative method that was developed as a systematic thematic cross-case analysis, which according to the originator Malterud (2012) is an easier way of qualitative analysis compared to a, for example, phenomenological approach. Rather than exploring the possible underlying meaning of what is said, the experiences of the patients as expressed by themselves are presented. STC, as well as most other qualitative methods, has the theoretical foundation of social conservatism (Malterud, 2012). In social conservatism, individuals develop subjective meanings of their experiences of certain objects or phenomenon (Creswell, 2012). The dynamic interpretation of multiple and varied versions of reality results in knowledge. Although obviously inspired by phenomenology, Malterud (2012) does not consider STC to be a phenomenological method and there is not a clearly stated phenomenological philosophy in STC. The method is considered to be more of a procedure rather than a specific theoretically devoted method. This means that many different theories, depending on the research question, can be used to support STC analysis (Malterud, 2012). In STC there is a risk that the individual context of data gets lost in the coding process where data is decontextualized. Information is always lost in a qualitative study design when data is reduced as part of the analyzing process. In order to reduce this risk of fragmentation, the STC process includes recontextualization where the researchers' interpretations are validated against the original transcripts (Malterud, 2012). Results from a qualitative study cannot be generalized, but study design and the selected population can often allow the results to be transferable to similar contexts. A common way to generalize data from a qualitative study is to do a quantitative follow-up study based on the qualitative results.

In the absence of high-quality evidence, the caregiver must turn to experience or, even better, the assembled experience, consensus of colleagues. The most commonly used consensus methods in health care are the nominal group technique, the conference consensus technique and the Delphi method (Murphy et al, 1998). The Delphi method was named after the famous Oracle in the ancient Greek city of Delphi. The Oracle was thought to deliver the God Apollo's knowledge through prophecies and advice. Policy makers in ancient Greece used the Oracles services in order to make decision, for example whether to go to war or not (Marchais-Roubelat and Roubelat, 2011). The Delphi method was developed as an experiment in the early 1950s at the RAND corporation in California, a think tank partly funded by the US military. The experiment was designed to apply expert opinions on a possible nuclear attack. The experiment was concealed to the public during a decade and was

first published in 1962 (Dalkey and Helmer, 1962). Since then the Delphi method has been used in a variety of areas, including dentistry and the field of TMD (Cramer et al, 2008, Durham et al, 2016). In current times, the Delphi method is a technique which strives for consensus of opinions in a group of experts, through a series of questionnaires in different “rounds”. In the classical Delphi method, the initial questionnaire consists of open-ended questions and collects opinions which are then analysed with a qualitative research method. The results from the first questionnaire are then returned to the experts in a second quantitative questionnaire (second round), where they can grade a set of statements with a Likert scale. Subsequently, the experts receive controlled feedback with the results, where they can see their own answers in relation to the other experts in the panel. The process is repeated in several rounds until consensus is achieved or a decrease in number of returned questionnaires is seen. Even though the key features of the method are intact, several modified Delphi techniques have been suggested (Hasson et al, 2000). One strength of the Delphi method, compared to other consensus techniques, is that the experts are anonymous to each another, which removes the possible social influence on opinions in a face to face setting (Bolger and Wright, 2011). Other advantages over the other consensus techniques are that it is easy to get a wide geographical dispersion of the experts and that the method is relatively cheap.

# AIMS

## *General aim*

The general aim of this thesis was to gain a better understanding concerning the different aspects of jaw exercises in the treatment of masticatory myofascial pain. Special emphasis was directed toward efficacy, cost-effectiveness, patients' views and dentists' experiences.

## *Specific aims*

- To investigate the self-perceived level of knowledge, attitudes and clinical experience in the treatment of children, adolescents and adults with TMD among GPDs and also to evaluate if these factors have changed over time (*study I*).
- To investigate patients' experiences of therapeutic jaw exercises in the treatment of masticatory myofascial pain (*study II and III*).
- To investigate if there is an international consensus among TMD/orofacial pain specialists regarding indications, performance, follow-ups and effectiveness of therapeutic jaw exercises in the treatment of TMD (*study IV*).
- To study the efficacy and cost-effectiveness of therapeutic jaw exercises in patients with masticatory myofascial pain in comparison to treatment with stabilization appliance and waiting-list controls (*study V*).

## MATERIAL AND METHODS

### *Study I*

In an earlier postal questionnaire study concerning GPDs attitudes and self-reported experience and knowledge in the field of TMD (Tegelberg et al, 2001) it was concluded that a majority of GPDs in Sweden lacked routines in diagnostics, choosing therapy and evaluating treatment results in children and adolescents with TMD. A web-based questionnaire was constructed based on this postal questionnaire. The web-based questionnaire consisted of 20 multiple-choice questions in the following categories: *Demographic information* – gender; number of years in profession. *Quality assurance* – presence of health declaration containing questions on the topic of orofacial pain and headache; regular case history of orofacial pain and headache; participation in post graduate TMD education. *Clinical experience and treatment* – self assessment of the GPDs own skills in diagnostics, therapy decision, various treatments and evaluation of treatment. The questions were answered using the following scale: 1 = lack of routine/unable, 2 = limited routine/unsure and 3 = good routine/confident. *Need for specialist resources in the field of TMD* – need for consultation visits in their own clinic; need for telephone consultations; need for the possibility to refer patients to an orofacial pain/TMD specialist; need for the possibility to auscultate at a specialist clinic; need for post graduate education. *Attitude* - The dentists were asked to finish each of the two sentences "To treat *adults* with TMD pain is ..." and "To treat *children/adolescents* with TMD pain is ..." with two of the ten following adjectives : interesting, educational, rewarding, worthwhile, challenging, stressful, difficult, frustrating, unpleasant and demanding. The first five adjectives were judged to be positive and the last five to be negative. If both selected adjectives were positive, the attitude was judged to be positive. If one adjective was positive and the other one was negative, the attitude was judged to be neutral and, consequently, two negative adjectives were judged as a negative attitude. The web-based questionnaire was sent to all GPDs in the Public Dental Health service in the County of Uppsala, Sweden (n=128) in September 2010. GPDs who did not answer the questionnaire received an e-mail reminder after two weeks. A maximum of three reminders were sent. In February 2014 a second web-based follow-up questionnaire was sent to all GPDs in the in the same county (n=113). Dentists who did not answer received an e-mail reminder according to the routines previously described. The follow-up questionnaire comprised eight of the original 20 multiple-choice questions in these categories: *Demographic information* – gender. *Quality assurance* – regular case history of orofacial pain and headache; participation in post graduate TMD education. *Clinical experience and treatment* – self assessment of the dentist's own ability in diagnostics, therapy decision, various treatments and evaluation of treatment. *Attitude* - The dentists were asked to finish each of these two sentences: "To treat *adults* with TMD pain is ..." and "To treat *children/adolescents* with TMD pain is ..." with two out of ten adjectives (see above). During the time period between the two questionnaires the Department of Stomatognathic

Physiology offered the GPDs, in the Public Dental Health service in Uppsala, five seminars, seven short courses and two major courses in TMD. These educations were part of the strategic educational program at the Public Dental Health service in the County of Uppsala. In January 2011 an optional examination template was introduced in the computer case files containing the following two questions; “Do you have pain in your temples, face, temporomandibular joints, or jaws once per week or more often?” and ”Do you have pain when you open your mouth wide or when you chew once per week or more often?” (Nilsson et al, 2006). The results from the present study (2010 and 2014) were also compared with the results from the previous study from 2001 (Tegelberg et al, 2001) in order to analyse if there were any major changes over a longer period of time.

### *Statistics*

The results are presented as frequencies and mean values (see next section below). For the statistical analyses of differences between variables and groups, Chi-square test was used (SigmaPlot). A p-value < 0.05 was considered as a statistically significant difference.

### *Ethical aspects*

After correspondence with the Regional Ethical Review Board in Uppsala, it was concluded that this study did not need an ethical vetting.

### *Methodological considerations*

The advantage of the web-based questionnaire study is that it is based on an earlier postal questionnaire from 2001 (Tegelberg et al, 2001) and that it has a cross sectional follow-up design. In this way, comparisons could be made over a long period of time (2001 to 2014). The fact that the earlier study (Tegelberg et al, 2001) was conducted in other counties, with other possible treatment traditions, should be taken under consideration when the results are interpreted. A disadvantage of the study, as already mentioned by Tegelberg et al (2001), is concerning the questions’ validity and reliability which has not been investigated. A weakness of the follow-up part of the material is that it is not known if the responders in the two questionnaires (2010 and 2014) were the same. We can therefore not treat the results as longitudinal prospective data. However, a relative low turnover on GPDs in the Public Dental Health service in the County of Uppsala and the high response rate to both questionnaires lend strength to the results and allow for comparison between the questionnaires.

### *Study II*

The experience of chronic masticatory myofascial pain is a very subjective feeling with components of a sensory, affective and cognitive nature (Dworkin et al, 2005, Turk et al,

2003). A qualitative research method, such as STC, that takes emotional, psychological, social and existential aspects into account is suitable to investigate these experiences (Britten et al, 1995, Malterud, 2001). Data were collected through semi-structured interviews. The interviewer was a dental hygienist and a researcher with experiences in both qualitative methods and interview technique. The interviewer had no connection with the patients' treatments and had very little experience and knowledge about the treatment studied. Prior to the interviews a TMD specialist therefore gave the interviewer a short (1.5 hours) education in TMD with special focus on therapeutic jaw exercises. The TMD specialist gave the patients careful verbal and written information about the study before the patients accepted to join. The interviewer also gave the informants verbal information about the study before the interviews. The voluntary participation was repeatedly emphasized. The information was given in this manner to foster trust between the researchers and the informants.

### *Study population*

The goal was to select a strategic sample of patients in order to get a variation in the study population concerning age and gender. The following inclusion and exclusion criteria were used: *Inclusion criteria:* 1) Myofascial pain with or without limited mouth opening according to the Research Diagnostic Criteria for TMD (RDC/TMD) axis I (Dworkin and LeResche, 1992). 2) Pain for a minimum of six months prior to performing therapeutic jaw exercises. 3) Age  $\geq$  18 years. 4) The patient should have performed therapeutic jaw exercises for at least three months and not longer than 12 months. *Exclusion criteria:* 1) Osteoarthritis, osteoarthrosis and disc displacement without reduction, in the TMJ, according to the RDC/TMD axis I (Dworkin and LeResche, 1992). 2) Dental pain. 3) Neuropathic pain. 4) Rheumatic disease or general inflammatory condition. 5) General myopathy, for example fibromyalgia. 6) Whiplash diagnosis. 7) Language difficulties. 8) Other treatment modalities than jaw exercises received at the specialist clinic. The number of participants was not decided on beforehand. Those patients who met the inclusion but not the exclusion criteria were asked to participate in the study. No patient declined to be a part of the study. Ten patients, one man and nine women, with a mean age of 35 years (range: 20-58 years) were enrolled. The patients had been referred to the Department of Stomatognathic Physiology, Public Dental Health Service, Uppsala and had only received treatment with therapeutic jaw exercises at the Department. The exercise program had been individually designed for each patient, according to clinical routine, and included jaw relaxation exercises, free movements of the mandible, movements of the mandible with a small resistance and stretching of the jaw muscles. All patients had been recommended to do the jaw exercises three times per day and were evaluated according to clinical routine after two and eight weeks. Six of the patients were completed and four were still under treatment when the interviews were conducted between April and September in 2014.

### *Semi-structured interviews*

An interview guide with 10 domains was constructed (Table 1) and tested in a test interview. The test interview was conducted in the same manner as the rest of the interviews and the interview guide was considered to be sufficient. The semi-structured interviews, which lasted 20-35 minutes, focused on the patient's experience of therapeutic jaw exercises as a treatment of masticatory myofascial pain. Both the open-ended and the follow up questions aimed to encourage the patient to reflect and freely comment on the different themes. Data collection was continued until no further relevant information emerged. The interviewer and one of the researchers listened to the interviews separately and decided together when no further data emerged. The interviews were then transcribed verbatim from spoken to written language. The interviews were conducted in a nonclinical environment at the department. The interviewer and the researchers involved in the analysis of data took care to bracketing their personal notions and expectations to reduce the risk of affecting the interviews and analysis, respectively. This was partially accomplished by writing down the researchers' expectations before the interviews and text analysis.

Table 1. Interview guide

| <b>Domain</b> | <b>Description</b>  |
|---------------|---|
| 1             | Experiences about the orofacial pain.   |
| 2             | Description and reflections on out-patient care until the patient received treatment at the unit for Stomathognatic Physiology. |
| 3             | Initial reflections on therapeutic jaw exercises.   |
| 4             | Expectations on the treatment.  |
| 5             | Experiences on the practical issues of treatment.   |
| 6             | The pros and cons of therapeutic jaw exercises.   |
| 7             | Patient adherence.  |
| 8             | Difficulties during treatment.  |
| 9             | Treatment effect.   |
| 10            | Reflections after treatment.  |

### *Text Analysis*

Two researchers were involved in the analysis of the text material. One of these is a TMD specialist and also the patients' former caregiver. The other researcher who was involved in the analysis is a dentist and professor in Cariology, with a long experience of qualitative research methods. STC according to Malterud (2012) was used in order to arrange and analyse the text material. In the *first phase*, reading all transcriptions from cover to cover attained a general impression of the whole and an overview of the data. Listening to the interviews and at the same time reading the transcripts were useful in quality testing of the transcribed texts. Small alterations of the texts were made. Preliminary themes of the informant's experiences were established and in this process the researchers carefully tried to bracket their preconceptions. In the *second phase*, meaning units were identified, coded and sorted under the previous preliminary themes. Coding involve decontextualization, where the



text is temporarily removed from its context for a cross case synthesis. Some of the preliminary themes were refined and sub-categorisations emerged. Recurrently the tacit rules used to code and sort the meaning units were identified and questioned. A research triangulation was carried out to test the themes legibility. The two researchers involved in the analysis and the interviewer separately sorted a number of randomly selected quotes under the different themes. Complete versions of the transcripts were kept in order to recontextualize the findings in the end of the analysis. In the *third phase* the meaning units in each subgroup were condensed to an artificial quotation, which summarized the different data in the subgroup. Some authentic quotations were selected to support the condensates. The names of the themes and subgroups were further adjusted as a result of evolving understanding. In the *fourth phase*, the content of each condensate was synthesized. Some authentic quotations were selected to support these descriptions. The validity of the synthesized results was checked by recontextualization of the results in relation to the original text. After the analysis, the quotes in Swedish were translated into English by a translator outside the research group and then translated back in order to ensure that no vital information was lost. Definite titles of the subgroups were chosen.

#### *Ethical aspects*

A written informed consent was obtained from all informants. No private data that would jeopardize their anonymity were published. In an interview situation there is always a risk of encroachment of the informant's integrity. It was clearly emphasized that the informants did not have to answer any questions that made them feel uneasy. During an interview, information can emerge that might need further management. A psychologist that could offer treatment, if required, was therefore attached to the project. Ethical approval was obtained from the Regional Ethical Review Board in Uppsala (2014/001).

#### *Methodological considerations*

According to Lincoln and Guba (1985) *trustworthiness* can be achieved in a qualitative research study through *credibility*, *dependability*, *transferability* and *confirmability*. In some extent *credibility* in qualitative research corresponds to internal validity in quantitative research. The criteria deal with whether the chosen methods produce credible and truthful findings in the data collection and interpretations in the analysis (Hamberg et al, 1994). A doctor-patient relationship might influence an interview, making the informant more prone to withhold information that they think would endanger further treatment. This threat to credibility was reduced in this study since the interviewer had no relations to the informants. Credibility was also strengthened by the use of an interview guide (Table 1), which ensured that the same domains were covered in all interviews. Preconceptions and personal notions can further influence and direct an interview. The fact that the interviewer had limited

experience and knowledge about the treatment studied reduced this threat and kept the interviews open minded. During the interviews follow-up questions aimed to give the informants an opportunity to further deepen their answers so that the true opinion of the informants emerged and could be apprehended by the interviewer. The transcription process, where the interviews are transcribed from spoken to written language, always pose a potential threat to credibility since transcripts are “impoverished, decontextualized reproductions of the living conversation” (Kvale, and Brinkmann, 2009). To reduce this risk, the corresponding author listened to the spoken interviews and read the transcripts simultaneously. Another threat to credibility in this study is the translation of the quotes from Swedish to English. To minimize this problem a translator outside the research group was engaged. The quotations were translated to English and then translated back to Swedish, only to verify that no vital information was lost in this process. In the analysis the researchers made an effort to stay close to the data and continuously making critical reflections regarding the findings of themes. This strengthens the credibility further. In qualitative research, complex connections are often studied in surroundings that is always changing. A criterion for “solidly performed research is *dependability*” (Hamberg et al, 1994). This means that a research project must adapt to new inputs and changes in the studied environment during the time period of the study (Hamberg et al, 1994). In *study II*, the number of participants was not decided on beforehand. The data collection was continued until no further relevant information emerged. This can be seen as an expression of dependability. The interview guide could have limited the dependability, but through the interviewer’s concern to use open-ended and follow-up questions the informant was encouraged to express his/her own perspectives and the interviewer could explore unexpected areas and adapt to these new inputs. The research process was also carefully described step by step so that it would be easy for others to follow. Results and conclusions from a qualitative study can never be generalized to a population level, but the results can in some instances be transferred to a different context. Thorough description of the research process, the study population and the context in which the study took place makes it possible for the reader to decide the degree of *transferability* (Graneheim and Lundman, 2004). Quantitative research strive for objectivity. The corresponding criterium in qualitative research is called *confirmability* and means that the study should include procedures that show that the findings originate from the data and are not fabrications due to poor analytic work or preconceived assumptions (Hamberg et al, 1994). Each finding in the present study was highlighted by actual quotes of the informants. Another way to enhance confirmability is to let several researchers independently review the interviews and then discuss the findings until consensus regarding themes is reached. The two researchers who were involved in the analysis had several meetings where the themes, sub categories and results were discussed. The researchers took care to bracketing their personal notions and expectations to reduce the risk of affecting the analysis. This was partially accomplished by frequently returning to the researchers written preconceived assumptions that had been made before the analysis. A research triangulation between three of the researchers concerning the different patterns and themes that emerged was also made to strengthen confirmability. The recontextualisation process in STC strengthens the confirmability further.

### *Study III*

As mentioned earlier, results from a qualitative study, such as *study II*, cannot be generalized, even if the study design and the selected population allows the results to be transferred to similar contexts. To be able to generalize data, a quantitative research approach is necessary. A postal questionnaire was constructed based on the findings in *study II* and then sent to 150 consecutive patients with masticatory myofascial pain who had received treatment with jaw exercises at a TMD specialist clinic during three to 12 months. The treatment with jaw exercises included both verbal and written information about the treatment as well as follow-ups concerning adherence and evaluations of treatment effect (Fig 1.). The patients could also have received other treatments such as interocclusal appliance therapy. The treatments were performed between April 2015 and November 2016. The *inclusion* and *exclusion criteria* were the same as in *study II* with the exception that participants that had received other treatment modalities than jaw exercises at the specialist clinic also were accepted. The postal questionnaire contained 24 statements concerning therapeutic jaw exercises in the categories *patient adherence, symptoms, treatment effect and participation* (Table 2). The participants were asked to answer each statement according to a five-item verbal Lickert scale that ranged from “Strongly agree” to “Strongly disagree”. A maximum of two reminders were sent to non-responders. The questionnaire, including possible reminders, were sent to the patients between April 2016 and March 2017. If 65% or more of the responders agreed or disagreed with a statement it was concluded that a *majority* of patients shared that opinion.

### *Statistics*

Data are generally presented as number (n) and frequencies (%), but for age, median (range) is reported. Differences between responders and non-responders in age and sex distribution were analysed with Mann-Whitney U-test and chi-two test, respectively.

### *Ethical aspects*

All patients received written information about the study and it was carefully emphasized that participation was voluntary. An answered questionnaire was considered to be a written informed consent. The questionnaire did not include any personal data and individual patients could therefore not be identified. No private data that would jeopardize the anonymity of the patients are published. It was clearly emphasized that the patients did not have to answer any questions that made them feel uneasy. Ethical approval was obtained from the Regional Ethical Review Board in Uppsala (2015/517).

Table 2. Statements in the postal questionnaire.

| Statements   |
|--|
| 1. It was difficult to add jaw exercises as a daily routine.   |
| 2. It was easier to remember the jaw exercises when I did it in connection with an already established routine (for example tooth brushing). |
| 3. The hardest was to remember/find the time to do the jaw exercises in the middle of the day.   |
| 4. The information concerning the jaw exercises was simple and structured.   |
| 5. The information and the instructions at follow-ups were good and useful.  |
| 6. I would have preferred a written information about the underlying cause of my symptoms.   |
| 7. I would rather not do jaw exercises among other people.   |
| 8. It is important that the jaw exercises does not feel like a burden or causes stress.  |
| 9. I had relatively severe jaw pain which motivated me to do the jaw exercises.  |
| 10. The jaw exercises were effective and reduced my symptoms.  |
| 11. The treatment effect motivated me to continue the training.  |
| 12. When my jaw symptoms were reduced, I did the jaw exercises less frequently.  |
| 13. In the beginning, I was skeptical/doubtful to the effectiveness of the jaw exercises.  |
| 14. The jaw exercises felt so "simple" and I had expected a more advanced treatment.   |
| 15. Considering the effectiveness of the jaw exercises, I was pleased not to have to go through with more advanced treatment.                |
| 16. The treatment effect of the jaw exercises came relatively fast.  |
| 17. It took a long time before I felt any effect of the jaw exercises.   |
| 18. My mouth opening capacity increased when I did the jaw exercises.  |
| 19. The treatment effect on my mouth opening capacity came more quickly than the effect on my jaw pain.                                      |
| 20. It feels good to be able to do the jaw exercises without any tools.  |
| 21. Before I got to know what caused my jaw pain, I was afraid that the pain was a symptom of a more serious disease.                        |
| 22. I understand that there is a connection between stress, general tension, jaw symptoms and pain.  |
| 23. It feels good to have tools to tackle the problems myself if the jaw symptoms should return.   |
| 24. To get information about the cause of the jaw symptoms and about how jaw exercises works made me more implicated in the treatment.       |

### *Methodological considerations*

Research methods where qualitative and quantitative methods are mixed in order to enhance generalisability are quite common (Polit and Beck, 2010). Depending on the research question, it might be suitable to either start with a quantitative questionnaire and then follow-up with qualitative interviews or vice versa (Adams and Cox, 2008). The postal questionnaire in *study III* was based on the qualitative results of *study II* in order to investigate if the experiences of the patients in *study II* could be generalized to a larger

population. When constructing a questionnaire, it must always be considered whether the questions will have the same meaning to all respondents or not. A pilot investigation of a questionnaire is one way to test this issue (Adams and Cox, 2008). Another vital question concerns the questions' reliability and validity. The reliability and validity of the statements in the postal questionnaire in *study III* have not been investigated. Still, the statements are considered to be simple, straightforward and hard to misinterpret.

