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**ORIGINAL ARTICLE**

# A two-year follow-up study of temporomandibular disorders in a female Sami population: validation of cases and controls as predicted by questionnaire

CHRISTINA STORM & ANDERS WÄNMAN

*Department of Odontology, Clinical Oral Physiology, Faculty of Medicine, Umeå University, Umeå, Sweden*

**Abstract**

***Objective.*** The first aim of this study was to validate persistent, severe symptoms of temporomandibular disorders (TMD) among Sami females, as predicted by questionnaire. The second aim was to establish diagnoses according to the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) Axis 1 among predicted cases and controls. The third aim was to compare subjects with severe TMD to controls in regard to dental occlusion, general health, and parafunctions. ***Material and Methods.*** The subjects, Sami females living in the Arctic region of northern Sweden, all with long-standing (≥1 year), intense (≥5 on NS), and frequent (≥once a week) symptoms of pain and dysfunction in the jaw–face region, were invited for clinical examination; 22 (63%) agreed to participate. Forty-six subjects with no symptoms in the jaw–face region were matched to these cases in accordance with five age groups. The examiner was blind to subject affiliation. ***Results.*** The positive predictive value of presenting with signs and symptoms of TMD at follow-up was 0.82; the negative value was 0.87. Cases reported impaired general health and awareness of parafunctions significantly more frequently than did controls. Registered dental occlusion factors did not distinguish cases from controls. ***Conclusions.*** Long-standing, intense, and frequent TMD symptoms remained essentially unchanged over the 2-year follow-up of females in a Sami population. Presence of severe TMD was related to impaired general health and awareness of oral parafunctions.

**Key Words:** *Females, pain, prospective, Sami, temporomandibular*

## Introduction

Over the past three to four decades, several population-based studies of the presence of pain and functional disturbance in the jaw–face region have been carried out [1–5]. Among the first in this field to apply epidemiology were Helkimo et al. [6] in a study of Sami populations around Lake Enare in northern Finland. The high observed prevalence of these conditions, together with the predominantly fluctuating pattern of their symptoms, has called into question the validity of epidemiological findings regarding relevant treatment needs and approaches [7,8]. From the clinical perspective, females are the dominant sex seeking care for these disturbances, though the reasons for this globally found pattern are not completely understood. The various qualities of the symptoms, such as frequency, intensity, and duration, seem to be factors of some importance [9,10].

It is essential not to rely solely on patient samples when examining the associated and contributing factors of a certain disease, because various elements may influence a patient's decision to seek health care. Therefore, population-based studies are needed, not only to determine the prevalence or incidence of a certain condition in a population, but also to explore possible co-morbidities and correlations. Surveying by means of telephone interviews or mailed questionnaires can be associated with some validity problems. Do the questions accurately measure or capture what we expect to measure? What does an affirmative answer to a question about the presence of pain in the jaws or face really mean? A recent study of adolescents [11] demonstrated that the predictive value of such a question in relation to subsequently establishing a temporomandibular disorder (TMD) diagnosis was only 21%; the finding may be a consequence of a pattern of predominantly occasional short-term pains [12,13]. It is therefore

Correspondence: Anders Wänman, Department of Odontology/Clinical Oral Physiology, Faculty of Medicine, Umeå University, Umeå, Sweden. Tel: +46 90 7856082. Fax: +46 90 13 25 78. E-mail: anders.wanman@odont.umu.se

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important to validate the outcome of any questionnaire used in screening possible TMD patients. We have previously conducted an epidemiological study of Sami females living in the Arctic region of northern Sweden [14]. The aim was to explore prevalence and co-morbidities of long-standing, frequent, and intense TMD symptoms to improve our understanding of their impact in the female Sami population, 8% of whom presented an intense level of frequent pain and dysfunction in the jaw–face region.

The first aim of this study was to analyze the validity of reported long-standing, intense, and frequent symptoms of pain and dysfunction in the jaw–face region with regard to signs and symptoms of temporomandibular disorders at a 2-year follow-up. The second aim was to establish a diagnosis according to the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) Axis 1 among predicted cases and controls. The third aim was to compare subjects with severe TMD to controls with regard to dental occlusion, general health, and parafunctions.

## Material and methods

### *Study population*

The study population derived from a sample of all 21 to 70-year-old females living in the Arctic