### *Study IV*

In the absence of evidence, the dental health professional must sometimes rely on experience in making treatment decisions. The assembled experience, consensus, of colleagues is better than the experience of one individual. One of the most common consensus methods in health care is the Delphi method (Murphy et al, 1998) which was used in *study IV*. The three main authors who were not a part of the expert panel, constructed a questionnaire with 31 statements regarding indications, performance, follow-ups and effectiveness of therapeutic jaw exercises in TMD patients. The statements were based on suggestions in the literature and the authors' own experiences. The questionnaire was electronic and created in the program Webropol© (Webropol Sverige AB, Linköping, Sweden). Fourteen international experts with a wide geographic dispersion (Table 3) were asked to participate in this Delphi study. An expert was defined as a person with at least 10 years of clinical experience as a TMD specialist and a research experience equivalent to at least associated professor status. The experts received carefully written information about the study before inclusion. All invited experts accepted to take part in the study.

Table 3. Geographic distribution of TMD experts.

| <b>TMD expert</b>               | <b>Geographic distribution</b> |
|---------------------------------|--------------------------------|
| Antoon De Laat                  | Belgium                        |
| Antonio Sergio Guimaraes        | Brazil                         |
| Merete Bakke                    | Denmark                        |
| Lene Baad-Hansen                | Denmark                        |
| Yrsa Le Bell                    | Finland                        |
| Nikolaos Nikitas Giannakopoulos | Germany                        |
| Ambra Michelotti                | Italy                          |
| Taro Arima                      | Japan                          |
| Frank Lobbezoo                  | The Netherlands                |
| Anders Johansson                | Norway                         |
| Anders Wänman                   | Sweden                         |
| Frauke Müller                   | Switzerland                    |
| Alan Glaros                     | USA                            |
| Richard Orbach                  | USA                            |

It was decided to continue the investigation until consensus was met or a stability between the different rounds was seen. Consensus was set to 80% (11 out of 14 experts) agreement or

disagreement (Green et al, 1999). A secondary threshold was created according to the following: If 9-10 (approximately 65-70%) out of 14 experts agreed or disagreed with the statement, consensus had not been reached but it was considered that a majority of experts agreed/disagreed. The experts were totally anonymous to each other. In *round 1*, the link to the questionnaire was e-mailed to the experts who were asked to respond to the statements according to a five-item verbal Likert scale that ranged from “Strongly agree” to “Strongly disagree”. There was also a possibility to leave free-text comments to each statement, which the experts were encouraged to do. Some of the statements had references to specific jaw exercise programs. These programs were attached to the e-mail as a pdf. A maximum of two reminders were sent if the expert did not answer. After the first round the expert received a compilation of the other experts’ responses and possible free-text comments. In this way, the expert could compare his/her answers with the answers from the other experts. Some statements for the second round were rephrased and divided after feed-back to clarify the essence of the statement. The experts were then asked to answer the refined questionnaire with 32 statements (Table 6:1, below) for *round 2*. In the same manner as earlier, the experts then received a compilation of the other experts’ responses and possible free-text comments. Subsequently, the experts were given the opportunity to check their answers and correct errors/mistakes in their input/response, concerning the statements in the questionnaire (round 2). Two experts made minor corrections of errors/mistakes in their input/response, concerning specific statements in the second questionnaire.

### *Ethical aspects*

All experts were carefully informed that participation was voluntary. In a Delphi study, the experts should be anonymous to each other and are therefore able to share their personal opinions unbiased by the possible social pressure of others. To be able to maintain a high response rate, for example through reminders, the main researcher must know the identity of the experts, which means that there is not a true anonymity in this kind of study. The method has been criticized to put pressure on the participants to converge their opinion into consensus (Keeney et al, 2001), since this is an explicit objective pronounced early in the research process. After correspondence with the Regional Ethical Review Board in Uppsala, it was concluded that this study did not need ethical vetting.

### *Methodological considerations*

The term “expert” and the assertion that a selected group of individuals represents an accurate and true “expert opinion” have been called into question (Hasson et al, 2000). Goodman (1987) emphasized the importance of selecting experts who have a great interest in the research topic. Still they must be relatively impartial so that the results reflect current knowledge (Hasson et al, 2000). In the present study the experts were chosen upon strict

criteria of clinical experience of TMD treatment, academic research experience and wide geographical dispersion. This selection process is of course exposed to bias. However, it is important to remember that these experts are chosen for a specific purpose; to share their knowledge on a specific problem. It would therefore be non-productive to randomly select experts to assure representativeness. Since there is not a high number of international TMD experts on this level, the results in this study are believed to be fairly representative. The number of participants in the expert panel has also been a subject for discussion over the years. In one study only five participants were used as an expert panel to identify serious drug interactions (Malone et al, 2005) and in another Delphi study, 2865 persons were invited to participate (only 1142 of them returned their questionnaires). Less than 10 participants are rarely seen in a Delphi study (Akins et al, 2005). A very large sample size might create more representative information, but it will also generate a larger amount of data which in turn can lead to analysis difficulties. This is particularly obvious if employing a qualitative part of the study concerning the experts' free text comments (Hasson et al, 2000). The number of participants is thus depending on the specific situation and research question. In the identified Delphi studies concerning TMD issues the number of participants was between 10-14 (John, 2010, Dawson et al, 2013, Durham et al, 2016). One of the key features and strengths of a Delphi study is the anonymity between the experts of the panel. The classical study of Asch (1956) showed how easily people change their judgement in order to fit a group majority. The normative social influence (Deutsch and Gerard, 1955) in these face to face settings, is regarded as a major influence on group judgement and decision making (Bolger and Wright, 2011). The anonymity between experts reduces this normative social influence and more true opinions are considered to be produced (Goodman, 1987). However, due to the outspoken goal of consensus and the feed-back with averages in opinions, even a Delphi method can put pressure on the participants to converge their opinions into a group consensus (Hasson et al, 2000, Bolger and Wright, 2011). True anonymity cannot be attained with the Delphi method due to the necessity that the researcher knows the panel members and their responses. True anonymity might also result in lack of accountability and thereby facilitate "ill-considered judgements" (Keeney et al, 2001). In a review by McKenna (1994) the term "quasi-anonymity" was suggested for Delphi studies which "implies that the respondents may be known to one another, but their judgements and opinions remain strictly anonymous". In the present study the main author alone could couple the experts and the graded opinions together. In the classical Delphi method, the first step is a questionnaire with only open-ended questions. These opinions are then analysed with a qualitative research method in order to produce a quantitative questionnaire for the following rounds (Keeney et al, 2001). In some modified forms of the Delphi method, this approach has been changed, where the experts in the first round receive a "pre-existing" questionnaire with information for ranking or response (Duffield, 1993). This approach could limit the possible options and also introduce bias in the responses (Hasson et al 2000). In the present study we started the first round with a pre-existing quantitative questionnaire based on the existing literature and the experience of the three main authors. Even though the degree of effectiveness of therapeutic jaw exercises in the management of TMD is still unclear (Armijo-Olivo et al, 2016), many studies have been

published concerning indications and effect of jaw exercises (Magnusson and Syrén, 1999, Michelotti et al 2004, Michelotti et al, 2005, List and Axelsson, 2010, Armijo-Olivo et al, 2016). Therefore, it was decided that it was not necessary to start with a hypothesis generating qualitative part with only open-ended questions. Nevertheless, the experts were encouraged to leave free-text comments to each statement. The feed-back from the experts after round 1, resulted in minor refinements of the questionnaire to clarify the essence of the statement. The most common way of defining consensus in a Delphi study is percentage agreement (Diamond et al, 2014). Different researchers have proposed different percentage agreement to be equated with consensus. Loughlin and Moore (1979) proposed 51%, Sumsion (1998) recommended 70% and Green et al (1999) suggested 80% agreement for the achievement of consensus. An alternative indicator of consensus used in some cases is stability of responses between a series of rounds (Diamond et al, 2014). In the present study it was decided that 11 out of 14 experts (approximately 80%) had to agree or disagree for *consensus* to be achieved. Since this decided percentage agreement for consensus is quite high, a *secondary threshold* was also created. If 9-10 (approximately 65-70%) out of 14 experts agreed or disagreed with the statement, consensus was not reached but it was concluded that a majority of experts agreed/disagreed with the statement. It is important to realize that even though consensus is met in and a majority of experts agree on different topics, this does not mean that the “truth” or correct belief has been found. The Delphi method does not replace RCTs, reviews and meta-analyses. It is merely a complement in areas where there is insufficient evidence. The classical Delphi Method consists of four rounds, but some studies recommend two or three rounds (Hasson et al, 2000, Keeney et al, 2001). Too few rounds will not generate meaningful data and too many rounds will risk sample fatigue with a decrease in response rate (Schmidt, 1997, Keeney et al, 2001). The present study was stopped after two rounds because either consensus was reach or a stability concerning responses between the rounds was seen. It might be questioned to talk about stability after only two rounds. However, it was judged that the graded opinions and free text comments showed a stability between the two rounds and therefore the study was ended even though all statements did not result in consensus. A response rate of 70% is considered to be acceptable in a Delphi study (Hasson et al, 2000). The Delphi method have been criticized for lack of reliability (Hasson et al, 2000, Keeney et al, 2001). Still, there are studies that have shown that the method indeed has a good reliability. Duffield (1993), for example, conducted a Delphi study with two expert panels of registered nurse managers. They found that the two panels agreed in 93% of the cases, which shows good reliability. The validity of the Delphi method has also been questioned. Goodman (1987) suggested that content validity can be assumed if the expert panel can be shown to be representative. With little research undertaken in this area, it can be summarized that the opinions still are divergent. Some argue that the method is both valid and reliable, while others claim that it is not (Hasson and Keeney, 2011). Considering the strict definition of an expert, the expert panels size and geographical dispersion, it can be concluded that the expert panel in the present study is representative of an international group of TMD-experts and therefore content validity in the results can be expected.



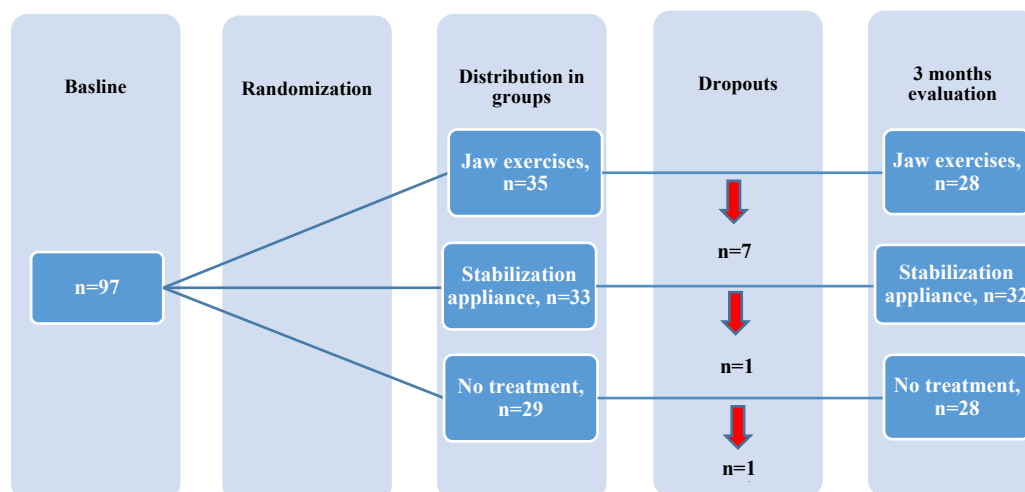
## *Study V*

In *study V* jaw exercises were compared to treatment with stabilization appliance and waiting-list controls in an RCT. The selection of patients was carried out according to the following criteria: *Inclusion criteria:* 1) Myofascial pain with or without limited mouth opening according to the RDC/TMD axis I (Dworkin and LeResche, 1992). 2) Pain (during rest and/or during jaw movements) > 40 mm on a 0-100 mm, VAS (McCormack et al, 1988). 3) Pain for a minimum of six months. 4) Age  $\geq$  18 years. The patient could also have: 1) Tension type headache. 2) Disc displacement with reduction and arthralgia of the TMJ according to the RDC/TMD axis I (Dworkin and LeResche, 1992). *Exclusion criteria:* 1) Osteoarthritis, osteoarthrosis and disc displacement without reduction according to RDC/TMD axis I (Dworkin and LeResche, 1992). 2) Complete dentures. 3) Treatment with an interocclusal appliance within the last five years. 4) Dental pain. 5) Neuropathic pain. 6) Rheumatic disease or general inflammatory condition. 7) Widespread pain, for example fibromyalgia. 8) WAD. 9) Severe morphological malocclusion (anterior open bite, pre-normality, forced cross-bite, post-normality where the horizontal over bite exceed 5 mm). 10) Known psychiatric disease. 11) Language difficulties. 12) Gross occlusal interferences hampering the possibility to make an optimal interocclusal appliance. Patients referred to the Department of Stomatognathic Physiology, the Public Dental Health service in Uppsala who fulfilled the inclusion criteria and wanted to participate were randomly divided into three groups: *therapeutic jaw exercises (group 1)*, *hard acrylic stabilization appliance (group 2)* and *“no treatment” (group 3)*. After statistical power calculation (based on earlier studies) a number of 174 patients, evenly distributed to the three groups, was considered to be enough to separate the treatment groups from each other (80% power, 1.75 SD and a significant level of 5%). The aim was to include 210 patients to control for possible dropouts. The randomisation was carried out through the use of sealed envelopes containing information about which treatment the patient should be allocated to. The preparation of the envelopes was done by a research assistant who did not participate in the other stages of the study. The 210 envelopes were constructed in one block. After including a patient in the study, the main author draw an envelope that decided which group the patient was included in and the patient was given a number between one and 210 and put on a list. This list was kept hidden to the research assistant who evaluated the patients through follow-up questionnaires. The patients could not be blind to the treatment intervention, but none of them knew which treatment that was being evaluated. The research assistant who evaluated the patients was blinded and did not know which treatment the patient had received.

Since there is no established placebo treatment for therapeutic jaw exercises, “no treatment”, i.e. waiting list patients, were chosen as a passive control. Hard acrylic stabilization appliance was chosen as an active control. The study started in September 2010 and was ended in late December 2017 due to the project’s settled time limit. During the course of this study, only 97 patients could be included according to the criteria above. After the described randomisation procedure above, these patients were distributed according to the following: 35 patients in *group 1*, 33 patients in *group 2* and 29 patients in *group 3*. Nine patients dropped

out before the final evaluation after three months (Fig 2). The mean age was 35 years (SD 18) and a majority of patients were women (77 women/79%). The mean pain duration was 25 months (SD 36).

Fig. 2. Flow chart of the study subjects. Randomisation was made through sealed envelopes. The reasons for dropout was either increased pain that demanded additional treatment or missed appointments. Data are presented as number of patients (n).



### Treatment procedure

The patients in *group 1 and 2* were all treated by the main author. The patients in *group 1* received relaxation exercises, free movements of the mandible, movements of the mandible with light resistance and finally stretching of the jaw muscles as described by Carlsson & Magnusson (1999), Fig 1. In *group 2* alginate impressions of the upper and lower jaw and an interocclusal bite registration with a wax wafer in centric relation were taken. The impressions and wax wafer were sent to a dental technician who made a hard acrylic stabilization appliance, which the patients received after two weeks. The stabilization appliance was carefully adjusted to optimal stability in centric relation allowing for a freedom in centric of approximately 0.5 to 1.0 mm. The teeth in the opposite arch were given point contact on the appliance both in centric relation and in the patient's own bite position ("centric occlusion"). It was strived for canine guidance in lateral excursions and in protrusion for group function in the frontal segment. The stabilization appliance was then polished. Carefully written patient information concerning the treatments was handed out to the patients in both treatment groups. All patients in the treatment groups were offered four appointments including the final evaluation appointment. The patients were also informed about the possibility to call the specialist clinic in case of questions or if they, due to some kind of inconvenience, needed extra appointments. The patients in the "no treatment" group

returned to the waiting list after examination. The three groups were evaluated after three months concerning change of subjective symptoms. In the two groups that received active treatment, patient adherence was also documented. After three months the “no treatment” group received indicated TMD therapy.

### *Instruments for evaluation of treatment effect*

All domains for evaluation of treatment effect according to IMMPACT (Dworkin et al, 2005 and 2008) were used, i.e. pain, global improvement, symptoms and adverse events, emotional functioning and physical functioning. 1) Pain intensity (during rest and during jaw movements) was registered by the patient on a 0-100 mm VAS with the endpoints marked with “no pain” and “worst imaginable pain” (McCormack et al, 1988). 2) Global improvement according to PGIC (Farrar et al, 2001) with the alternatives: Pain free, Much improved, Improved, Unchanged, Worse, Much worse, Very much worse. 3) Depression and anxiety according to HADS (Zigmond and Snaith, 1983). 4) JFLS-20 (Ohrbach et al, 2008). 5) Consumption of analgesics (type of analgesic, number of pills during the last month). 6) frequency of tension type headache (Almost never, 1-2 times/month, 1 time/week, Several times/week, Daily). 7) Adverse events. Pain intensity was chosen as the primary evaluation variable and a pain reduction of 30% on the VAS was considered to be a clinically relevant improvement (Farrar et al, 2001). Cost-effectiveness was measured by treatment time (minutes) and number of visits.

### *Statistics*

For test of categorical measures at one time point, the Chi-square test and the Fisher’s exact test was used. For test of continuous measures at one time point, the student’s independent t-test was used. In order to account for the repeated measures in the normal distributed outcomes (e.g. VAS at baseline and at three months) a generalized linear mixed effect model was applied included fixed effects. In a next step, a generalized estimation equation model was fitted for repeated categorical outcomes. The generalized linear mixed effect model and the generalized estimation equation model included the estimates for each visit, but also the interaction between the visits and the treatments. All the models used an unstructured covariance matrix (Breslow and Clayton, 1993, Fitzmaurice et al, 2004). These analyses were conducted using SAS 9.3 and a p-value below 5% was considered statistically significant.

### *Ethical aspects*

Normal waiting list time at the Department of Stomatognathic Physiology, Public Dental Health service, Uppsala was, at the time for the investigation, approximately 6-8 months for non-acute patients. Immediately when the referrals were received by the clinic, they were

assessed and patients who were likely to fulfil the inclusion criteria were summoned for an initial examination. This means that the patients in the control group “no treatment” did not wait longer for TMD treatment than other patients on the waiting list. Ethical approval was obtained from the Regional Ethical Review Board in Uppsala (2010/067).

### *Methodological considerations*

The power calculation concluded that a number of 174 patients, evenly distributed to the three groups, should be enough to separate the groups from each other. The aim was to include 210 patients to control for possible drop outs. During the course of the study, it was only possible to include 97 patients. The main reason for this difficulty to include patients was the exclusion criteria “Treated with an interocclusal appliance within the last five years”. Interocclusal appliance therapy is a very common treatment in general dental practice (Klasser and Greene, 2009) and therefore most patients that are referred to the Department of Stomatognathic Physiology in Uppsala have received this treatment in close time proximity. A pre-requisite for long-term treatment with a hard acrylic stabilization appliance is that the patient is skeletally fully grown and that all teeth have erupted. For this reason, only patients 18 years and older were included in the study. The randomization process with sealed envelopes was constructed after the desired number of patients (n=210) in one block and that is the reason why the number of patients was not equal between groups. A randomization in smaller blocks could have controlled the distribution between groups even though the desired number of patients was not reached. When the groups are small, as in our study, the risk for unbalanced known and unknown factors increases (Swedish Council on Health Technology Assessment in Health Care, 2018). Even though a material is randomized, different disproportions in certain patient factors can sometimes be seen (Lindh, 2013). Randomisation minimizes differences between groups in the beginning of a study, but it does not prevent differences in the treatment or assessment of outcomes between the groups. This may result in biased estimates of treatment effect. Blinding of as many involved individuals as possible in a study minimize the likelihood of such bias (Karanicolas et al, 2010). In the present study, the patients were not blind to the treatment intervention but none of the patients knew that jaw exercises was the treatment that was being evaluated. The research assistant who evaluated the patients through follow-up questionnaires was blinded. The drop-out rate in the present study was below 10% and is therefore not considered to affect the reliability of the results (Swedish Council on Health Technology Assessment in Health Care, 2018).

## RESULTS AND DISCUSSION

### *Study I*

#### *Demographics and response rate*

The response rate in the 2010 questionnaire was 71% (n=91). In the follow-up questionnaire in 2014 the response rate was 73% (n=82). The majority of the dentists were women, 70% (n=64) in 2010, 72% (n=59) in 2014, as compared to 71% (n=177) in 2001. The mean number of years in profession was 17.4 years both in 2010 (range: 1-39 years) and 2001. There was no statistically significant difference between the responders in respect of gender and working experience. In a large comprehensive review, concerning the response rate of general practicing physicians to postal questionnaires, it was concluded that the overall response rate was 61% (Creavin et al, 2011). The response rates in the present study, 71% in 2010 and 73% in 2014, must therefore be considered good for a questionnaire study. The agreement concerning the demographic factors *gender* and *number of years in profession* in the present questionnaire study and the earlier study (Tegelberg et al, 2001) was very good. Figures from the Swedish Association of Local Authorities and Regions (2014), the Swedish Association of Public Dental Officers (Member register, 2015) and the National Board of Health and Welfare (Statistical database) concerning gender (67.8 % women), working experience (mean 17.1 years) and age distribution of Swedish dentists in the public dental health care on a national level correspond well with the responders in the present study. Thus, the responders in the present study can be considered representative for dentists in the public dental health care on a national level.

#### *Quality assurance*

In 2010, only 13% of the GPDs stated that they used a health declaration containing questions on facial pain and headache. An increase in the frequency of “*regular case history of facial pain and headache*” was seen between 2010 and 2014 both in children/adolescents (28% and 45% respectively,  $p=0.027$ ) and in adults (70% and 89%, respectively,  $p=0.004$ ). Both in 2010 and 2014 significantly fewer GPDs reported taking “*regular case history of facial pain and headache*” in children/adolescents compared to in adults ( $p<0.001$ ). In 2010, 51% of the GPDs stated that they had received continuing post-graduate education about TMD. At the follow-up in 2014, 35% of the GPDs stated that they had received education about TMD during the time-period 2011-2014. A majority of these dentists (83%) had attended the TMD education program offered by the Public Dental Health service in the County of Uppsala. Due to a probable under-treatment of TMD in both adults and children/adolescents (Nilsson et al, 2005, The National Board of Health and Welfare, 2011), it is important to enhance the identification of these patients. It has been suggested that standardized questions in a health declaration could improve the detection of patients with TMD pain (Tegelberg et al, 2001).

One explanation for the difference concerning frequency of taking “*regular case history of facial pain and headache*” in children/adolescents compared with adults, might be that, in Sweden, children and adolescents are commonly examined by dental hygienists or dental assistants, and that the GPDs therefore do not ask the young patients about this anamnestic information. In 2014, the proportion of dentists that reported that they took regular case histories of facial pain and headache had increased both in adults and children/adolescents. This increase might be the result of the introduction of questions about TMD pain in the optional examination template in the computer case files. Nilsson et al (2006) have shown that these questions have a good reliability and validity in adolescents. Another factor that might have influenced the increased frequency is the strategic educational TMD program in the Public Dental Health service in Uppsala County. However, definite conclusions concerning such a connection is not possible to make due to study design and the lack of true longitudinal prospective data. In 2010, half of the GPDs reported that they had attended postgraduate education in TMD and in 2014, one third reported further continuing education in TMD (mainly by attending the internal educational program in TMD). Education and training have been shown to increase the adoption rate of new treatment technique and the frequency of good-quality care in dentistry (Dahlström et al, 2015). Continuing postgraduate TMD education is of probable importance to increase the identification of TMD patients and to improve patients’ care.

#### *Clinical experience and treatment*

Compared to 2001 fewer dentists reported in 2010 and 2014, respectively, that they had good routines and confidence in treating children/adolescents with *interocclusal appliances* (Table 4). Nevertheless, interocclusal appliance treatment was the treatment alternative in which most of the GPDs felt that they had good clinical routines and confidence both in 2010 and 2014. This finding is not surprising since earlier studies (Glass et al, 1991 and 1993, Lindfors et al, 2006) have shown that this treatment is one of the most commonly used TMD therapies. In 2010 and 2014 fewer GPDs also reported that they had a good clinical routine and confidence in *occlusal adjustments* in children/adolescents compared to 2001 (Tegelberg et al, 2001). Occlusal adjustment has been questioned as a TMD therapy for many years (Koh and Robinson, 2004, List and Axelsson, 2010). To perform reversible TMD treatments is the predominant treatment concept in Scandinavia (Carlsson and Magnusson, 1999). These two facts might have influenced and reduced the frequency of occlusal adjustment performed and thereby also the self-reported frequency of good clinical routine and confidence for this treatment. Also in adults, a majority of dentists reported that they felt insecure and did not have good clinical routines in occlusal adjustment. According to the National Guidelines for Adult Dental Care (The National Board of Health and Welfare, 2011), there are still indications for occlusal adjustment in the treatment of some types of TMD patients. It is therefore a problem that a majority of GPDs reported that they lack good clinical routines and confidence in this treatment modality.

Concerning *jaw exercises* and *pharmacological intervention* (i.e. mostly analgesics and NSAIDs), the opposite trend was found (Table 4). The proportion of GPDs that reported good clinical routines and confidence in jaw exercises when treating children/adolescents increased over time. This corroborates well with the already mentioned Scandinavian concept of reversible TMD treatments (Carlsson and Magnusson, 1999).

Table 4. Self-evaluation of clinical experience and skill concerning good routines and confidence in TMD diagnostics, therapy decision, performance of different treatments and assessment of treatment results in *children/adolescents and adults with TMD*. Comparison between different years and groups. Figures express number of responders (percentage distribution within brackets).

|                                    | 2014 (County I)<br>n= 82  |         | 2010 (County I)<br>n=91   |         | 2001 (County II-IV)*<br>n=250 | p Value  |
|------------------------------------|---------------------------|---------|---------------------------|---------|-------------------------------|--|
|                                    | Children /<br>Adolescents | Adults  | Children /<br>Adolescents | Adults  | Children / Adolescents        |  |
| Diagnostics                        | 27 (32)                   | 44 (54) | 25 (27)                   | 43 (47) | 89 (36)                       | 0.002 <sup>A</sup><br>0.001 <sup>B</sup>   |
| Therapy decision                   | 23 (28)                   | 36 (44) | 16 (18)                   | 35 (38) | 71 (28)                       | <0.001 <sup>A</sup>  |
| Performance of<br>treatment        |                           |         |                           |         |                               |  |
| Interocclusal<br>appliance         | 49 (60)                   | 68 (83) | 64 (70)                   | 75 (82) | 195 (78)                      | 0.002 <sup>B</sup><br>0.005 <sup>C</sup>   |
| Occlusal adjustment                | 15 (18)                   | 23 (28) | 17 (19)                   | 16 (18) | 105 (42)                      | <0.001 <sup>C</sup>  |
| Jaw exercises                      | 38 (46)                   | 52 (63) | 35 (38)                   | 44 (48) | 66 (26)                       | 0.041 <sup>B</sup><br>0.002 <sup>C</sup>   |
| Pharmacological<br>intervention    | 11 (13)                   | 25 (30) | 3 (3)                     | 12 (13) | 16 (6)                        | 0.031 <sup>A</sup><br>0.014 <sup>B</sup><br>0.029 <sup>D</sup><br>0.033 <sup>E</sup><br>0.011 <sup>F</sup> |
| Evaluation of<br>treatment results | 28 (34)                   | 38 (46) | 20 (22)                   | 31 (34) | 75 (30)                       | NS   |

A = between children/adolescents and adults in 2010, B = between children/adolescents and adults in 2014, C = between children/adolescents in 2001 and 2010/2014, D = between children/adolescents in 2001 and 2014, E = between children/adolescents in 2010 and 2014, F = between adults in 2010 and 2014.

**County I** = County of Uppsala.

**County II-IV** = Counties of Östergötland, Västmanland and Göteborg. \* Data from Tegelberg et al (2001).

A small, but statistically significant, increase in the frequency of good clinical routines and confidence in pharmacological intervention in children/adolescents was also seen over time. Still, the great majority of GPDs reported that they lacked good clinical routines and confidence in this treatment. Again, postgraduate continuing education in different kinds of TMD treatments is probably important.

There were no statistically significant changes concerning *diagnostics, therapy decision and evaluation of treatment results* in children/adolescents over time (2001-2010-2014). In general, few GPDs reported good clinical routines and confidence concerning these variables (Table 4). The GPDs felt more insecure concerning TMD diagnostics, therapy decisions and treatment with interocclusal appliance, jaw exercises and pharmacological intervention in children/adolescents than in adults (Table 4). During the years 2009 to 2014 the incidence of interocclusal appliances in adults increased, whereas the incidence in children/adolescents did not change (Public Dental Health Service, Uppsala. Database register). One can speculate that the more patients a dentist examines and treats, the more confident and skilled the dentist gets. This might in part explain that the GPDs felt more secure in the diagnostics and treatment of adult TMD patients. In 2010, 13% of the GPDs reported good clinical routine and confidence in pharmacological intervention of adults with TMD. The corresponding figure in 2014 was 30%. The internal strategic educational program in TMD might partly explain this increase. A majority of GPDs wanted to have the possibility to consult or refer TMD patients to an orofacial pain/TMD specialist. The complexity of TMD, self-perceived lack of knowledge and the feeling that TMD treatment is non-profitable might be some factors that can explain this demand. A vast majority of the GPDs also wanted to use the specialist for continuing education and for acquiring new knowledge. The figures from 2010 were almost identical to the figures reported in 2001 (Tegelberg et al, 2001). This means that the high need for orofacial pain/TMD specialists has been solid over a long period of time. A majority of the GPDs were positive to treat adults and about half of them were positive to treat children/adolescents with TMD. There were no statistically significant changes concerning attitudes over time. Attitudes of the GPDs have been suggested to be the most important factor in the guidance of care (Tegelberg et al, 2001). Attitudes are probably better investigated with a qualitative research method, for example a focus group study, than by a questionnaire study. The validity of the questions used in this study can be questioned. However, as mentioned earlier, the strength of this study lies in the cross-sectional follow-up design as well as in the fact that the data can be compared with earlier studies.

## *Study II*

In the systematic process of analysing the qualitative data according to STC, four main themes were identified. In the research triangulation concerning the different patterns and themes that emerged, the intersubjective agreement was 98%. The first theme ***Patient adherence*** was divided into six subgroups: *Routines, Instructions, Social context, Personality, Treatment effect and View on treatment*. The second theme ***Symptoms*** was divided into four subgroups: *Debut, Experience, Own explanations and Reactions*. The third theme ***Treatment effect*** was divided into three subgroups: *Pain, Physical impairment and Time*. The fourth theme ***Participation*** was divided into three subgroups: *Empowerment, Create trust and Knowledge and comprehension*. Below the main results are presented and discussed.



## ***Patient adherence***

### *Routines*

The patients found it difficult to incorporate the jaw exercises as a natural part of their everyday life. To find recurrent opportunities (taking a walk, go by car, tooth brushing etc.) in the already established routines (when the patients had a moment for him-/herself) and do the exercises in conjunction with these routines seemed to be a key to success. Many patients did their jaw exercises twice a day. In the middle of the day around lunchtime, many patients felt that they were too stressed to do the exercise.

*"... either when I get up in the morning or when I'm going to bed, that's when it's easiest ... you remember because it's a kind of routine when you're getting ready for bed or before you get up."*

### *Social context*

Several informants felt uncomfortable doing the jaw exercises among other people, above all in public, but also in a more private setting. The reasons for this could be that the jaw made noises during exercise, children became afraid or the informants felt that they looked peculiar when doing the exercises. One informant stressed the importance of not making the exercises too demanding and burdensome. The informant said that patients may have a lot of other exercises, for example from a physical therapist, and it is then important, especially in the beginning, to be content with oneself even if you only manage to do the jaw exercises once a day instead of the recommended three times a day.

*"... I don't do this near other people and the children are frightened when I do the stretching exercise as they think I look like a lion."*

*"... you don't want to sit next to someone who can hear what you sound like ... people would wonder what you were up to."*

### *View on treatment*

Many informants stated that they were suspicious and sceptical towards the jaw exercises in the beginning of the treatment. The jaw exercises seemed too simple and the informants had expected a more advanced treatment. Some informants thought that a surgical treatment was necessary. Patients that earlier had received physical therapy for other pain conditions reported that jaw exercises seemed reasonable and stated that they understood the connection between muscle pain and exercise as a treatment modality. When the patients started to experience a treatment effect, they were happy that "advanced treatment" such as surgery was not needed. At the end of treatment, the simplicity of therapeutic jaw exercises was what

most informants valued most. One informant stressed the positive fact that no tools are needed to do the exercises and that the exercises can be done at any place.

*“At first I was a bit skeptical about whether a couple of exercises would really help my jaw... it was good that I only needed to do a few exercises ... I didn't need an operation and all that ... that would have been awful.”*

The interviews showed that a majority of informants found it hard to incorporate the jaw exercises in their daily routine. In a previous study (Sluijs et al, 1993), concerning home exercises, it was concluded that 64% of the patients reported varying degrees of non-adherence to the prescribed exercises. Considering the effect non-adherence has on treatment outcome, it is of utmost importance to have strategies that aims to enhance adherence in clinical practice (McLean et al, 2010). Doing the jaw exercises in conjunction with an established routine seemed to increase the frequency of training. It was also important for the informants to do their training when they were alone. Consequently, it seems important to recommend the patient to do the exercises in conjunction with an already established routine and in privacy. Tooth brushing twice per day seems to be a good routine to connect the jaw exercises to. Brewer et al (2013) have shown that patients complete fewer home rehabilitation exercises on days when they feel stressed. If the patient report high levels of stress from other daily activities, they could be informed not to despair if they cannot manage to do the exercises three times per day. In such a case, once per day may be satisfactory in the beginning and then to increase the frequency later when a natural routine has been established. It has been suggested that jaw exercises should be performed several times per day for optimal effect (Michelotti et al, 2005, Moraes et al 2013). Most informants described that they were sceptical to the jaw exercises in the beginning due to its simplicity. Later on, the simplicity of the treatment was what the majority of the patients valued most. This aspect is very important to address in the beginning of treatment. A patient that is too sceptical towards a treatment is not likely to follow the instructions in an optimal way.

## ***Symptoms***

### *Experience*

Most informants had pronounced pain and fatigue localised to the cheeks, temples and neck. Jaw function aggravated their pain and they felt restricted in their mouth opening capacity. Headache was a frequently described symptom.

*“Around lunch time you get a headache that radiates from your jaw muscles all the way up to your temples and then down again to the base of your skull.”*

### *Own explanations*

Some informants said that they initially thought that the pain was a symptom of a more serious disease such as cancer. One informant was certain that the pain came from her sinus and another informant suspected that the symptoms were related to the eruption of wisdom teeth. However, most informants saw a connection between, stress, general tension and pain.

*"... when I get stressed I've noticed that the problem gets worse, plus I start breathing from my chest instead and I start to tense my jaws ... So everything is linked to stress".*

### *Reactions*

The patients reported feelings of fear of a more serious illness, panic especially in conjunction with loss of function and feelings of being unfortunate or ill-fated.

*"... then I couldn't open my mouth wide and I panicked ... " " ... I felt really unhappy about how I was being affected ... "*

Most informants initially described restricted mouth opening, a pronounced pain and fatigue localised to cheeks, temples and neck. This is in line with the description of masticatory myofascial pain in the literature (Carlsson and Magnusson, 1999). Some informants were afraid that the pain was a symptom of a more severe disease, such as cancer. Initial reassuring information about the reason for muscle pain, the treatment protocol and prognosis of treatment is of utmost importance. This has also been suggested in an earlier review (Michelotti et al, 2005). Some patients had explanatory models of their own, even though they had received information about the disorder. It seems important to ask the patients about their own thoughts about the cause of pain at an early stage and to discuss these alternative explanations. It is possible that a patient that does not believe the caregivers explanation for cause and effect might be less prone to follow the treatment recommendation. Adequate information to assist the patient in making choices and overcoming unhelpful beliefs is therefore important (Michelotti et al, 2005).

### ***Treatment effect***

#### *Pain*

Many of the patients experienced a positive sensation of the treatment already from the beginning and reported that the pain started to decrease after a couple of weeks. The pain then decreased during a longer period of time.

*“... I have less pain now, which is a great improvement. And the pain in my temples ... I haven't felt that for a long time.”*

### *Physical impairment*

Therapeutic jaw exercises had a very distinct effect on maximal mouth opening. Several of the informants said that their mouth opening capacity gradually had increased.

*“... I have noticed a difference - that I have more mobility (in my jaw) and that I can open my mouth wider.”*

A majority of the patients described a good treatment effect on both restricted mouth opening capacity and pain. These findings are in line with earlier quantitative studies on the subject (Magnusson and Syrén, 1999, Maloney et al, 2002, Nicolakis et al 2002, Michelotti et al, 2004).

### ***Participation***

#### *Empowerment*

A majority of the patients wanted to continue with their jaw exercises in the long-term in order to reduce the risk of regaining the symptoms. Some of the informants stated that they felt safe and secure and also strengthened by having the possibility to manage potential jaw symptoms themselves in the future.

*“... It feels pretty good to know that I don't need to call in as soon as I feel a bit of stiffness as I now actually have some solutions available to me.”*

#### *Knowledge and comprehension*

Simple explanation of the underlying causes of the pain and how the treatment with jaw exercises work was important to motivate patients to participate in the treatment regime. Knowledge about the underlying causes increased acceptance and awareness and also reduced the feeling of fear. In the end of the treatment, information about the prognosis was important to reduce the risk that patients would worry about a possible recurrence of their symptoms.

*“... I thought the advice was good ... they explained what the problem was and all that, and I felt like I understood”*

*“... it increases awareness, and you understand why you have to do it three times a day.”*

A majority of patients wanted to continue with the jaw exercises and some reported that they felt secure and that they, if the symptoms returned, had tools to tackle the problems themselves. In a previous study (Moraes et al, 2013), it was suggested that jaw exercises can prevent relapse of TMD pain. In all kind of pain therapy, it is important to motivate the patient to participate and take own responsibility for his/her well-being, i.e. locus of control (Michelotti et al, 2005). Positive long-term effects have been described in self-management pain programs (Lorig et al, 2001). Well-informed patients are more likely to participate in the treatment and show better adherence (Epstein et al, 2004). The information should be both verbal and written and it is important to keep the information simple and to have a clear structure.

### *Study III*

The response rate to the postal questionnaire was 73% (n=109). The majority of the patients were women (79%). The median age of the patients was 48 years (range: 18-83). There was no significant difference between responders (n=109) and non-responders (n=41) in respect of sex distribution (p=0.891) but the non-responders were significantly (p<0.001) younger than the responders with a median age of 30 years (range 18-68). The national public health survey in Sweden is a national questionnaire study on health and living conditions that comprise a random sample of 20,000 individuals (16-84 years old) and has been conducted annually between 2004 and 2016. In the beginning of the study (2004) the response rate was almost 61% but it has dropped annually and was only 47% in 2016 (The Public Health Agency of Sweden, 2016). These results show a possible downward tendency of response rates to questionnaires in Sweden. The response rate of 73% in the present study must therefore be considered as good. Both sex and age distribution among the responders corresponded well to what can be expected in patients at a TMD specialist clinic (Anastassaki and Magnusson, 2004). The non-responders were younger than the responders in the present study. In a survey from the Swedish National Board for Youth Affairs (2007) it was shown that the response rate in the age group 16-29 was only 46% compared to the age group 35-74 where the response rate was 60%. One might speculate that younger people to a greater extend have a life situation with less stability and therefore are more prone not to answer questionnaires. Thirty-five patients (32%) only received jaw exercises as treatment for their masticatory myofascial pain. Thus, a majority of patients (n=74) received jaw exercises as a part of a more extensive treatment regime. The most common combination of treatments was hard acrylic stabilization appliance, jaw exercises and information and 40 patients only received these treatments. The mean treatment time was six months (range: 3-12 months) and the mean number of appointments was five (range: 1-19). A majority of patients (71%) thought it was easier to remember the jaw exercises when they did it in connection with an already established routine and stated that it was most difficult to remember/find the time to do the jaw exercises in the middle of the day. There are many different factors that may influence a patient's acceptance and adoption of a self-management regime. The importance

of how to promote the treatment's significance in relation to e.g. cost, motivational and social aspects has been stressed (Harvey et al, 2015). In the present study 70% of the responders reported that it was difficult to find the time to do the jaw exercises in the middle of the day. This might be explained by a combination of stress at work and the fact that a majority of patients did not want to do the exercises among other people. Enhancing patient adherence should always be a goal in a treatment plan considering that low patient adherence has been reported in the treatment of chronic pain patients (Lutz et al, 1983). A number of studies have reported an increased frequency of depression, somatization, stress, anxiety, sleep dysfunction and catastrophizing thoughts in patients with TMD (including masticatory myofascial pain) compared to healthy individuals (Rollman and Gillespie, 2000, Kotiranta et al, 2015). However, in a recent systematic review (Wieckiewicz et al, 2017) it was concluded that there is not a clearly established causal relationship between mental status and masticatory myofascial pain. Half of the patients in *study III* reported that they were afraid that the pain was a symptom of a more serious disease, such as cancer. This is an example of pain catastrophizing and emphasizes the importance of initial structured information about the cause of the symptoms and their benign character. A majority of patients (78%) in the present study also reported that the information about the cause of the jaw symptoms and the mechanisms behind jaw exercises made them more involved in the treatment. The patient should always be an active part in the development of the treatment plan to optimize the chances that the patient becomes the "motor" of his/her own rehabilitation (Swedish Council on Health Technology Assessment in Health, 2006). Regarding jaw exercises, the caregiver can only provide the patient with tools for treatment. It is always the patient that has to do the job. After information, 87% of the patients reported that they understood the connection between stress, general tension, jaw symptoms, and pain. In a systematic review (Rees and Williams, 2009) it was concluded that patients with chronic illness need information to enable good self-care management at the time for diagnosis and from then onwards. In the present study a vast majority of the responders perceived the written and verbal information given as structured, good, and useful (Table 5). Seventy-seven per cent of the patients believed that it is important that the jaw exercises do not feel like a burden or cause stress and a majority did not want to do the jaw exercises among other people. A basic definition of stress is that there is "an imbalance between the demands placed on us and our ability to manage them" (Danielsson et al, 2012). If the patient has many other active treatment interventions (e.g. physiotherapy for neck problems, interocclusal appliance for TMD, psychological intervention, appointments to a general physician etc.), additional jaw exercises might be perceived as a burden. Thus, an inability to cooperate to the treatment regime might produce stress and a feeling of inadequacy. Therefore, it is very important to discuss these issues with the patient and to achieve mutual agreement concerning a tenable tailored treatment plan.

Table 5. The distribution of responses (n) to each of the 24 statements as well as the frequency of agreement (%) in 109 patients with masticatory myofascial pain answering a postal questionnaire 3 to 12 months after treatment with jaw exercises.

| Statements   | Answers        |              |         |                 |                   |           | Agreement |
|--|----------------|--------------|---------|-----------------|-------------------|-----------|-----------|
|  | Strongly agree | Partly Agree | Neutral | Partly Disagree | Strongly disagree | No answer | %         |
| 1. It was difficult to add jaw exercises as a daily routine.   | 19             | 46           | 11      | 11              | 22                | 0         | 60        |
| 2. It was easier to remember the jaw exercises when I did it in connection with an already established routine (for example tooth brushing). | 34             | 41           | 20      | 4               | 7                 | 1         | 71        |
| 3. The hardest was to remember/find the time to do the jaw exercises in the middle of the day.   | 48             | 28           | 14      | 6               | 12                | 1         | 70        |
| 4. The information concerning the jaw exercises was simple and structured.   | 85             | 18           | 1       | 4               | 1                 | 0         | 94        |
| 5. The information and the instructions at follow-ups were good and useful.  | 71             | 22           | 11      | 3               | 2                 | 0         | 85        |
| 6. I would have preferred a written information about the underlying cause of my symptoms.   | 31             | 27           | 28      | 6               | 17                | 0         | 53        |
| 7. I would rather not do jaw exercises among other people.   | 60             | 29           | 10      | 1               | 7                 | 2         | 82        |
| 8. It is important that the jaw exercises does not feel like a burden or causes stress.  | 56             | 28           | 18      | 1               | 4                 | 2         | 77        |
| 9. I had relatively severe jaw pain which motivated me to do the jaw exercises.  | 52             | 30           | 10      | 10              | 5                 | 2         | 75        |
| 10. The jaw exercises were effective and reduced my symptoms.  | 42             | 36           | 16      | 4               | 9                 | 2         | 72        |
| 11. The treatment effect motivated me to continue the training.  | 46             | 27           | 20      | 6               | 8                 | 2         | 67        |
| 12. When my jaw symptoms were reduced, I did the jaw exercises less frequently.  | 21             | 44           | 22      | 7               | 13                | 2         | 60        |
| 13. In the beginning, I was skeptical/doubtful to the effectiveness of the jaw exercises.  | 17             | 30           | 26      | 9               | 26                | 1         | 43        |
| 14. The jaw exercises felt so "simple" and I had expected a more advanced treatment.   | 22             | 21           | 19      | 13              | 33                | 1         | 39        |
| 15. Considering the effectiveness of the jaw exercises, I was pleased not to have to go through with more advanced treatment.                | 53             | 18           | 20      | 9               | 7                 | 2         | 65        |
| 16. The treatment effect of the jaw exercises came relatively fast.  | 27             | 30           | 24      | 9               | 16                | 3         | 52        |
| 17. It took a long time before I felt any effect of the jaw exercises.   | 12             | 22           | 30      | 13              | 30                | 2         | 31        |
| 18. My mouth opening capacity increased when I did the jaw exercises.  | 44             | 16           | 32      | 3               | 10                | 4         | 55        |
| 19. The treatment effect on my mouth opening capacity came more quickly than the effect on my jaw pain.                                      | 17             | 27           | 44      | 2               | 15                | 4         | 40        |
| 20. It feels good to be able to do the jaw exercises without any tools.  | 86             | 10           | 9       | 1               | 1                 | 2         | 88        |
| 21. Before I got to know what caused my jaw pain, I was afraid that the pain was a symptom of a more serious disease.                        | 25             | 28           | 14      | 9               | 31                | 2         | 49        |
| 22. I understand that there is a connection between stress, general tension, jaw symptoms and pain.  | 81             | 14           | 9       | 1               | 3                 | 1         | 87        |
| 23. It feels good to have tools to tackle the problems myself if the jaw symptoms should return.   | 77             | 10           | 15      | 0               | 6                 | 1         | 80        |
| 24. To get information about the cause of the jaw symptoms and about how jaw exercises works made me more implicated in the treatment.       | 55             | 30           | 15      | 3               | 6                 | 0         | 78        |

Agreement = frequency of patients (%) responding either "Strongly agree" or "Partly agree".

A majority (75%) of the patients in *study III* stated that severe jaw pain at the beginning of treatment, motivated them to do the jaw exercises and 67% stated that the perceived treatment

effect, made them continue with the exercises. These motivational factors have also been reported in patients who received treatment with hard acrylic interocclusal appliances for TMD problems (Lindfors et al, 2011). It is therefore important to pay attention to and reinforce a positive progress of treatment effect, such as reduction of pain and increased mouth opening capacity, even if it is minor. This approach will probably increase the patient adherence to the prescribed jaw exercises. Wig et al (2004) have also shown that patients with more severe TMD pain and limitation of jaw function, at the beginning of treatment, were more adherent to recommended treatment regimes. A majority of patients (72%) in the current study reported that the jaw exercises were effective and reduced their symptoms. This finding supports the findings in earlier clinical studies (Magnusson and Syrén, 1999, Michelotti, 2004). Considering the effectiveness of the jaw exercises, the majority of patients (65%) in the present study were pleased that they did not have to go through more “advanced” treatment. Eighty per cent of the patients reported that it felt good to have tools to tackle the problems themselves if the jaw symptoms should return. This process of becoming more confident, stronger and in control of the own life can be defined as empowerment. Empowerment can play a significant role in pain treatment and rehabilitation since the patients’ own commitment and active participation in a therapeutic program are critical factors for the treatment success (Okifuji et al, 2007). The results are presented in detail in Table 5.

### *Study IV*

In medicine and dentistry, there are divergent attitudes toward opinions that are based on clinical experience. In the hierarchy of evidence presented by Rinchuse et al (2008), “consensus opinion of experts” was considered to be number ten on a 11-point scale, where only “anecdotal reports and testimonies” was considered to be of less research quality. Clinical experience, on the other hand, is considered to be very important when research findings are applied to individual patients (Haynes et al, 2002) and also in areas such as treatment with jaw exercises, where there is insufficient research-based evidence (Murphy et al, 1998). The response rates in *study IV* was 100%. After the second round, consensus was found in 18 out of 32 statements (56%) and a majority of experts agreed/disagreed with the statements in another 8 out of 32 cases (25%). The Delphi process was stopped after the second round because either consensus was achieved or a stability between the rounds concerning graded opinions and free text comments was seen. The results are presented in Table 6:1-2. There was a consensus among the experts that jaw exercises can be recommended to patients with myalgia in the jaw muscles, chronic arthritis (in order to reduce the risk of hypomobility of the jaw), restricted mouth opening capacity due to hyperactivity in the jaw closing muscles and in cases with disc displacement without reduction.



Table 6:1. Statements used in the final Delphi questionnaire.

| <b>Statements</b> |  |
|-------------------|--|
| 1.                | I often recommend jaw exercises to my patients with temporomandibular disorders.   |
| 2.                | I recommend jaw exercises to patients with restricted mouth opening capacity due to hyperactivity in jaw closing muscles.  |
| 3.                | I recommend jaw exercises to patients with restricted mouth opening capacity due to disc displacement without reduction.   |
| 4.                | I recommend jaw exercises to patients with restricted mouth opening capacity due to radiation therapy.   |
| 5.                | I recommend jaw exercises to patients with myalgia in the jaw muscles.   |
| 6.                | I recommend jaw exercises to patients with catching/ temporarily locking (short duration) of the jaw associated with disc displacement.  |
| 7.                | I recommend jaw exercises to patients with TMJ arthralgia (not arthritis).   |
| 8.                | I recommend jaw exercises to patients with acute painful TMJ arthritis.  |
| 9.                | I recommend jaw exercises to patients with chronic TMJ arthritis (due to e.g. rheumatic disease) in order to reduce the risk of restricted mouth opening.  |
| 10.               | The patient is always instructed and given useful verbal advice on how and when to perform the jaw exercises.  |
| 11.               | The patient should always be given written information on how and when to perform the jaw exercises.   |
| 12.               | The jaw exercise program should, if possible, be individualized according to the patient's symptoms.   |
| 13.               | The patient is usually given a combination of free movements, movements against a slight resistance as well as stretching (see description under Jaw Exercise Program I in enclosed PDF).  |
| 14.               | In patients with severe pain from the jaw system, jaw exercises will in most cases aggravate the pain. Therefore, these patients only receive relaxation exercises and careful stretching.   |
| 15.               | In cases of catching/ temporarily locking (short duration) of the jaw due to suspected disk displacement, I recommend an exercise program where the patient is instructed to open and close the jaw in a protruded position (See Jaw Exercise program II in enclosed PDF). The aim of this exercise is to reduce or eliminate catching/temporarily locking of the jaw. |
| 16.               | Jaw exercises are usually not very successful in eliminating clicking of the jaw due to disk displacement.   |
| 17.               | Patients with clicking of the TMJ should not provoke these sounds when using jaw exercises, because that increases the risk of aggravating the condition of disc displacement. Thus, the patient is instructed to use only small movements that don't provoke the clicking sound.  |
| 18.               | Jaw exercises are well suited for a delegated way of working, where, for instance, a dental assistant or a physiotherapist may instruct the patient and also follow up the result of the training.   |
| 19.               | A patient who receives jaw exercises is normally followed-up after 2-3 weeks concerning cooperation/adherence. Depending on condition and severity of the symptoms, recall might deviate from this "normal standard".  |
| 20.               | In patients with inadequate adherence, re-instruction and additional check-ups (for example by telephone) may prove valuable.  |
| 21.               | A patient who has received jaw exercises is normally evaluated after 6-8 weeks. Depending on condition and severity of the symptoms, follow-up evaluation might deviate from this "normal standard".   |
| 22.               | Jaw exercises are effective in increasing the mouth opening capacity in patients with restricted mouth opening capacity due to hyperactivity in jaw closing muscles.   |
| 23.               | Jaw exercises are effective in increasing the mouth opening capacity in patients with restricted mouth opening capacity due to disc displacement without reduction.  |
| 24.               | Jaw exercises are effective in increasing the mouth opening capacity in patients with restricted mouth opening capacity due to radiation therapy.  |
| 25.               | Jaw exercises are effective in reducing or eliminating myalgia in the jaw muscles.   |
| 26.               | Jaw exercises are effective in reducing or eliminating catching/ temporarily locking (short duration) of the jaw due to disc displacement.   |
| 27.               | Jaw exercises are effective in reducing or eliminating arthralgia (not arthritis) of the TMJs.   |
| 28.               | Jaw exercises are effective in reducing or eliminating arthritis of the TMJs.  |
| 29.               | Jaw exercises might aggravate the TMD pain in some cases. Still, in general jaw exercises is a treatment without any major adverse effects.  |
| 30.               | The treatment is often begun with counseling and jaw exercises which, if necessary, may be complemented with other treatments in a later stage.  |
| 31.               | Jaw exercises are often used in combination with other treatments.   |
| 32.               | Jaw exercises are used as a sole treatment if the patient has TMD problems that evolve during daytime.   |

Table 6:2. Frequency of answers (n) by 14 TMD experts on each of the 32 statements in the final Delphi questionnaire (Table 6:1).

| Statement no. | Answers (n = 14)  |       |         |          |                      | Majority<br>9-10/14 | Consensus<br>≥11/14 |
|---------------|-------------------|-------|---------|----------|----------------------|---------------------|---------------------|
|               | Strongly<br>agree | Agree | Neutral | Disagree | Strongly<br>disagree |                     |                     |
| 1.            | 9                 | 4     | 1       |          |                      |                     | Yes                 |
| 2.            | 10                | 2     | 1       |          | 1                    |                     | Yes                 |
| 3.            | 11                | 1     | 1       | 1        |                      |                     | Yes                 |
| 4.            | 6                 | 4     | 4       |          |                      | Yes                 |                     |
| 5.            | 11                | 2     | 1       |          |                      |                     | Yes                 |
| 6.            | 4                 | 2     | 3       | 4        | 1                    |                     |                     |
| 7.            | 2                 | 7     | 3       | 1        | 1                    | Yes                 |                     |
| 8.            | 1                 | 2     | 2       | 5        | 4                    | Yes                 |                     |
| 9.            | 6                 | 5     | 3       |          |                      |                     | Yes                 |
| 10.           | 12                | 2     |         |          |                      |                     | Yes                 |
| 11.           | 8                 | 6     |         |          |                      |                     | Yes                 |
| 12.           | 12                | 2     |         |          |                      |                     | Yes                 |
| 13.           | 4                 | 4     | 5       |          | 1                    |                     |                     |
| 14.           | 4                 | 6     | 3       |          | 1                    | Yes                 |                     |
| 15.           | 2                 | 7     | 2       | 2        | 1                    | Yes                 |                     |
| 16.           | 2                 | 11    | 1       |          |                      |                     | Yes                 |
| 17.           | 2                 | 7     | 2       | 2        | 1                    | Yes                 |                     |
| 18.           | 4                 | 9     | 1       |          |                      |                     | Yes                 |
| 19.           | 6                 | 7     |         | 1        |                      |                     | Yes                 |
| 20.           | 4                 | 8     | 2       |          |                      |                     | Yes                 |
| 21.           | 4                 | 5     | 2       | 3        |                      | Yes                 |                     |
| 22.           | 7                 | 6     | 1       |          |                      |                     | Yes                 |
| 23.           | 8                 | 3     | 2       | 1        |                      |                     | Yes                 |
| 24.           | 3                 | 2     | 9       |          |                      |                     |                     |
| 25.           | 5                 | 6     | 3       |          |                      |                     | Yes                 |
| 26.           | 2                 | 4     | 3       | 4        | 1                    |                     |                     |
| 27.           | 3                 | 4     | 6       | 1        |                      |                     |                     |
| 28.           |                   | 1     | 3       | 8        | 2                    | Yes                 |                     |
| 29.           | 3                 | 10    | 1       |          |                      |                     | Yes                 |
| 30.           | 2                 | 10    | 2       |          |                      |                     | Yes                 |
| 31.           | 5                 | 9     |         |          |                      |                     | Yes                 |
| 32.           | 1                 | 4     | 3       | 3        | 3                    |                     |                     |

Majority = 9-10 out of 14 TMD experts either agreed or disagreed with the statement. Consensus = ≥11 out of 14 TMD experts either agreed or disagreed with the statement.

A majority of experts also recommended jaw exercises to patients with arthralgia of the TMJs (not arthritis) and patients with restricted mouth opening capacity due to radiation therapy. The majority did not recommend jaw exercises in patients with acute painful arthritis. Jaw exercises were considered to be effective (consensus) in the treatment of myalgia in the jaw muscles and in increasing mouth opening capacity due to hyperactivity in jaw closing muscles and disc displacement without reduction, but the treatment was considered not to be very successful in eliminating clicking of the joints due to disc displacement.

Two RCTs support the opinion that jaw exercises are effective in the treatment of TMD myalgia (Magnusson & Syrén 1999, Michelotti et al 2004). The sample size in these studies were small and it is therefore difficult to draw general conclusions from an evidence-based point of view. The studies of Tegelberg et al (1988, 1996) lend support to the achieved consensus that jaw exercises could be recommended to patients with chronic arthritis (due to e.g. rheumatic disease) to reduce the risk of hypomobility of the jaw. In cases of acute painful arthritis, the first choice of treatment is pharmacological management (Häggman-Henrikson et al, 2017) and a majority of the experts concluded that jaw exercises are not to be recommended in these patients. Studies have shown that jaw exercises can be effective in patients with restricted mouth opening capacity due to disc displacement without reduction (Yuasa and Kurita, 2001, Minakuchi et al, 2001, Haketa et al, 2010) and hyperactivity in jaw closing muscles (Maloney et al, 2002). Other studies have not been able to demonstrate that jaw exercises produce a significant increase of mouth opening capacity compared to controls in these patient-groups (Craane et al 2012). Despite these contradictory results, the expert panel came to a consensus that jaw exercises are effective and can be recommended in patients with restricted mouth opening capacity due to these two conditions. A majority of experts also recommended jaw exercises to patients with restricted mouth opening capacity due to radiation therapy, but there was no consensus/majority for the effectiveness of jaw exercises in this condition. In a study on patients that had been treated with radiotherapy for head and neck cancer, Loorents et al (2014) showed that there was no significant difference in effect on mouth opening capacity between jaw training with TheraBite<sup>®</sup> compared to a control group. In contrast, Pauli et al (2016) showed that jaw exercises with emphasis on stretching with TheraBite<sup>®</sup> or the Jaw Trainer (Engström device), had a significant positive effect on mouth opening capacity in patients with head and neck cancer after treatment with radiotherapy compared to matched controls. In a systematic review, Kamstra et al (2017) concluded that a large variation of stretching techniques and performances of jaw exercises have been presented in studies on trismus in head and neck cancer patients. No stretching/exercise technique was more effective than the other and a majority of studies reported an increase in mouth opening capacity after treatment. Clicking of the TMJs due to disc displacement is a common condition (Elfving et al 2002) and it seldom progresses into more serious conditions such as locking of the TMJs (Magnusson et al, 1994). Some studies have shown that jaw exercises can reduce the frequency of clicking of the TMJs to a great extent (Au and Klineberg, 1993, Yoda et al, 2003) while another study showed only minor effect on the clicking sounds (Nicolakis et al 2000). There was a consensus in the expert

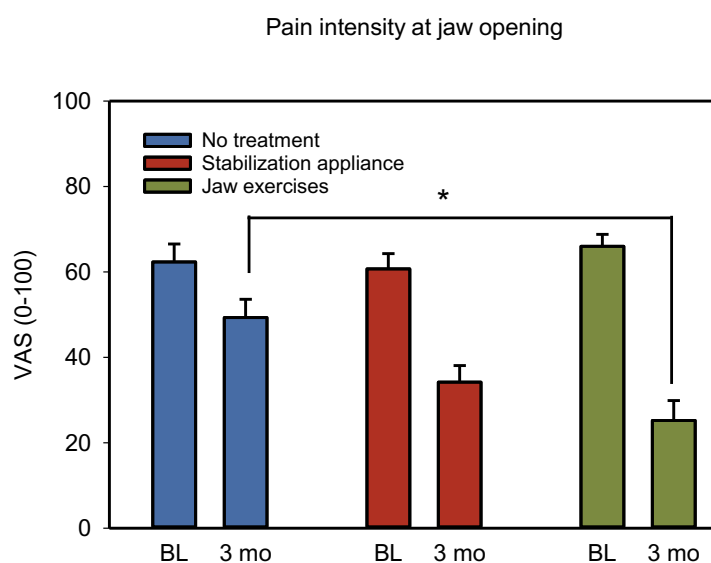
panel that jaw exercises are usually not very successful in eliminating clicking of the joints due to disc displacement. Even though the clicking sound is not harmful, it might give the patient discomfort. In *study II* it was shown that some patients are afraid that TMD pain is a symptom of a more serious disease. It can be speculated that a TMJ symptom such as clicking, can provoke similar fears. Initial reassuring information about the cause of the clicking sound and their benign character are therefore important. There was no consensus among the experts regarding the actual way of doing the exercises. It has been reported that exercise intervention for spinal pain based on patient-specific tailored intervention, compared to standardized protocols has the potential to improve treatment outcome (Falla and Hodges, 2017). In exercise treatment for patients with fibromyalgia and related syndromes, it is stressed that the patient population is heterogeneous and because of that, the prescription of exercise must be individualized (Mannerkorpi and Iversen, 2003). In TMD patients the individualized prescription of jaw exercises to each patient and his/her condition and severity of symptoms is probably as important as in other pain conditions. Therefore, it might be difficult to reach consensus concerning a standardized treatment protocol. The recommendation of short term follow-up of jaw exercises after 2-3 weeks met the definition of consensus and a majority of experts also recommended evaluation of the exercises after 6-8 weeks. Follow-ups and evaluations must also always be individualized depending on the patient's adherence, on the specific condition and on severity of symptoms. Even though jaw exercises might aggravate TMD pain in some cases, it was considered to be a treatment without any major adverse effects (consensus). In the current study there was also consensus that jaw exercises are well suited for delegated work. Chronic pain management is often team-based (Driscoll and Kerns, 2016). Because of a scarcity of TMD experts, teamwork in the management of TMD is a necessity. A dental assistant or a physiotherapist may well instruct the patient in jaw exercises and also follow up the results (Durham et al, 2016). In the light of the results from *study IV*, there is obviously substantial clinical experience and knowledge that points towards the conclusion that jaw exercises are effective and highly indicated in a number of different TMD conditions. These results, based on the knowledge and clinical experience of a group of TMD experts, are important as guidelines for the general practitioner in absence of solid evidence for the effectiveness of jaw exercises in the management of TMD (Armijo-Olivo et al, 2016).

### *Study V*

Pain intensity was the primary outcome variable in *study V* and a pain reduction of 30% on the VAS was considered to be a clinically relevant improvement (Farrar et al, 2001). VAS pain scores during rest decreased in all three groups and there were no statistically significant differences between groups. Only jaw exercises showed a statistically significant difference compared to no treatment ( $p < 0.001$ ) for reduction of pain intensity during jaw movement according to VAS scores (Fig. 3). It might be tempting to conclude, that jaw exercises therefore are more efficient than stabilization appliances in reducing pain due to

masticatory myofascial pain, but there was no statistically significant difference between the treatment groups supporting such a claim. In conformity with these results, other studies (Magnusson and Syrén, 1999, Michelotti et al, 2012) comparing jaw exercises and occlusal appliance therapy in patients with TMD myalgia have not been able to show statistically significant differences between the groups. In the current study, a statistically significant higher number of patients in the jaw exercise group received a 30% and 50% reduction of pain intensity on VAS, compared to the no treatment group ( $p=0.011$  and  $p<0.001$ , respectively). The same difference was shown between stabilization appliance and no treatment, but only at the 50% level ( $p=0.007$ ). Systematic reviews (Swedish Council on Health Technology Assessment in Health Care, 2006, Friction et al, 2010) have earlier concluded that stabilization appliance is more effective in reducing TMD pain than no treatment. There were no statistically significant differences between the treatment groups concerning number of patients who received 30% or 50% pain reduction (Fig. 4).

Fig 3. Changes in primary outcome variable pain intensity according to Visual Analog Scale (VAS) scores (during jaw movement). Mean values are presented. The asterisk indicates the following statistically significant differences between jaw exercises and no treatment; \*  $p<0.001$ .

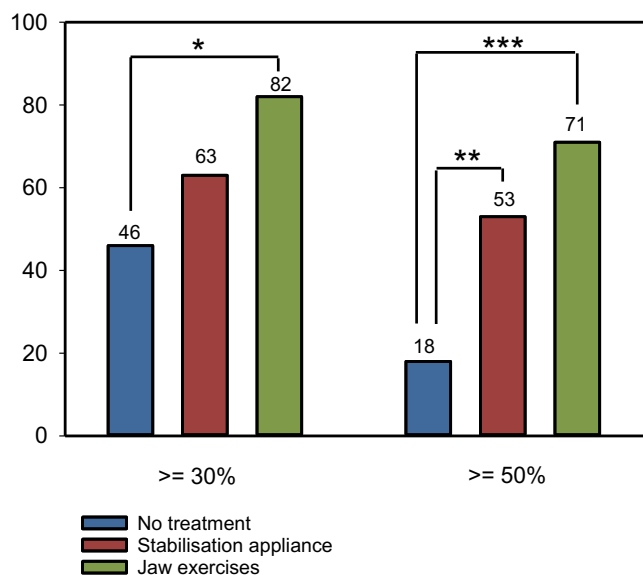


According to PGIC, the patients in the treatment groups reported a greater improvement compared to the no treatment group ( $p<0.001$ ), in which most patients stated an unchanged or worsened situation. There was no statistically significant difference between treatment groups in this aspect. Pain is a very subjective feeling and in the absence of objective outcomes, PGIC is considered to be a clinically relevant tool to assess the subjective perception of treatment impact in pain management (Rampakakis et al, 2015). There was a statistically significant decrease in the frequency of both headache and consumption of analgesics in the jaw exercise group compared to the no treatment group ( $p=0.028$  and  $p=0.007$ , respectively). In the stabilization appliance group there was also a statistically

significant decrease in consumption of analgesics but not in headache frequency compared to the no treatment group ( $p=0.02$ , Table 7).

The comorbidity between primary headaches such as tension-type headache, migraine and TMD is well established (Speciali and Dach, 2015). Several studies (Magnusson T, 1981, Caspersen et al, 2013) have reported a correlation between tension-type headache and TMD and recommend TMD treatment in patient with TMD and recurrent headache. Ekberg et al (2002) reported a positive effect on tension-type headache after treatment with stabilization appliances in patients with TMD of mainly arthrogenous origin. Considering these interactions, multidisciplinary teams of both physicians and orofacial pain specialists has been recommended in headache cases in order to attain the most precise diagnosis and efficient treatment (Marklund et al, 2014, Conti et al, 2016).

Fig 4. Changes in pain intensity according to the highest original Visual Analog Scale (VAS) scores (during rest or jaw movement) at evaluation and after 3 months. Equal to or more than 30 and 50% reduction of pain intensity are presented. Numbers in per cent. The asterisk indicates the following statistically significant differences between treatment groups and the no treatment group; \*  $p=0.011$ , \*\*  $p=0.007$  \*\*\*  $p<0.001$ .



Both treatment groups showed a reduction of analgesic consumption compared with the no treatment group. It is reasonable that a reduction in the need of analgesics is a sign of a reduction of pain intensity which were, as mentioned above, shown for both treatments. Wright et al (2006) concluded that stabilization appliance and self-management therapies reduced the consumption of analgesics with 18% in patients with TMD and headache. There was a statistically significant reduction of JFLS scores in the jaw exercise group compared to the no treatment group ( $p=0.008$ ), but there were no differences over time concerning HADS scores (Table 7). Doepel et al (2012) have shown that occlusal appliances reduce the scores

of JFLS-20 in patients with myofascial pain according to RDC/TMD (Dworkin and LeResche, 1992).

Table 7. Secondary outcome variables at examination/baseline (BL) and evaluation after 3 months (3 mo). Data are presented as number of patients (per cent) or median (range). If there is an even number of numbers in the set, then median is set as the average of the two numbers in the middle.

|                                  | No treatment<br>(n=28) |             | Stabilization appliance<br>(n=32) |             | Jaw exercises<br>(n=28) |             | p-value            |
|----------------------------------|------------------------|-------------|-----------------------------------|-------------|-------------------------|-------------|--------------------|
|                                  | BL                     | 3 mo        | BL                                | 3 mo        | BL                      | 3 mo        |                    |
| <b>Tension type headache</b>     |                        |             |                                   |             |                         |             |                    |
| Missing data                     | 1 (4)                  | 0           | 0                                 | 0           | 1 (4)                   | 1 (4)       | A. 0.028           |
| Almost never                     | 1 (4)                  | 4 (14)      | 3 (9)                             | 3 (9)       | 2 (7)                   | 8 (29)      |                    |
| 1-2 times/month                  | 5 (18)                 | 3 (11)      | 7 (22)                            | 9 (28)      | 10 (36)                 | 11 (39)     |                    |
| 1 time/week                      | 7 (25)                 | 7 (25)      | 6 (19)                            | 10 (31)     | 8 (29)                  | 4 (14)      |                    |
| Several times / week             | 8 (29)                 | 8 (29)      | 5 (16)                            | 6 (19)      | 3 (11)                  | 2 (7)       |                    |
| Daily                            | 6 (21)                 | 6 (21)      | 11 (34)                           | 4 (13)      | 4 (14)                  | 2 (7)       |                    |
| <b>Consumption of analgesics</b> |                        |             |                                   |             |                         |             |                    |
| Missing data                     | 0                      | 0           | 0                                 | 0           | 0                       | 1 (4)       | A. 0.007           |
| Never                            | 3 (11)                 | 4 (14)      | 4 (13)                            | 8 (25)      | 4 (14)                  | 11 (39)     |                    |
| 1-2 times/month                  | 7 (25)                 | 9 (32)      | 5 (16)                            | 14 (44)     | 10 (36)                 | 10 (36)     | B. 0.02            |
| 1 time/week                      | 4 (14)                 | 2 (7)       | 8 (25)                            | 2 (6)       | 6 (21)                  | 2 (7)       |                    |
| Several times/week               | 8 (29)                 | 6 (21)      | 10 (31)                           | 7 (22)      | 6 (21)                  | 3 (11)      |                    |
| Daily                            | 6 (21)                 | 7 (25)      | 5 (16)                            | 1 (3)       | 2 (7)                   | 1 (4)       |                    |
| <b>HADS</b>                      |                        |             |                                   |             |                         |             |                    |
| Depression                       | 3.5 (0-16)             | 2.0 (0-18)  | 2.5 (0-9)                         | 3.0 (0-10)  | 1.0 (0-7)               | 1.0 (0-6)   | C. <0.001, D. 0.02 |
| Anxiety                          | 6.5 (0-18)             | 6.5 (0-19)  | 8.0 (0-18)                        | 7.5 (0-17)  | 5.0 (0-13)              | 4.0 (0-14)  | D. 0.03            |
| <b>JFLS-20</b>                   |                        |             |                                   |             |                         |             |                    |
| Median score                     | 1.5 (0-8.3)            | 1.6 (0-6.1) | 1.1 (0-6.3)                       | 0.5 (0-6.0) | 2.0 (0-5.7)             | 0.4 (0-5.7) | A. 0.008           |

The letters indicate the following statistically significant differences: A. Between jaw exercises and no treatment over time. B. Between stabilization appliance and no treatment over time. C. Between jaw exercises and no treatment at baseline. D. Between jaw exercises and stabilization appliance at baseline. HADS = Hospital Anxiety and Depression Scale; JFSL = Jaw function Limitation Scale.

Concerning treatment costs, jaw exercises are cheaper than the stabilization appliance which is associated with a dental technician fee which in Sweden is approximately 170 US dollars. In *study V*, both treatment groups were offered four appointments including the final evaluation. Some patients called and wanted more appointments and some cancelled appointments because they felt they were not needed. In the jaw exercise group, the patients had fewer appointments (mean three appointments) and a shorter mean treatment time (24 minutes, range: 14-52) compared to the group that received stabilization appliance treatment (mean four appointments and 47 minutes, range: 29-69). These differences were statistically significant ( $p < 0.001$ ) for both comparisons. It can be concluded that jaw exercises is a more cost-effective treatment compared to stabilization appliance with fewer visit to the clinic, lower overhead costs and a lower mean treatment time. This finding is interesting since Magnusson & Syrén (1999) found the opposite relation concerning treatment time. This difference between studies can perhaps be explained by differences in number of patients and local clinical routines. The no treatment group was returned to the waiting list after examination and therefore only received two appointments. Since the

control group (no treatment) was not offered as many appointments as the treatment groups, it is not known if the treatment effect reported in this study, is due to the specific effect of the actual treatment or if it is due to more general effects such as e.g. empathic therapists (Karlsson and Bergmark, 2015). It has also been suggested that asking patients to wait for treatment puts them in a stalled stage of change where they become passive and do not move toward action on their own. This might result in less improvement than in a simply untreated group where the patients might seek treatment elsewhere or attempt behavioral changes on their own (Cunningham et al, 2013). Still, reduction of pain intensity was also seen in the no treatment group. This can probably be explained by the regression to the mean (Bland and Altman, 1994).



## GENERAL DISCUSSION

Since pain is a very subjective experience, it is difficult to quantify. The importance to study pain and pain treatment both in a qualitative and a quantitative way has therefore been stressed (Osborn and Rodham, 2010). Consequently, pain conditions and their treatments should be illustrated both objectively through RCTs and subjectively through qualitative research methods where a deeper understanding of the patients' experiences can be investigated. The caregivers' experiences are also important in the evaluation of a treatment modality and should be included to produce a fair overall impression of a treatment. The focus of this thesis was to gain a better understanding concerning the different aspects of jaw exercises in the treatment of masticatory myofascial pain. Both quantitative and qualitative research methods were used with special emphasis directed toward efficacy, cost-effectiveness, patients' views and dentists' experiences.

### *Self-reported experiences of general practicing dentists*

In *study I* it was concluded that the GPDs felt more insecure concerning TMD diagnostics, therapy decisions and treatment in children/adolescents compared to adults. This kind of insecurity can probably be coupled to lack of education and clinical experience. A majority of dentists also reported that they felt insecure and did not have good clinical routines in occlusal adjustment in both adults and children/adolescents. According to the National Guidelines for Adult Dental Care (The National Board of Health and Welfare, 2011), there are still indications for occlusal adjustment in the treatment of some types of TMD patients. It is therefore a problem that a majority of GPDs reports a lack of good clinical routines and confidence in this treatment modality. The importance of continuing professional development to improve clinical practice in dentistry has been emphasized (Belfield et al, 2001). Continuing professional development can be undertaken in many different ways, for example theoretical and practical courses, conferences, specialist consultations in the clinic of the GPDs, auscultation or clinical work with a specialist and journal reading groups (NHS, 1999). It is not only the individual performance of the caregiver that improves with continuing professional development. The performance of colleagues in the dental team is also likely to improve due to favourable effects of working with a capable colleague. There might also be positive economic effects in the dental clinic since capable and skilled practitioners are likely to have lower frequency of retreatments, shorter treatment times and more efficient patient care (Belfield et al, 2001). The European Parliament has recommended compulsory continuing postgraduate education for dentists and in many European countries it has been introduced, but Sweden is an exception in this aspect (Suslick J, 2013). In the present thesis, it was shown that GPDs reported a high need for orofacial pain/TMD specialists and a majority of the GPDs wanted the specialists to offer continuing education in

TMD. The responsibility of continuing professional development lies both on the individual GPD and the possible employer.

Most GPDs reported that they felt confident and had good clinical routines in interocclusal appliance therapy. This finding is not surprising since earlier studies have shown that this treatment is one of the most commonly used TMD therapies (Glass et al, 1991 and 1993, Lindfors et al, 2006). The proportion of GPDs that reported good clinical routines and confidence in jaw exercises when treating children/adolescents increased over time. This corroborates well with the already mentioned Scandinavian concept of reversible TMD treatments (Carlsson and Magnusson, 1999). Also, the proportion of GPDs that reported that they ask their patients about facial pain and headache increased over time. This increase might partly be explained by the introduction of questions about TMD pain in an optional examination template in the computer case files. Nilsson et al (2006) have shown that these questions have a good reliability and validity in adolescents. Lövgren et al (2016) have also shown that these questions are a valid tool to identify adult TMD patients in need of supplementary examination and treatment. However, even though TMD patients are identified, a majority of patients still do not receive indicated TMD treatment (Lövgren et al, 2017). It is obvious that it is not enough only to have tools to identify a TMD patient, you also need clinical guidelines that describe the management of such a patient.

#### *Patient experiences of jaw exercises*

The results of *study II* and *III* indicate that it is important to recommend the patient to do the jaw exercises in conjunction with an already established routine, for example tooth brushing, to enhance adherence. Treatments can never be effective if the patient do not follow the treatment recommendations (Davies, 1968, Geboy and Ingersoll, 1983). Insufficient adherence is an extensive problem where studies have shown that 30% to 70% of all patients fail to adhere to recommended health instructions (Sackett, 1976, Kirscht and Rosenstock, 1979, Roter et al, 1998). Low patient adherence has also been found in the treatment of chronic pain. In patients who had completed a multidisciplinary pain program, patient adherence was low, averaging around 42% for individual regimens (Lutz et al, 1983). All possible ways of increasing adherence are therefore important and working with already established routines seems to be an easy way of doing it. The results also show that a large proportion of patients are afraid that the pain due to TMD myalgia is a symptom of a serious disease. Initial information about the cause of the symptoms, their benign character, as well as the favourable prognosis of the treatment are therefore mandatory parts for reassurance of the patient. Information also makes the patient more involved in the treatment and likely to adhere to the treatment (Epstein et al, 2004), which probably is important for treatment success. Consequently, it is important to incorporate well designed and structured information in all stages of examination and treatment of patients with TMD. A majority of the patients reported that jaw exercises were effective in reducing symptoms of TMD myalgia and the patients felt safe and secure because they had a tool (jaw exercises) to tackle the problems

themselves if the symptoms should return. Due to the complexity of pain and the lack of objective measurements, the researcher has to rely on self-reported outcome variables of the patient (Younger et al, 2009). The subjective reports from the patients in this thesis clearly states that jaw exercises are effective. The patients also seem to become more confident, stronger and in control when using jaw exercises. This process can be defined as empowerment and can play a significant role in pain treatment and rehabilitation, since the patients' own commitment and active participation in a treatment are critical factors for the long-term success (Okifuji et al, 2007).

### *The consensus of TMD specialists*

The results of *study IV* show that there is an international consensus among TMD specialists/experts that jaw exercises are effective in the treatment of myalgia in the jaw muscles and in increasing mouth opening capacity due to hyperactivity in jaw closing muscles and disc displacement without reduction. Even though jaw exercises might aggravate TMD-pain in some cases, the experts considered it to be a treatment without any major adverse effects. These results are important as guidelines for the general practitioner in absence of solid evidence for the effectiveness of exercises in the treatment of TMD. Most TMD specialists in Sweden work in the Public Dental Health service (Håkansson, 2006). As a part of a larger organization, the specialist often acts as a sounding board for the GPDs in difficult clinical situations where advice and recommendations are given. The results of this thesis give strength to the recommendation of jaw exercises in the management of TMD (Carlsson and Magnusson, 1999). In *study IV*, there was also consensus regarding the following practical issues of jaw exercises: 1) The patients should always get verbal and written information about the treatment, 2) short term follow-up is recommended after 2-3 weeks and 3) jaw exercises are well-suited for delegated work. This kind of results are important in the construction of practical guidelines concerning jaw exercises.

### *The efficacy of jaw exercises*

The results of the RCT (*study V*) showed that that jaw exercises are more effective than no treatment in reducing pain intensity, headache and consumption of analgesics in patients with TMD myalgia. Jaw exercises are also cost-effective when compared to treatment with stabilization appliance. These results strengthen earlier studies showing that jaw exercises seem to be effective in the management of TMD myalgia (Magnusson and Syrén, 1999, De Laat et al, 2003, Michelotti et al, 2004). Jaw exercises as a standalone treatment also seems to have similar effect as occlusal appliance on the reduction of headache (Ekberg et al, 2002) and occlusal appliance/self-management treatment on consumption of analgesics (Wright et al, 2006). Chronic myofascial pain in the orofacial region can lead to large expenses for the community as well as for the individual patient. In the US the annual treatment-cost for TMD

is estimated to approximately 4 billion dollars (Gatchel, et al, 2006). Thus, early cost-effective treatments are necessary to reduce health care costs and unnecessary suffering for the patients. Considering the results of *study V*, jaw exercises, in combination with information/counselling, should be recommended as a first line of treatment in the management of masticatory myofascial pain.

### *Methodological limitations*

The main weakness of the questionnaire in *study I* is that the questions' validity and reliability has not been investigated. Another weakness of the follow-up part of the material is that it is not known if the responders in the two questionnaires were the same and the results can therefore not be seen as longitudinal prospective data. Still, the relative low turnover rate of GPDs in the Public Dental Health service in the County of Uppsala, the high response rate in both questionnaires and the cross-sectional design lend strength to the results and allow for comparison of results between the questionnaires. The central criticism of all qualitative studies, including *study II*, is that results and conclusions never can be generalized to a population level. However, the results can in some instances be transferred to a different context. Through a thorough description of the research process, the study population and the context in which the study took place, it is possible for the reader to decide the degree of transferability of the results of *study II*. *Study III* has the same weakness as *study I* concerning the validity and reliability of the questions in the questionnaire. Nevertheless, the statements that were based on the results of *study II* were considered to be simple, straightforward and hard to misinterpret. The major methodological weakness in *study IV* is the modified Delphi approach, where the study was stopped after only two rounds because either consensus was reached or a stability concerning responses between the rounds was seen. It can be questioned if stability can be reached after only two rounds. Finally, in *study V* the main weakness is the number of participants. The power calculation concluded that a number of 174 patients, evenly distributed to the three groups, should be enough to separate the treatment groups from each other. During the course of the study, only 97 patients were included.

### *Implications*

The results of this thesis emphasize continuing professional development and the importance of quality assurance concerning the GPDs knowledge and clinical experience. The Public Dental Health service or private employers must, through quality assurance, identify areas in need of improvement and offer tailor-made education to their employees. The need of TMD-specialists has been high during a long period of time. Counties that do not have their own specialists must try to solve this unmet need by e.g. trying to employ consultant TMD-specialists from other counties or educate own dentists to become specialists. The findings concerning the patients' experiences of jaw exercises in treatment of masticatory myofascial

pain indicate that there are several areas that must be emphasized in treatment with jaw exercises. These results, in combination with the consensus of TMD-specialists seen in the Delphi study, can create a basis for the development of practical guidelines for the treatment with jaw exercises. The patients' experiences, the consensus of TMD-specialists and the results of the RCT suggest that jaw exercises are effective in the treatment of TMD myalgia. In Sweden there is an increasing shortage of experienced dentists and a large part of the working dentists will retire shortly. The long-term prognoses to the year 2035 also reveal an unchanged situation concerning the supply and demand of dentists (The National Board of Health and Welfare, 2018). Because of a scarcity of both general dentists and TMD experts, teamwork in the management of TMD is a necessity. Jaw exercises is a treatment that is well suited for a delegated way of working. A dental assistant may well instruct the patient in jaw exercises and also follow up the results (Durham et al, 2016). Considering the findings that jaw exercises are effective in the treatment of TMD myalgia and also was shown to be more cost-effective compared to stabilization appliance, it can be recommended as a first treatment of choice in many patients with TMD myalgia.

#### *Future research*

Continuous studies concerning the GPDs knowledge, experiences and clinical routines in TMD treatment is needed as quality assurance to identify areas in need of improvement. Due to a probable under-treatment of TMD in both adults and children/adolescents (Nilsson et al, 2005, The National Board of Health and Welfare, 2011), it is important to enhance the identification of these patients. Studies concerning both identification of TMD patients and factors influencing further assessment and treatment of identified TMD patients are warranted. As mentioned earlier a combination of qualitative and quantitative research methods are important in the study of pain. More studies with this combination are needed in the research field of TMD pain to get a deeper and more versatile understanding of the condition. The present RCT showed that jaw exercises were more effective than no treatment in reducing pain intensity in patients with masticatory myofascial pain. Still, the number of participants were too few to achieve enough power to be able to separate the two treatment groups from each other. More RCTs with higher number of participants and meta-analyses concerning the efficacy of jaw exercises are needed.

## CONCLUSIONS

- General practicing dentists seem to be more insecure concerning treatment with jaw exercises in children/adolescents compared to adults, but their confidence with the treatment increased over time. There is a high need for orofacial pain/TMD specialists and a majority of the GPDs wants the specialists to offer continuing education in TMD (*study I*).
- To enhance adherence, patients should be recommended to perform jaw exercises in conjunction with an already established routine (*study II and III*).
- Information about the cause of the symptoms, their benign character, as well as the favourable prognosis of the treatment of TMD is important for reassurance of the patient and for treatment success (*Study II and III*).
- Jaw exercises empower patients and give them self-confidence (*study II and III*).
- There is an international consensus among TMD specialists/experts that jaw exercises is an effective treatment of myalgia in the jaw muscles, reduced mouth opening and disc displacement without reduction. Even though jaw exercises might aggravate TMD-pain in some cases, it is considered to be a treatment without any major adverse effects (*study IV*).
- Treatment with jaw exercises is more effective than no treatment in reducing pain intensity, headache and consumption of analgesics in patients with TMD myalgia. Jaw exercises are also more cost-effective than treatment with stabilization appliance (*study V*).

## POPULÄRVETENSKAPLIG SAMMANFATTNING

Långvarig smärta i ansikte och käkar är vanligt förekommande och studier har visat att ungefär 5-12% av den vuxna befolkningen drabbas. Käkmuskelsmärta är ett av de vanligaste tillstånden som orsakar smärta i ansiktsregionen. Flertalet av de drabbade är kvinnor och smärtan är ofta förenad med svårigheter att gapa, tuggproblem, huvudvärk och trötthet i käkarna. Orsaken till varför man får käkmuskelsmärta är inte helt klarlagd, även om flera faktorer har föreslagits. Genetiska faktorer, stress, hormonella orsaker och tandgnissling/tandpressning är några möjliga orsaker som diskuterats. Smärtan påverkar ofta både livssituationen och livskvaliteten negativt då flera viktiga funktioner störs och besvären ibland leder till sjukskrivning. Om smärtan inte behandlas kan de leda till stora kostnader för både individ och samhälle i form av exempelvis inkomstförlust och sjukskrivningskostnader. Om patienterna inte får effektiv behandling av besvären inom tandvården är det även vanligt att de istället belastar andra delar av sjukvården, ofta utan tillfredställande behandlingsresultat och med ökade vårdkostnader som följd. Tidiga, kostnadseffektiva behandlingsmetoder är alltså viktiga för att både minska kostnader och patientens lidande. Behandling av smärta i ansikte och käke har beskrivits i över ett sekel. Två av de vanligaste behandlingsformerna är bettskena och rörelseträning för käken. Bettskena är den behandling som har det bästa vetenskapliga stödet och man vet att behandlingen är effektiv när det gäller att minska både käkmuskelsmärta och smärta från käkleder. Flera studier har även utvärderat effekten av rörelseträning vid behandling av käkmuskelsmärta. Rörelseträningen syftar till att uppnå avslappning i käkmuskulaturen, förbättra funktionen och minska smärtan. Vissa studier har visat mycket lovande resultat som talar för att rörelseträning kan vara en effektiv behandling. Studierna som gjorts är dock inte tillräckligt omfattande för att man ska kunna dra några säkra slutsatser. Den övergripande målsättningen med denna avhandling är att förbättra kunskapen kring rörelseträning som behandling av käkmuskelsmärta. Fokus har lagts på att undersöka behandlingens effekt och kostnadseffektivitet i jämförelse med behandling med bettskena respektive ingen behandling. Patienternas upplevelser av behandlingsformen och tandläkarnas erfarenheter ur både ett allmäntandvårds- och specialisttandvårdsperspektiv har undersökts. I *studie I* analyserades allmäntandläkarnas självrapporterade erfarenhet av diagnostik, behandlingsval och behandling av patienter med smärta i ansikte och käke. Attityder kring behandling av denna patientgrupp och behovet av specialistresurser undersöktes också. Informationsinhämtning skedde via en web-enkät som skickades ut till samtliga allmäntandläkare inom folktandvården i Uppsala år 2010 (128 st) och 2014 (113 st). Enkäten skickades ut vid dessa två tillfällen för att möjliggöra jämförelser över tid och resultaten jämfördes även med en tidigare studie från 2001. Det visade sig att tandläkarna generellt kände sig mer osäkra när det gällde diagnostik, behandlingsval och behandling av barn/ungdomar jämfört med vuxna med smärta i ansikte och käkar. Betydligt färre tandläkare rapporterade också att de frågade sina barn/ungdomspatienter om ansiktssmärta och huvudvärk jämfört med vuxna patienter. Över tid ökade antalet tandläkare som rapporterade att de ställde frågor till sina patienter kring ansiktssmärta/huvudvärk. Över en längre

tidsperiod uppgav tandläkarna att de blev säkrare på behandling med rörelseträning. Bettskena var den behandling där flest tandläkare kände sig säkra och uppgav att de hade en god klinisk rutin. En majoritet av tandläkarna hade en positiv alternativt en neutral attityd till behandling av såväl barn/ungdomar som vuxna med smärta i ansikte och käkar. Det visade sig även att det finns ett stort behov av specialister på området och en majoritet av tandläkarna vill att specialisterna erbjuder vidareutbildning. I *studie II* studerades vilka upplevelser patienter med käkmuskelsmärta har av behandling med rörelseträning. Smärta är en av de mest subjektiva och komplexa upplevelser som finns. För att erhålla djupare förståelse och kunskap om ett smärttillstånd och dess behandling är det viktigt att få kunskap om patientens upplevelser och erfarenheter. Dessa fenomen undersöks bäst med en så kallad kvalitativ forskningsmetod, exempelvis en intervjustudie, som tar hänsyn till känslomässiga, psykologiska, sociala och existentiella aspekter. Tio patienter intervjuades enligt en fastställd intervjuguide. Intervjuerna spelades in och överfördes senare till text som analyserades med metoden systematisk textkondensering. Resultaten visar att flera patienter till en början trodde att smärtan var ett symptom på en mer allvarlig sjukdom, exempelvis cancer, och att en del patienter var skeptiska till rörelseträning som behandlingsmetod på grund av dess ”enkelhet”. Innan behandling påbörjas är det därför viktigt att informera patienterna om att käkmuskelsmärta är ett ofarligt tillstånd med god prognos. Patienterna upplevde med tiden att behandlingen gav smärtreduktion. Rörelseträningens enkelhet var en av de saker som patienterna i slutändan uppskattade mest. Att göra rörelseträningen i anslutning till en redan etablerad rutin, exempelvis tandborstning, verkade vara viktigt för att patienterna lättare skulle komma ihåg att utföra träningen. Flera patienter uppgav att de ville fortsätta med rörelseträningen och att de kände sig stärkta genom att ha ”verktyg” att själva kunna ta till om besvären skulle återkomma. Resultaten från kvalitativa intervjustudier kan inte generaliseras, men det är möjligt att resultaten kan överföras till en liknande kontext. I *studie III* undersöktes därför om resultaten från studie II kunde generaliseras till en större grupp patienter med käkmuskelsmärta som fått behandling med rörelseträning. Utifrån resultaten i intervjustudien skapades en enkät som skickades till 150 patienter som behandlats för käkmuskelsmärta inom specialisttandvården i Uppsala. Enkäten bestod av 24 påståenden och patienterna ombads gradera hur väl dessa överensstämde med deras uppfattning enligt en femgradig skala (från *instämmer helt* till *instämmer inte alls*). Svarefrekvensen var 73% vilket är bra för en enkätstudie. En majoritet av patienterna rapporterade att det var lättare att komma ihåg rörelseträningen om de satte den i samband med en redan etablerad rutin. Patienterna uppgav att information om den bakomliggande orsaken till besvären gjorde dem mer involverade i behandlingen. Hälften av patienterna misstänkte till en början att deras symptom berodde på en allvarlig sjukdom, exempelvis cancer. Flertalet patienter rapporterade att rörelseträningen var effektiv och reducerade deras symptom och uppgav även att det kändes bra att själva kunna ta till rörelseträningen om besvären skulle återkomma. Flertalet resultat från studie II kunde således generaliseras till en större grupp patienter. Sammanfattningsvis upplever patienter med käkmuskelsmärta att rörelseträning är en effektiv behandling. Information är viktig både i lugnande syfte och för att göra patienten mer delaktig i behandlingen. Rörelseträning stärker patienter och ger dem verktyg att själva kunna



påverka sin situation om besvären skulle återkomma. I situationer där det inte finns tillräckligt med vetenskapligt stöd för en behandling så måste vårdgivaren använda sin erfarenhet eller ännu hellre, den samlade erfarenheten hos kollegiet d.v.s. konsensus, för att fatta behandlingsbeslut. I *studie IV* undersöktes vilka erfarenheter och uppfattningar en grupp internationella experter hade och om konsensus kunde uppnås gällande rörelseträning som behandling av smärta och funktionsstörningar i käksystemet. Det finns flera olika metoder för att undersöka konsensus. En av de vanligaste metoderna är Delfi-metoden som fått sitt namn efter oraklet i Delfi. Metoden går ut på att en expertpanel svarar på en enkät i olika omgångar. Mellan varje omgång får panelens deltagare information om hur de övriga svarat i förhållande till de egna svaren. I de flesta fall konvergerar svaren över tid till en gemensam uppfattning, en konsensus. En fördel med metoden är att deltagarna är anonyma, vilket minskar risken för negativ social påverkan. Fjorton internationella experter tillfrågades om de ville ingå i expertpanelen och samtliga accepterade. En web-baserad enkät konstruerades utifrån befintlig litteratur och skickades till experterna. Experterna instruerades att svara på varje påstående enligt en femgradig skala (från *instämmer helt* till *instämmer inte alls*) och uppmuntrades även att lämna synpunkter i fri text. Svarsfrekvensen var 100 %. Efter första omgången fick deltagarna en sammanställning där de egna svaren kunde ses i relation till de övriga experternas svar. Därefter fick experterna svara på enkäten i en andra omgång. Studien avslutades efter två omgångar. Resultaten från denna studie visar att det finns konsensus för att rörelseträning är en effektiv behandling av käkmuskelsmäta och vid gapomfångsinskränkning orsakad av överaktivitet i käkmuskulaturen eller diskförskjutningsproblematik. Rörelseträning bör alltid individanpassas och även om rörelseträning ibland kan öka käkmärtan anses det vara en behandling utan några allvarliga biverkningar. I avsaknad av vetenskapligt stöd för rörelseträning kan dessa resultat utgöra riktlinjer för allmäntandläkare vid val av behandling. I *studie V* studerades behandlingseffekten av rörelseträning vid behandling av käkmuskelsmäta i en randomiserad kontrollerad studie. Behandlingen jämfördes med bettskena och ”ingen behandling”. Nittiosju patienter med käkmuskelsmäta fördelades slumpvis till de tre grupperna och utvärderades efter tre månader avseende subjektiva besvär. Behandlingarna jämfördes även avseende kostnadseffektivitet. Resultaten visar att rörelseträning effektivt minskar smärta, huvudvärk och konsumtion av smärtstillande läkemedel hos patienter med käkmuskelsmäta. Behandling med rörelseträning kräver färre besök och mindre behandlingstid i jämförelse med behandling med bettskena och är således en kostnadseffektiv behandling.

Sammanfattningsvis visar resultaten av denna avhandling att allmäntandläkare har blivit säkrare på behandling med rörelseträning över tid. Patienter med käkmuskelsmäta upplever att rörelseträning är en effektiv behandling och experter på området rekommenderar denna behandling vid käkmuskelsmäta och inskränkt gapomfång. Rörelseträning minskar käkmuskelsmäta, huvudvärk och behovet av smärtstillande läkemedel och är en kostnadseffektiv behandling jämfört med bettskena.

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## REFERENCES

- Adams A, and Cox AL. Questionnaires, In-depth Interviews and Focus Groups. In: Cox, A. L., and Cairns, P. (Eds.). *Research Methods Human-Computer Interaction*. Cambridge University Press, Cambridge. 2008: 17–34.
- Aggarwal VR, Lovell K, Peters S, et al. Psychosocial interventions for the management of chronic orofacial pain. *Cochrane Database Syst Rev* 2011; 11: CD008456.
- Akins RB, Tolson H, Cole BR. Stability of response characteristics of a Delphi panel: application of bootstrap data expansion. *BMC Med Res Methodol*. 2005; 1; 5:37.
- Al-Baghdadi M, Durham J, Araujo-Soares V, Robalino S, Errington L, Steele J. TMJ Disc Displacement without Reduction Management: A Systematic Review. *J Dent Res*. 2014; 93: 37S-51S.
- Al-Jundi MA, John MT, Setz JM, Szentpétery A, Kuss O. Meta-analysis of treatment need for temporomandibular disorders in adult nonpatients. *J Orofac Pain*. 2008; 22:97-107.
- Anastassaki A, Magnusson T. Patients referred to a specialist clinic because of suspected temporomandibular disorders: a survey of 3194 patients in respect of diagnoses, treatments, and treatment outcome. *Acta Odontol Scand*. 2004; 62: 183-192.
- Armijo-Olivo S, Pitance L, Singh V, Neto F, Thie N, Michelotti A. Effectiveness of Manual Therapy and Therapeutic Exercise for Temporomandibular Disorders: Systematic Review and Meta-Analysis. *Phys Ther*. 2016; 96: 9-25.
- Asch SE. Studies of independence and conformity: a minority of one against a unanimous majority. *Psychological Monographs: General and Applied*, 1956; 70: 1-70.
- Asmundson, GJ et al. Let's get physical: a contemporary review of the anxiolytic effects of exercise for anxiety and its disorders. *Depress Anxiety*. 2013; 30: 362-73.
- Au AR, Klineberg JJ. Isokinetic exercise management of temporomandibular joint clicking in young adults. *J Prosthet Dent*. 1993; 70: 33-9.
- Austin BD, Shupe SM. The role of physical therapy in recovery after temporomandibular joint surgery. *J Oral Maxillofac Surg*. 1993; 51: 495-8.
- Bandura A. Self-efficacy: toward a unifying theory of behavioral change. *Psychol Rev*. 1977; 84: 191-215.
- Bandura A. *Self-efficacy: The Exercise of Control*. New York: Freeman. 1997.
- Belfield CR, Morris ZS, Bullock AD, Frame JW. The benefits and costs of continuing professional development (CPD) for general dental practice: a discussion. *Eur J Dent Educ*. 2001; 5: 47-52.
- Berger M, Szalewski L, Bakalczuk M, et al. Association between estrogen levels and temporomandibular disorders: A systematic literature review. *Prz Menopauzalny* 2015; 14: 260–270.
- Bjelland I, Dahl AA, Haug TT, Neckelmann D. The validity of the Hospital Anxiety and Depression Scale. An updated literature review. *J Psychosom Res* 2002; 52: 69–77.
- Bland JM, Altman DG. Regression towards the mean. *BMJ*. 1994; 308: 1499.

- Bolger F, Wright G. Improving the Delphi process: Lessons from social psychological research. *Technol Forecast Soc Change*. 2011; 78: 1500-1513.
- Breivik H, et al. Survey of chronic pain in Europe: prevalence, impact on daily life, and treatment. *Eur J Pain*. 2006; 10: 287-333.
- Breivik H, Borchgrevink PC, Allen SM, Rosseland LA, Romundstad L, Breivik Hals EK, Kvarstein G, Stubhaug A. Assessment of pain. *Br J Anaesth*. 2008; 101: 17–24.
- Breivik H, Eisenberg E, O'Brien T. The individual and societal burden of chronic pain in Europe: the case for strategic prioritisation and action to improve knowledge and availability of appropriate care. *BMC Public Health*. 2013; 13: 1229.
- Breslow NE, Clayton DG. Approximate inference in generalized linear mixed models. *Journal of the American Statistical Association* 1993; 88: 9–25.
- Brewer BW, Cornelius AE, Van Raalte JL, Tennen H, Armeli S. Predictors of adherence to home rehabilitation exercises following anterior cruciate ligament reconstruction. *Rehabil Psychol*. 2013 ; 58: 64-72.
- Britten N, Jones R, Murphy E, Stacy R. Qualitative research methods in general practice and primary care. *Fam Pract* 1995; 12: 104-14.
- Broberg C, Tyni-Lenné R. Sjukgymnastik som vetenskap och profession. Legitimerade sjukgymnasters riksförbund. (In Swedish). 2009.
- Buchbinder D, Currivan RB, Kaplan AJ, Urken ML. Mobilization regimens for the prevention of jaw hypomobility in the radiated patient: a comparison of three techniques. *J Oral Maxillofac Surg*. 1993; 51: 863-7.
- Burgess JA, Sommers EE, Truelove EL, Dworkin SF. Short-term effect of two therapeutic methods on myofascial pain and dysfunction of the masticatory system. *J Prosthet Dent*. 1988; 60: 606-10.
- Carlson CR, Bertrand PM, Ehrlich AD, Maxwell AW, Burton RG. Physical self-regulation training for the management of temporomandibular disorders. *J Orofac Pain*. 2001; 15: 47-55.
- Carlson CR, Okeson JP, Falace DA, et al. Comparison of psychologic and physiologic functioning between patients with masticatory muscle pain and matched controls. *J Orofac Pain* 1993; 7: 15–22.
- Carlsson GE, Magnusson T. Management of Temporomandibular Disorders in the General Dental Practice. Chicago (IL): Quintessence Publishing; 1999. p. 93–121.
- Carlsson C. Grundläggande Akupunktur. Studentlitteratur AB, in Swedish, 1992. ISBN 91-44-34461-9.
- Caspersen N, Hirsvang JR, Kroell L, Jadidi F, Baad-Hansen L, Svensson P, Jensen R. Is There a Relation between Tension-Type Headache, Temporomandibular Disorders and Sleep? *Pain Res Treat*. 2013; 2013: 845684.
- Carmeli E, Sheklow SL, Bloomenfeld I. Comparative study of repositioning splint therapy and passive manual range of motion techniques for anterior displaced temporomandibular discs with unstable excursive reduction. *Physiotherapy* 2001; 87: 26–36.

Clark GT. Classification, causation and treatment of masticatory myogenous pain and dysfunction. *Oral Maxillofac Surg Clin North Am.* 2008; 20: 145-57.

Conti PC, Costa YM, Gonçalves DA, Svensson P. Headaches and myofascial temporomandibular disorders: overlapping entities, separate managements? *J Oral Rehabil.* 2016; 43: 702-15.

Costen JB. A syndrome of ear and sinus symptoms dependent upon disturbed function of the temporomandibular joint. *Ann Otol Rhinol Laryngol* 1934; 43: 1-15.

Craane B, Dijkstra PU, Stappaerts K, De Laat A. One-year evaluation of the effect of physical therapy for masticatory muscle pain: a randomized controlled trial. *Eur J Pain.* 2012; 16: 737-47.

Craane B, Dijkstra PU, Stappaerts K, De Laat A. Randomized controlled trial on physical therapy for TMJ closed lock. *Journal of dental research.* 2012; 91: 364-9.

Cramer CK, Klasser GD, Epstein JB, Sheps SB. The Delphi process in dental research. *J Evid Based Dent Pract.* 2008; 8: 211-20.

Creavin ST, Creavin AL, Mallen CD. Do GPs respond to postal questionnaire surveys? A comprehensive review of primary care literature. *Fam Pract.* 2011; 28: 461-7.

Creswell JW. *Qualitative Inquiry and Research Design: Choosing Among Five Traditions.* 2nd ed. London: Sage Publications; 2012: 7-11, 20-4.

Cunningham JA, Kypri K, McCambridge J. Exploratory randomized controlled trial evaluating the impact of a waiting list control design. *BMC Med Res Methodol.* 2013; 13: 150.

Dahlström L, Molander A, Reit C. The impact of a continuing education programme on the adoption of nickel-titanium rotary instrumentation and root-filling quality amongst a group of Swedish general dental practitioners. *Eur J Dent Educ.* 2015; 19: 23-30.

Dalkey N, Helmer O. An experimental application of the Delphi method to the use of experts. Memorandum RM-727/1-Abrigaded. 1962.

Danielsson M, Heimerson I, Lundberg U, Perski A, Stefansson CG, Akerstedt T. Psychosocial stress and health problems: Health in Sweden: The National Public Health Report 2012. Chapter 6. *Scand J Public Health.* 2012; 40: 121-134.

Dawson A, Raphael KG, Glaros A, et al. Development of a quality-assessment tool for experimental bruxism studies: reliability and validity. *J Orofac Pain.* 2013; 27: 111-22.

De Felício CM, de Oliveira MM, da Silva MA. Effects of orofacial myofunctional therapy on temporomandibular disorders. *J Craniomand Pract.* 2010; 28: 249-59.

De Felício CM, Melchior Mde O, Ferreira CL, Da Silva MA. Otologic symptoms of temporomandibular disorder and effect of orofacial myofunctional therapy. *J Craniomand Pract.* 2008; 26: 118-25.

De Kanter RJ, Truin GJ, Burgersdijk RC, Van 't Hof MA, Battistuzzi PG, Kalsbeek H, Käyser AF. Prevalence in the Dutch adult population and a meta-analysis of signs and symptoms of temporomandibular disorder. *J Dent Res.* 1993; 72: 1509-18.

De La Torre Canales G, Câmara-Souza MB, Muñoz Lora VRM, Guarda-Nardini L, Conti PCR, Garcia RMR, Del Bel Cury AA, Manfredini D. Prevalence of psychosocial impairment in Temporomandibular Disorder patients: a systematic review. *J Oral Rehabil.* 2018. doi: 10.1111/joor.12685. [Epub ahead of print].

De Laat A, Stappaerts K, Papy S. Counseling and physical therapy as treatment for myofascial pain of the masticatory system. *J Orofac Pain.* 2003; 17: 42-9.

De Leeuw R, Bertoli E, Schmidt JE, et al. Prevalence of traumatic stressors in patients with temporomandibular disorders. *J Oral Maxillofac Surg* 2005; 63: 42–50.

Deutsh M, Gerard HG. A study of normative and informational social influence upon individual judgment. *J. Abnorm. Soc. Psychol.* 1955; 51: 629–36.

Diamond IR, Grant RC, Feldman BM, Pencharz PB, Ling SC, Moore AM, Wales PW. Defining consensus: a systematic review recommends methodologic criteria for reporting of Delphi studies. *J Clin Epidemiol.* 2014; 67: 401-9.

Dinas, PC, Koutedakis, Y, Flouris AD. Effects of exercise and physical activity on depression. *Ir J Med Sci.* 2011; 180: 319-25.

Doepel M, Nilner M, Ekberg E, LE Bell Y. Long-term effectiveness of a prefabricated oral appliance for myofascial pain. *J Oral Rehabil.* 2012; 39: 252-60.

Driscoll MA, Kerns RD. Integrated, team-based chronic pain management: Bridges from theory and research to high quality patient care. *Adv Exp Med Biol.* 2016; 904: 131-47.

Durham J, Al-Baghdadi M, Baad-Hansen L, et al. Self-management programmes in temporomandibular disorders: results from an international Delphi process. *J Oral Rehabil.* 2016; 43: 929-936.

Duffield C. The Delphi technique: a comparison of results obtained using two expert panels. *International journal of nursing studies.* 1993; 30: 227-37.

Dworkin RH, Turk DC, Farrar JT et al. Core outcome measures for chronic pain clinical trials: IMMPACT recommendations. *Pain.* 2005; 113: 9-19.

Dworkin RH, Turk DC, Wyrwich KW et al. Interpreting the clinical importance of treatment outcomes in chronic pain clinical trials: IMMPACT recommendations. *J Pain.* 2008; 9: 105-21.

Dworkin SF, LeResche L. Research diagnostic criteria for temporomandibular disorders: review, criteria, examinations and specifications, critique. Review. *J Craniomandib Disord* 1992; 6: 301-55.

Dworkin SF, LeResche L, DeRouen T, Von Korff M. Assessing clinical signs of temporomandibular disorders: reliability of clinical examiners. *J Prosthet Dent.* 1990; 63: 574-9.

Edwards RR, et al. The role of psychosocial processes in the development and maintenance of chronic pain. *The Journal of Pain.* 2016; 17: 70-92.

Ekberg EC, Nilner M. The influence of stabilisation appliance therapy and other factors on the treatment outcome in patients with temporomandibular disorders of arthrogenous origin. *Swed Dent J.* 1999; 23: 39-47.

- Ekberg E, Vallon D, Nilner M. Treatment outcome of headache after occlusal appliance therapy in a randomised controlled trial among patients with temporomandibular disorders of mainly arthrogenous origin. *Swed Dent J* 2002; 26: 115-24.
- Ekberg E, Nilner M. Treatment outcome of appliance therapy in temporomandibular disorder patients with myofascial pain after 6 and 12 months. *Acta Odontol Scand* 2004; 62: 343-9.
- Elfving L, Helkimo M, Magnusson T. Prevalence of different temporomandibular joint sounds, with emphasis on disc-displacement, in patients with temporomandibular disorders and controls. *Swed Dent J*. 2002; 26: 9-19.
- Enthoven P, Skargren E, Oberg B. Clinical course in patients seeking primary care for back or neck pain: a prospective 5-year follow-up of outcome and health care consumption with subgroup analysis. *Spine*. 2004; 29: 2458-2465.
- Epstein RM, Alper BS, Quill TE. Communicating evidence for participatory decision making. *J Am Med Assoc*; 2004; 291: 2359-66.
- Eriksson PO, Häggman-Henrikson B, Nordh E, Zafar H. Co-ordinated mandibular and head-neck movements during rhythmic jaw activities in man. *J Dent Res*. 2000; 79: 1378–1384.
- Eversole LR, Machado L. Temporomandibular joint internal derangements and associated neuromuscular disorders. *J Am Dent Assoc*. 1985; 110: 69-79.
- Falla D, Hodges PW. Individualized exercise interventions for spinal pain. *Exerc Sport Sci Rev*. 2017; 45: 105-115.
- Farrar JT, Young JP Jr, LaMoreaux L, Werth JL, Poole RM. Clinical importance of changes in chronic pain intensity measured on an 11-point numerical pain rating scale. *Pain* 2001; 94: 149-58.
- Fillington RB, Ohrbach R, Greenspan JD, et al. Psychological factors associated with development of TMD: The OPPERA prospective cohort study. *J Pain* 2013; 14: T75–90.
- Fitzmaurice GM, Laird NM, Ware JH. *Applied Longitudinal Analysis*. New York, Wiley, 2004.
- Flor H, Turk DC. *Chronic pain: an Integrated Biobehavioral Approach*. 2015: Lippincott Williams & Wilkins.
- Fornasari D. Pain mechanisms in patients with chronic pain. *Clinical Drug Investigation*. 2012. 32: 45-52.
- Forsell H, Kalsö E. Application of principles of evidence-based medicine to occlusal treatment for temporomandibular disorders: are there lessons to be learned? *J Orofac Pain*. 2004; 18: 9-22; discussion 23-32.
- Fricton JR, Schiffman EL. The craniomandibular index: validity. *J Prosthet Dent*. 1987; 58: 222-8.
- Fricton J, Look JO, Wright E, et al. Systematic review and meta-analysis of randomized controlled trials evaluating intraoral orthopedic appliances for temporomandibular disorders. *J Orofac Pain* 2010; 24: 237–254.
- Fysioterapeuterna. *Specialistordning för fysioterapeuter*. (In Swedish). Inrättad 1993. Reviderad 2014.



- Gagnon CM, Scholten P, Atchison J. Multidimensional Patient Impression of Change Following Interdisciplinary Pain Management. *Pain Pract.* 2018. doi: 10.1111/papr.12702. [Epub ahead of print]
- Gatchel RJ, Stowell AW, Wildenstein L, et al. Efficacy of an early intervention for patients with acute temporomandibular disorder-related pain: a one-year outcome study. *J Am Dent Assoc* 2006; 137: 339-47.
- Gavish A, Winocur E, Astandzelov-Nachmias T, Gazit E. Effect of controlled masticatory exercise on pain and muscle performance in myofascial pain patients: A pilot study. *J Craniomand Pract.* 2006; 24: 184-90.
- Geboy MJ, Ingersoll BD. Patient adherence to dental regimens. *Compend Contin Educ Dent* 1983; 4: 185-90.
- Geisser ME, Clauw DJ, Strand V, Gendreau RM, Palmer R, Williams DA. Contributions of change in clinical status parameters to Patient Global Impression of Change (PGIC) scores among persons with fibromyalgia treated with milnacipran. *Pain.* 2010; 149: 373-8.
- Geneen LJ et al. Physical activity and exercise for chronic pain in adults: an overview of Cochrane Reviews. *Cochrane Database Syst Rev.* 2017; 24; 4:CD011279.
- Gerdle B, Bjork J, Henriksson C, Bengtsson A. Prevalence of current and chronic pain and their influences upon work and healthcare-seeking: a population study. *J Rheumatol.* 2004; 31: 1399-1406.
- Gesch D, Bernhardt O and Kirbschus A. Association of malocclusion and functional occlusion with temporomandibular disorders (TMD) in adults: A systematic review of population-based studies. *Quintessence Int* 2004; 35: 211–221.
- Gesch D, Bernhardt O, Mack F, et al. Association of malocclusion and functional occlusion with subjective symptoms of TMD in adults: Results of the Study of Health in Pomerania (SHIP). *Angle Orthod* 2005; 75: 183–190.
- Glass EG, Glaros AG, McGlynn FD. Myofascial pain dysfunction: treatments used by ADA members. *J Craniomandib Pract.* 1993; 11: 25-9.
- Glass EG, McGlynn FD, Glaros AG. A survey of treatments for myofascial pain dysfunction. *J Craniomandib Pract.* 1991; 9: 165-8.
- Goodman C M. The Delphi technique: a critique. *Journal of advanced nursing.* 1987; 12: 729-734.
- Goodwillie DH. Arthritis of the temporo-maxillary articulation. *Arch Med* 1881; 5: 259-63.
- Grace EG, Sarlani E, Reid B. The use of an oral exercise device in the treatment of muscular TMD. *J Craniomand Pract.* 2002; 20: 204-8.
- Graneheim UH, Lundman B. Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Educ Today.* 2004; 24: 105-12.
- Green B, Jones M, Hughes D, Willimas A. Applying the Delphi technique in a study of GP's information requirements. *Health and Social Care in the Community.* 1999; 7: 198-205.
- Gustavsson C. Self-Management of Persistent Neck Pain – A Multi-Component Group Intervention in Primary Health Care. Uppsala: Uppsala university; 2011.

- Hadhazy VA, Ezzo J, Creamer P, Berman BM. Mind-body therapies for the treatment of fibromyalgia. A systematic review. *J Rheumatol*. 2000; 27: 2911-2918.
- Haketa T, Kino K, Sugisaki M, Takaoka M, Ohta T. Randomized clinical trial of treatment for TMJ disc displacement. *Journal of Dent Res*. 2010; 89: 1259-63.
- Hamberg K, Johansson E, Lindgren G, Westman G. Scientific rigour in qualitative research - examples from a study of women's health in family practice. *Fam Pract*. 1994; 11: 176-81.
- Hansson P, Ekblom A. Transcutaneous electrical nerve stimulation (TENS) as compared to placebo TENS for the relief of acute oro-facial pain. *Pain* 1983; 15: 157-65.
- Harvey J, Dopson S, McManus RJ, Powell J. Factors influencing the adoption of self-management solutions: an interpretive synthesis of the literature on stakeholder experiences. *Implement Sci*. 2015; 13: 10:159.
- Hasson F, Keeney S, McKenna H. Research guidelines for the Delphi survey technique. *J Adv Nurs*. 2000; 32: 1008-15.
- Hasson F, Keeney S. Enhancing rigour in the Delphi technique research. *Technological Forecasting & Social Change*. 2011; 78: 1695–1704.
- Haynes RB, Devereaux PJ, Guyatt GH. Clinical expertise in the era of evidence-based medicine and patient choice. *ACP J Club*. 2002; 136: 11-14.
- Helkimo M. Studies on function and dysfunction of the masticatory system. II. Index for anamnestic and clinical dysfunction and occlusal state. *Sven Tandlak Tidskr*. 1974; 67: 101-21.
- Herrmann C. International experiences with the Hospital Anxiety and Depression Scale—a review of validation data and clinical results. *J Psychosom Res* 1997; 42: 17–41.
- Hogg-Johnson S, van der Velde G, Carroll LJ, et al. The burden and determinants of neck pain in the general population: results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders. *Spine*. 2008; 33: 39-51.
- Holdgate A, Asha S, Craig J, Thompson J. Comparison of a verbal numeric rating scale with the visual analogue scale for the measurement of acute pain. *Emerg Med* 2003; 15: 441–6.
- Huang GJ, LeResche L, Critchlow CW, Martin MD, Drangsholt MT. Risk factors for diagnostic subgroups of painful temporomandibular disorders (TMD). *J Dent Res*. 2002; 81: 284-8.
- Hurwitz EL, Carragee EJ, van der Velde G, et al. Treatment of neck pain: noninvasive interventions: results of the Bone and Joint Decade 2000-2010 Task Force on Neck Pain and Its Associated Disorders. *Spine*. 2008; 33: S123-152.
- Håkansson G. Kartläggning av Specialisttandvården. Avd för Vård och Omsorg. Sveriges Kommuner och Landsting. Dnr 2006/1906.
- Häggman-Henrikson B, Alstergren P, Davidson T, Högestätt ED, Östlund P, Tranaeus S, Vitols S, List T. Pharmacological treatment of orofacial pain - health technology assessment including a systematic review with network meta-analysis. *J Oral Rehab*. 2017; 44: 800-26.
- Häggman-Henrikson B, List T, Westergren HT, Axelsson SH. Temporomandibular disorder pain after whiplash trauma: a systematic review. *J Orofac Pain*. 2013; 27: 217–226.

- Häggman-Henrikson B, Rezvani M, List T. Prevalence of whiplash trauma in TMD patients: a systematic review. *J Oral Rehabil.* 2014; 41: 59-68.
- Häggman-Henrikson B, Wiesinger B, Wänman A. The effect of supervised exercise on localized TMD pain and TMD pain associated with generalized pain. *Acta Odontol Scand.* 2018; 76: 6-12.
- Ioannidis JP, Evans SJ, Gøtzsche PC, O'Neill RT, Altman DG, Schulz K, Moher D; CONSORT Group. Better reporting of harms in randomized trials: an extension of the CONSORT statement. *Ann Intern Med.* 2004; 141: 781-8.
- Johannes CB, et al. The prevalence of chronic pain in United States adults: results of an Internet-based survey. *J Pain.* 2010; 11: 1230-9.
- John MT. Improving TMD classification using the Delphi technique. *J Oral Rehabil.* 2010; 37: 766-70.
- Kalamir A, Graham PL, Vitiello AL, Bonello R, Pollard H. Intra-oral myofascial therapy versus education and self-care in the treatment of chronic, myogenous temporomandibular disorder: A randomised, clinical trial. *Chiropr Man Therap* 2013 ; 21: 17.
- Kamstra JJ, van Leeuwen M, Roodenburg JLN, Dijkstra PU. Exercise therapy for trismus secondary to head and neck cancer: A systematic review. *Head Neck.* 2017; 39: 2352-2362.
- Karanicolas PJ, Farrokhyar F, Bhandari M. Practical tips for surgical research: blinding: who, what, when, why, how? *Can J Surg.* 2010; 53: 345-8.
- Karlsson L. Chronic pain and exercises – studies on pain, biochemistry, adherence and attitudes. Linköping University medical dissertations No.1575. Linköping 2017.
- Karlsson P, Bergmark A. Compared with what? An analysis of control-group types in Cochrane and Campbell reviews of psychosocial treatment efficacy with substance use disorders. *Addiction.* 2015; 110: 420-8.
- Karolyi M. Beobachtungen über Pyorrhoea Alveolaris. *Oesterreich-Ungarische Vierteljahrsschrift für Zahnheilkunde* 1901; 17: 279-83.
- Keeney S, Hasson F, McKenna HP. A critical review of the Delphi technique as a research methodology for nursing. *Int J Nurs Stud.* 2001; 38: 195-200.
- Kirscht JP, Rosenstock IM. Patients' Problems in Following Recommendations of Health Experts. In : Stone GC, Cohen F, Adler N, eds. *Health Psychology.* San Fransisco: Jossey-Bass, 1979: 189-215.
- Kirveskari P, Jamsa T, Alanen P. Occlusal adjustment and the incidence of demand for temporomandibular disorder treatment. *J Prosthet Dent.* 1998; 79: 433-8.
- Kirveskari P, Jämsä T. Health risk from occlusal interferences in females. *Eur J Orthod.* 2009; 31: 490-5.
- Klasser GD, Greene CS. Oral appliances in the management of temporomandibular disorders. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; 107: 212-223.
- Klobas L, Axelsson S, Tegelberg A. Effect of therapeutic jaw exercise on temporomandibular disorders in individuals with chronic whiplash-associated disorders. *Acta Odontol Scand.* 2006; 64: 341-7.

- Koh H, Robinson PG. Occlusal adjustment for treating and preventing temporomandibular joint disorders. *J Oral Rehabil.* 2004; 31: 287-92.
- Kohno S, Matsuyama T, Medina R, Arai Y. Functional rhythmical coupling of head and mandibular movements. *J Oral Rehabil.* 2001; 28: 161–167.
- Kotiranta U, Suvinen T, Kauko T, Le Bell Y, Kemppainen P, Suni J, Forssell H. Subtyping patients with temporomandibular disorders in a primary health care setting on the basis of the research diagnostic criteria for temporomandibular disorders axis II pain-related disability: a step toward tailored treatment planning? *J Oral Facial Pain Headache.* 2015; 9: 26-34.
- Kraaijenga S, van der Molen L, van Tinteren H, Hilgers F, Smeele L. Treatment of myogenic temporomandibular disorder: a prospective randomized clinical trial, comparing a mechanical stretching device (TheraBite®) with standard physical therapy exercise. *J Craniomand Pract.* 2014; 32: 208-16.
- Kuttala M, Le Bell Y, Alanen P. The concepts prevalence, need for treatment, and prevention of temporomandibular disorders: a suggestion for terminology. *Acta Odontol Scand.* 1996; 54: 332-6.
- Kvale S, Brinkmann S. (2009). *InterViews: Learning the Craft of Qualitative Interviewing.* 2nd ed. Los Angeles: Sage Publications. P. 177-178.
- Könönen M, Nyström M. A longitudinal study of craniomandibular disorders in Finnish adolescents. *J Orofacial Pain.* 1993; 7: 329-36.
- Leadley RM, et al. Chronic diseases in the European Union: the prevalence and health cost implications of chronic pain. *J Pain Palliat Care Pharmacother.* 2012; 26: 310-25.
- Lerman SF, Rudich Z, Brill S, Shalev H, Shahar G. Longitudinal associations between depression, anxiety, pain, and pain-related disability in chronic pain patients. *Psychosom Med.* 2015; 77: 333-41.
- Lincoln YS, Guba EG. *Naturalistic Inquiry.* Beverly Hills, CA. Sage 1985. P. 289-332.
- Linde C, Isacson G, Jonsson BG. Outcome of 6-week treatment with transcutaneous electric nerve stimulation compared with splint on symptomatic temporomandibular joint disk displacement without reduction. *Acta Odontol Scand.* 1995; 53: 92-8.
- Lindfors E, Helkimo M, Magnusson T. Patients' adherence to hard acrylic interocclusal appliance treatment in general dental practice in Sweden. *Swed Dent J.* 2011; 35: 133-42.
- Lindfors E, Magnusson T, Tegelberg A. Interocclusal appliances - indications and clinical routines in general dental practice in Sweden. *Swed Dent J.* 2006; 30: 123-34.
- Lindh J. [Evaluating treatment effect]. *Läkartidningen.* 2013; 110: 159-62. In Swedish.
- Ling PH. *Gymnastikens Allmänna Grunder.* Uppsala. 1834.
- Lipton JA, Ship JA, Larach-Robinson D. Estimated prevalence and distribution of reported orofacial pain in the United States. *J Am Dent Assoc.* 1993; 124: 115-21.
- List T, Axelsson S. Management of TMD: Evidence from systematic reviews and meta-analyses. *J Oral Rehabil.* 2010; 37: 430–451.

- List T, Jensen RH. Temporomandibular disorders: Old ideas and new concepts. *Cephalalgia*. 2017; 37: 692-704.
- List T, Tegelberg Å, Lundeberg T, Orbach R. Smärtlindring i Ansikte och Huvud ur Bettfysiologiskt Perspektiv. Stockholm: Gothia, 1999. pp.76-81.
- Loeser JD, Treede RD. The Kyoto protocol of IASP Basic Pain Terminology. *Pain*. 2008; 137: 473-7.
- Look JO, John MT, Tai F, Huggins KH, Lenton PA, Truelove EL, Ohrbach R, Anderson GC, Shiffman EL. The Research Diagnostic Criteria For Temporomandibular Disorders. II: reliability of Axis I diagnoses and selected clinical measures. *J Orofac Pain*. 2010; 24: 25-34.
- Loorents V, Rosell J, Karlsson C, Lidbäck M, Hultman K, Börjeson S. Prophylactic training for the prevention of radiotherapy-induced trismus - a randomised study. *Acta Oncol*. 2014; 53: 530-8.
- Lorig KR, Holman H. Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med*. 2003; 26: 1-7.
- Lorig KR, Ritter P, Stewart AL, Sobel DS, Brown BW Jr, Bandura A, Gonzalez VM, Laurent DD, Holman HR. Chronic disease self-management program: 2-year health status and health care utilization outcomes. *Med Care*. 2001; 39: 1217-23.
- Loughlin KG, Moore LF. Using Delphi to achieve congruent objectives and activities in a pediatrics department. *J Med Educ*. 1979; 54: 101-6.
- Lundh H, Westesson PL, Kopp S. A three-year follow-up of patients with reciprocal temporomandibular joint clicking. *Oral Surg Oral Med Oral Pathol*. 1987; 63: 530-3.
- Lutz RW, Silbret M, Olshan N. Treatment outcome and compliance with therapeutic regimens: long-term follow-up of a multidisciplinary pain program. *Pain* 1983; 17:301-8.
- Lövgren A, Marklund S, Visscher CM, Lobbezoo F, Häggman-Henrikson B, Wänman A. Outcome of three screening questions for temporomandibular disorders (3Q/TMD) on clinical decision-making. *J Oral Rehabil*. 2017; 44: 573-579.
- Lövgren A, Visscher CM, Häggman-Henrikson B, Lobbezoo F, Marklund S, Wänman A. Validity of three screening questions (3Q/TMD) in relation to the DC/TMD. *J Oral Rehabil*. 2016; 43: 729-36.
- Macfarlane TV, Glenny AM, Worthington HV. Systematic review of population-based epidemiological studies of oro-facial pain. *J Dentistry*. 2001; 29: 451-467.
- Macfarlane TV, Kenealy P, Kingdon HA, et al. Orofacial pain in young adults and associated childhood and adulthood factors: Results of the population study, Wales, United Kingdom. *Commun Dent Oral Epidemiol*. 2009; 37: 438-450.
- Macfarlane TV, Kenealy P, Kingdon HA, et al. Twenty- year cohort study of health gain from orthodontic treatment: Temporomandibular disorders. *Am J Orthod Dentofacial Orthop*. 2009; 135: 692 e1-8. (discussion-3).
- Magnusson T. Mandibular dysfunction and recurrent headache. Thesis. 1981, University of Göteborg.

Magnusson T, Carlsson GE, Egermark I. Changes of clinical signs of craniomandibular disorders from the age of 15 to 25 years. *J Orofac Pain*. 1994; 8: 207-15.

Magnusson T, Syrén M. Therapeutic jaw exercises and interocclusal appliance therapy. A comparison between two common treatments of temporomandibular disorders. *Swed Dent J*. 1999; 23: 27-37.

Makino I, Arai YC, Aono S, et al. The effects of exercise therapy for the improvement of jaw movement and psychological intervention to reduce parafunctional activities on chronic pain in the craniocervical region. *Pain Pract*. 2014; 14: 413-8.

Malone DC, Abarca J, Hansten PD, et al. Identification of serious drug-drug interactions: results of the partnership to prevent drug-drug interactions. *Am J Geriatr Pharmacother*. 2005; 3: 65-76.

Maloney, GE, Mehta, N, Forgione, AG, Zawawi, KH, Al-Badawi, EA, Driscoll, SE. Effect of a passive jaw motion device on pain and range of motion in TMD patients not responding to flat plane intraoral appliances. *J Craniomandib Pract*. 2002; 20: 55-66.

Malterud K. Qualitative research: standards, challenges, and guidelines. *Lancet*. 2001; 358: 483-8.

Malterud K. Systematic text condensation: A strategy for qualitative analysis. *Scand J Public Health*. 2012; 40: 795-805.

Manchikanti L, Singh V, Datta S, Cohen SP, Hirsch JA. Comprehensive review of epidemiology, scope, and impact of spinal pain. *Pain Physician*. 2009; 12: 35-70.

Manfredini D, Landi N, Bandettini Di Poggio A, et al. A critical review on the importance of psychological factors in temporomandibular disorders. *Minerva Stomatol*. 2003; 52: 321-326.

Manfredini D, Lobbezoo F. Relationship between bruxism and temporomandibular disorders: A systematic review of literature from 1998 to 2008. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2010; 109: e26-50.

Mann AW, Pankey LD. Concepts of Occlusion: the Philosophy of Occlusion Rehabilitation. In: Courtade GL, editor. *Occlusal rehabilitation*. Philadelphia: W B Saunders 1963: 621-36.

Mannerkorpi K, Iversen MD. Physical exercise in fibromyalgia and related syndromes. *Best Pract Res Clin Rheumatol*. 2003; 17: 629-47.

Marchais-Roubelat A, Roubelat F. The Delphi method as a ritual: Inquiring the Delphic Oracle. *Technol Forecast Soc Change*. 2011; 78: 1491-1499.

Marchand, S. The physiology of pain mechanisms: from the periphery to the brain. *Rheum Dis Clin North Am*. 2008; 34: 285-309.

Marklund S, Häggman-Henrikson B, Wänman A. Risk factors associated with incidence and persistence of frequent headaches. *Acta Odontol Scand*. 2014; 72: 788-94.

Marklund S, Wänman A. Incidence and prevalence of temporomandibular joint pain and dysfunction. A one-year prospective study of university students. *Acta Odontol Scand*. 2007; 65: 119-27.

McCollum BB. Factors that make the mouth and teeth a vital organ (articulation orthodontia). *J Am Dent Assoc*. 1927; 14: 1261-71.

- McCormack HM, Horne DJ, Sheather S. Clinical applications of visual analogue scales: a critical review. *Psychol Med.* 1988; 18: 1007-19.
- McKenna HP. The Delphi technique: a worthwhile research approach for nursing? *J Adv Nurs.* 1994; 19: 1221-25.
- McLean SM, Burton M, Bradley L, Littlewood C. Interventions for enhancing adherence with physiotherapy: a systematic review. *Man Ther.* 2010; 15: 514-21.
- McNeill C. History and evolution of TMD concepts. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1997; 83: 51-60.
- Medlicott, MS, Harris, SR. A systematic review of the effectiveness of exercise, manual therapy, electrotherapy, relaxation training, and biofeedback in the management of temporomandibular disorder. *Phys Ther.* 2006; 86: 955-73.
- Melzack R, Wall PD. Pain mechanisms: a new theory. *Science.* 1965; 150: 971-9.
- Merskey H., Bogduk N. Classification of Chronic Pain, 2<sup>nd</sup> ed. Part III: Pain Terms, A Current List with Definitions and Notes on Usage, pp 209-214. IASP Task Force on Taxonomy. Seattle, WA: IASP Press: 1994.
- Michelotti A, De Wijer A, Steenks M, Farella M. Home exercises regimes for the management of non-specific temporomandibular disorders. *J Oral Rehabil.* 2005; 32: 779-85.
- Michelotti A, Iodice G, Vollaro S, Steenks MH, Farella M. Evaluation of the short-term effectiveness of education versus an occlusal splint for the treatment of myofascial pain of the jaw muscles. *J Am Dent Assoc.* 2012; 143: 47-53.
- Michelotti, A, Steenks, MH, Farella, M, Parisini, F, Cimino, R, Martina, R. The additional value of a home physical therapy regimen versus patient education only for the treatment of myofascial pain of the jaw muscles: short-term results of a randomized clinical trial. *J of Orofac Pain.* 2004; 18: 114-25.
- Minakuchi H, Kuboki T, Matsuka Y, Maekawa K, Yatani H, Yamashita A. Randomized controlled evaluation of non-surgical treatments for temporomandibular joint anterior disk displacement without reduction. *J Dent Res.* 2001; 80: 924-8.
- Moher D, Hopewell S, Schulz KF, Montori V, Gøtzsche PC, Devereaux PJ, Elbourne D, Egger M, Altman DG; CONSORT. CONSORT 2010 explanation and elaboration: updated guidelines for reporting parallel group randomised trials. *Int J Surg.* 2012; 10: 28-55.
- Moraes A da R, Sanches ML, Ribeiro EC, Guimarães AS. Therapeutic exercises for the control of temporomandibular disorders. *Dental Press J Orthod.* 2013; 18: 134-9.
- Møystad A, Krogstad BS, Larheim TA. Transcutaneous nerve stimulation in a group of patients with rheumatic disease involving the temporomandibular joint. *J Prosthet Dent* 1990; 64: 596-600.
- Mulet M, Decker KL, Look JO, Lenton PA, Schiffman EL. A randomized clinical trial assessing the efficacy of adding 6 x 6 exercises to self-care for the treatment of masticatory myofascial pain. *J Orofac Pain.* 2007; 21: 318-28.

Murphy MK, Black NA, Lamping DL, McKee CM, Sanderson CF, Askham J, Marteau T. Consensus development methods, and their use in clinical guideline development. *Health Technol Assess.* 1998; 2: 1-88.

National Institute of Dental and Craniofacial Research. Facial Pain. 2018. <https://www.nidcr.nih.gov/research/data-statistics/facial-pain>. Accessed April 30, 2018.

Naugle KM, Fillingim RB, Riley JL. A Meta-Analytic Review of the hypoalgesic effects of exercise. *The Journal of Pain.* 2012; 13: 1139-1150.

Naugle KM, Riley JL. Self-reported physical activity predicts pain inhibitory and facilitatory function. *Med Sci Sports Exerc.* 2014; 46: 622-9.

NHS. Continuing Professional Development: Quality in the New NHS. London: Department of Health. 1999.

Nicolakis P, Erdogmus CB, Kollmitzer J, Kersch-Schindl K, Sengstbratl M, Nuhr M, et al. Long-term outcome after treatment of temporomandibular joint osteoarthritis with exercise and manual therapy. *J Craniomand Pract.* 2002; 20: 23-7.

Nicolakis P, Erdogmus B, Kopf A, Ebenbichler G, Kollmitzer J, Piehslinger E, Fialka-Moser V. Effectiveness of exercise therapy in patients with internal derangement of the temporomandibular joint. *J Oral Rehabil.* 2001; 28: 1158-64.

Nicolakis P, Erdogmus B, Kopf A, Nicolakis M, Piehslinger E, Fialka-Moser V. Effectiveness of exercise therapy in patients with myofascial pain dysfunction syndrome. *J Oral Rehabil.* 2002; 29: 362-8.

Nicolakis P, Burak EC, Kollmitzer J, Kopf A, Piehslinger E, Wiesinger GF, Fialka-Moser V. An investigation of the effectiveness of exercise and manual therapy in treating symptoms of TMJ osteoarthritis. *J Craniomand Pract.* 2001; 19: 26-32.

Nicolakis P, Erdogmus B, Kopf A, Djaber-Ansari A, Piehslinger E, Fialka-Moser V. Exercise therapy for craniomandibular disorders. *Arch Phys Med Rehabil.* 2000; 81: 1137-42.

Nijs J, Kosek E, et al. Dysfunctional endogenous analgesia during exercise in patients with chronic pain: to exercise or not to exercise? *Pain Physician.* 2012; 15: ES205-13.

Nilsson IM, List T, Drangsholt M. Prevalence of temporomandibular pain and subsequent dental treatment in Swedish adolescents. *J Orofac Pain.* 2005; 19: 144-50.

Nilsson IM, List T, Drangsholt M. The reliability and validity of self-reported temporomandibular disorder pain in adolescents. *J Orofac Pain.* 2006; 20: 138-44.

Okeson JP. (editor). *Orofacial Pain: Guidelines for Assessment, Diagnosis & Management*, American Academy of Orofacial Pain, Quintessence Publishing (IL), 3rd Edition, 1996.

Ohrbach R, Fillingim RB, Mulkey F, et al. Clinical findings and pain symptoms as potential risk factors for chronic TMD: Descriptive data and empirically identified domains from the OPPERA case-control study. *J Pain.* 2011; 12: T27-45.

Ohrbach R, Larsson P, List T. The jaw functional limitation scale: development, reliability, and validity of 8-item and 20-item versions. *J Orofac Pain.* 2008; 22: 219-30.

Ohrbach R, Knibbe W. *Diagnostic Criteria for Temporomandibular Disorders: Scoring Manual for Self-Report Instruments*. Version 29 May 2016. [www.rdc-tmdinternational.org](http://www.rdc-tmdinternational.org) Accessed on 2 July 2018.



- Okifuji A, Ackerlind S. Behavioral medicine approaches to pain. *Anesthesiol Clin*. 2007; 25: 709-719.
- Onizawa K, Yoshida H. Longitudinal changes of symptoms of temporomandibular disorders in Japanese young adults. *J Orofac Pain*. 1996; 10: 151-6.
- Osborn M, Rodham K. Insights into Pain: A Review of Qualitative Research. *Rev Pain*. 2010; 4: 2-7.
- Paço M, Peleteiro B, Duarte J, Pinho T. The Effectiveness of Physiotherapy in the Management of Temporomandibular Disorders: A Systematic Review and Meta-analysis. *J Oral Facial Pain Headache*. 2016; 30: 210-20.
- Pauli N, Svensson U, Karlsson T, Finizia C. Exercise intervention for the treatment of trismus in head and neck cancer - a prospective two-year follow-up study. *Acta Oncol*. 2016; 55: 686-92.
- Peddareddygar LR, Oberoi K, Grewal RP. Congenital insensitivity to pain: a case report and review of the literature. *Case Rep Neurol Med*. 2014; 2014: 141953.
- Peterson M. Chronic Tennis Elbow: Aspects on Pathogenesis and Treatment in a Soft Tissue Pain Condition. Uppsala: Uppsala University; 2011.
- Phillips C, et al. Prioritising pain in policy making: The need for a whole systems perspective. *Health Policy*. 2008; 88: 166-175.
- Plesh O, Adams SH and Gansky SA. Temporomandibular joint and muscle disorder-type pain and comorbid pains in a national US sample. *J Orofac Pain*. 2011; 25: 190–198.
- Polit DF, Beck CT. Generalization in quantitative and qualitative research: myths and strategies. *Int J Nurs Stud*. 2010; 47: 1451-8.
- Progiante PS, Pattussi MP, Lawrence HP, Goya S, Grossi PK, Grossi ML. Prevalence of Temporomandibular Disorders in an Adult Brazilian Community Population Using the Research Diagnostic Criteria (Axes I and II) for Temporomandibular Disorders (The Maringá Study). *Int J Prosthodont*. 2015; 28: 600-9.
- Public Dental Health Service, Uppsala, Sweden. Database register.
- Quartana PJ, Buenaver LF, Edwards RR, et al. Pain catastrophizing and salivary cortisol responses to laboratory pain testing in temporomandibular disorder and healthy participants. *J Pain*. 2010; 11: 186–194.
- Rampakakis E, Ste-Marie PA, Sampalis JS, Karellis A, Shir Y, Fitzcharles MA. Real-life assessment of the validity of patient global impression of change in fibromyalgia. *RMD Open*. 2015 14;1(1).
- Rashid A, Matthews NS, Cowgill H. Physiotherapy in the management of disorders of the temporomandibular joint - perceived effectiveness and access to services: a national United Kingdom survey. *Br J Oral Maxillofac Surg*. 2013; 51: 52-7.
- Rees S, Williams A. Promoting and supporting self-management for adults living in the community with physical chronic illness: A systematic review of the effectiveness and meaningfulness of the patient-practitioner encounter. *JB Libr Syst Rev*. 2009; 7: 492-582.
- Reid KJ, et al. Epidemiology of chronic non-cancer pain in Europe: narrative review of prevalence, pain treatments and pain impact. *Curr Med Res Opin*. 2011; 27: 449-62.

Rigon M, Pereira LM, Bortoluzzi MC et al. Arthroscopy for temporomandibular disorders. The Cochrane database of systematic reviews. 2011; 5: CD006385.

Rinchuse DJ, Kandasamy S, Ackerman MB. Deconstructing evidence in orthodontics: making sense of systematic reviews, randomized clinical trials, and meta-analyses. *World J Orthod.* 2008; 9: 167-76.

Rollman GB, Gillespie JM. The role of psychosocial factors in temporomandibular disorders. *Curr Rev Pain.* 2000; 4: 71-81.

Roter DL, Hall JA, Merisca R, Nordstrom B, Cretin D, Svarstad B. Effectiveness of interventions to improve patient compliance: a meta-analysis. *Med Care.* 1998 ; 36: 1138-61.

Sackett DL. The Magnitude of Compliance and Noncompliance. In: Sackett DL, Haynes RB (eds): *Compliance with Therapeutic Regimens.* Baltimore, Johns Hopkins University Press. 1976.

Sagheer MA, Khan MF, Sharif S. Association between chronic low back pain, anxiety and depression in patients at a tertiary care centre. *J Pak Med Assoc.* 2013; 63: 688-90.

Sajedi H, Bas M. The evaluation of the aerobic exercise effects on pain tolerance. *Sport Science.* 2016; 9: 7-11.

Sakuma S, Yamaguchi Y, Taguchi N, Ogi N, Kurita K, Ito Y. Pilot study of the short-term effects of range-of-motion exercise for the temporomandibular joint in patients with temporomandibular joint disc displacement without reduction. *J Phys Ther Sci.* 2017; 29: 274-277.

Sale H, Isberg A. Delayed temporomandibular joint pain and dysfunction induced by whiplash trauma: a controlled prospective study. *J Am Dent Assoc.* 2007; 138: 1084–1091.

Sato S, Goto S, Nasu F, Motegi K. Natural course of disc displacement with reduction of the temporomandibular joint: changes in clinical signs and symptoms. *J Oral Maxillofac Surg.* 2003; 61: 32-4.

Scherpenhuizen A, van Waes AM, Janssen LM, Van Cann EM, Stegeman I. The effect of exercise therapy in head and neck cancer patients in the treatment of radiotherapy-induced trismus: A systematic review. *Oral Oncol.* 2015; 51: 745-50.

Schiffman EL, Look JO, Hodges JS, Swift JQ, Decker KL, Hathaway KM, Templeton RB, Friction JR. Randomized effectiveness study of four therapeutic strategies for TMJ closed lock. *J Dent Res.* 2007; 86: 58-63.

Schiffman E, Ohrbach R, Truelove E, Look J, Anderson G, Goulet JP et al. Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) for Clinical and Research Applications: recommendations of the International RDC/TMD Consortium Network\* and Orofacial Pain Special Interest Group. *J Oral Facial Pain Headache.* 2014; 28: 6-27.

Schmidt R.C. Managing Delphi surveys using nonparametric statistical techniques. *Decision Sciences.* 1997; 28: 763-774.

Schmitter M, Ohlmann B, John MT, Hirsch C, Rammelsberg P. Research diagnostic criteria for temporomandibular disorders: a calibration and reliability study. *J Craniomand Pract.* 2005; 23: 212-8.

- Schwartz L. Disorders of the Temporomandibular Joint. Philadelphia: W B Saunders, 1959: 28-43.
- Seligman DA, Pullinger AG. The role of functional occlusal relationships in temporomandibular disorders: a review. *J Craniomandib Disord.* 1991; 5: 265-79.
- Slade GD, Ohrbach R, Greenspan JD, et al. Painful temporomandibular disorder: Decade of discovery from OPPERA studies. *J Dent Res.* 2016; 95: 1084–1092.
- Sluijs EM, Kok GJ, van der Zee J. Correlates of exercise compliance in physical therapy. *Physical Therapy* 1993; 73: 771e82.
- Smith BH, Elliott AM. Active self-management of chronic pain in the community. *Pain.* 2005; 113: 249-250.
- Smith SB, Maixner DW, Greenspan JD, et al. Potential genetic risk factors for chronic TMD: Genetic associations from the OPPERA case control study. *J Pain.* 2011; 12: T92–101.
- Snaith RP. The Hospital Anxiety and Depression Scale. *Health Qual Life Outcomes.* 2003; 1: 29.
- Speciali JG, Dach F. Temporomandibular dysfunction and headache disorder. *Headache.* 2015; 55: 72-83.
- Stallard H. Functions of the occlusal surfaces of the teeth. *J Am Dent Assoc.* 1930; 13: 401.
- Stern AF. The hospital anxiety and depression scale. *Occup Med (Lond).* 2014; 64: 393-4.
- Story WP, Durham J, Al-Baghdadi M, et al. Self- management in temporomandibular disorders: A systematic review of behavioural components. *J Oral Rehabil.* 2016; 43: 929–936.
- Stuart CE. Articulation of human teeth. In: McCollum BB, Stuart CE, editors. A research report. South Pasadena (CA): Scientific Press, 1955.
- Sumsion T. The Delphi technique: an adaptive research tool. *British Journal of Occupational Therapy.* 1998; 61: 153 -156.
- Suslick J. Männen skippar norsk efterutbildning. *Tandläkartidningen.* 2013. <http://www.tandlakartidningen.se/arkivet/nyhet/mannen-skippar-norsk-efterutbildning/>
- Swedish Association of Local Authorities and Regions. Table 6 LP 2014.
- Swedish Council on Health Technology Assessment in Health Care (SBU). Assessment of Methods in Health Care - A handbook. 2018; 48-49.
- Swedish Council on Health Technology Assessment in Health Care (SBU). Methods of Treating Chronic Pain. Stockholm. 2006. SBU report no 177/1 (in Swedish): 22, 168.
- Tegelberg A, List T, Wahlund K, Wenneberg B. Temporomandibular disorders in children and adolescents: a survey of dentists' attitudes, routine and experience. *Swed Dent J.* 2001; 25: 119-27.
- Tegelberg A, Kopp S. Short-term effect of physical training on temporomandibular joint disorder in individuals with rheumatoid arthritis and ankylosing spondylitis. *Acta Odontol Scand.* 1988; 46: 49-56.

Tegelberg A, Kopp S. A 3-year follow-up of temporomandibular disorders in rheumatoid arthritis and ankylosing spondylitis. *Acta odontologica Scandinavica*. 1996; 54: 14-8.

The National Board of Health and Welfare. Bedömning av Tillgång och Efterfrågan på Personal i Hälso- och Sjukvård och Tandvård. Nationella planeringsstödet 2018.

The National Board of Health and Welfare. National guidelines for adult dental care. 2011. In Swedish. ISBN: 978-91-86885-09-0.

The National Board of Health and Welfare. Statistical database.

The Public Health Agency of Sweden. Facts on the national public health survey. <https://www.folkhalsomyndigheten.se/globalassets/statistik-uppfoljning/enkater-undersokningar/nationella-folkhalsoenkaten/fakta-nationella-folkhalsoenkaten-2016.pdf>

The Swedish Association of Public Dental Officers. Member register. 2015.

The Swedish National Board for Youth Affairs. Young people with attitude. The Swedish National Board for Youth Affairs – Attitude and value survey 2007;17. [https://www.mucf.se/sites/default/files/publikationer\\_uploads/unga-med-attityd.pdf](https://www.mucf.se/sites/default/files/publikationer_uploads/unga-med-attityd.pdf)

Thong ISK, Jensen MP, Miró J, Tan G. The validity of pain intensity measures: what do the NRS, VAS, VRS, and FPS-R measure? *Scand J Pain*. 2018; 18: 99-107.

Treede RD. The International Association for the Study of Pain definition of pain: as valid in 2018 as in 1979, but in need of regularly updated footnotes. *Pain Rep*. 2018; 3: e643.

Treede RD, Rief W, Barke A, et al. A classification of chronic pain for ICD-11. *Pain*. 2015; 156: 1003-1007.

Truelove E, Huggins KH, Mancl L, Dworkin SF. The efficacy of traditional, low-cost and nonsplint therapies for temporomandibular disorder: a randomized controlled trial. *J Am Dent Assoc*. 2006; 137: 1099-107.

Truelove E, Pan W, Look JO, Mancl LA, Ohrbach RK, Velly AM, Huggins KH, Lenton P, Shiffman EL. The Research Diagnostic Criteria for Temporomandibular Disorders. III: validity of Axis I diagnoses. *J Orofac Pain*. 2010; 24: 35-47.

Tuncer AB, Ergun N, Tuncer AH, Karahan S. Effectiveness of manual therapy and home physical therapy in patients with temporomandibular disorders: A randomized controlled trial. *J Bodyw Mov Ther*. 2013; 17: 302-8.

Turk DC, Dworkin RH, Allen RR, et al. Core outcome domains for chronic pain clinical trials: IMMPACT recommendations. *Pain*; 2003; 106: 337-45.

Turk DC, Wilson HD, Cahana A. Treatment of chronic non-cancer pain. *Lancet*. 2011; 377: 2226-35.

Van Hecke O, Torrance N, Smith BH. Chronic pain epidemiology and its clinical relevance. *British Journal of Anaesthesia*. 2013; 111: 13-18.

Velly AM, Gornitsky M, Philippe P. Contributing factors to chronic myofascial pain: a case-control study. *Pain*. 2003; 104: 491-9.

Visscher CM, Ohrbach R, van Wijk AJ, et al. The Tampa Scale for Kinesiophobia for temporomandibular disorders (TSK-TMD). *Pain* 2010; 150: 492–500.

- Von Korff M, Jensen MP, Karoly P. Assessing global pain severity by self-report in clinical and health services research. *Spine*. 2000; 25: 3140–51.
- Wessberg GA, Carroll WL, Dinham R, Wolford LM. Transcutaneous electrical stimulation as an adjunct in the management of myofascial pain-dysfunction syndrome. *J Prosthet Dent*. 1981; 45: 307-14.
- Wieckiewicz M, Zietek M, Smardz J, Zenczak-Wieckiewicz D, Grychowska N. Mental Status as a Common Factor for Masticatory Muscle Pain: A Systematic Review. *Front Psychol*. 2017; 9: 8:646.
- Wig AD, Aaron LA, Turner JA, Huggins KH, Truelove E. Short-term clinical outcomes and patient compliance with temporomandibular disorder treatment recommendations. *J Orofac Pain*. 2004; 18: 203-213.
- Wright EF, Clark EG, Paunovich ED, Hart RG. Headache improvement through TMD stabilization appliance and self-management therapies. *J Craniomand Pract*. 2006; 24: 104-11.
- Wänman A. Craniomandibular disorders in adolescents. A longitudinal study in an urban Swedish population. *Swed Dent J Suppl*. 1987; 44: 1-61.
- Wänman A, Ernberg M and List T. Guidelines in the management of orofacial pain/TMD: An evidence- based approach. *Tandlaegebladet*. 2016; 11: 828–838.
- Yoda T, Sakamoto I, Imai H, Honma Y, Shinjo Y, Takano A, et al. A randomized controlled trial of therapeutic exercise for clicking due to disk anterior displacement with reduction in the temporomandibular joint. *J Craniomandib Pract*. 2003; 21: 10-6.
- Younger J, McCue R, Mackey S. Pain outcomes: a brief review of instruments and techniques. *Curr Pain Headache Rep*. 2009; 13: 39-43.
- Yuasa, H, Kurita, K; Treatment Group on Temporomandibular Disorders. Randomized clinical trial of primary treatment for temporomandibular joint disk displacement without reduction and without osseous changes: a combination of NSAIDs and mouth-opening exercise versus no treatment. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2001; 91: 671-5.
- Zigmond AS, Snaith RP. The Hospital Anxiety and Depression Scale. *Acta Psychiatr Scand*. 1983; 67: 361–370.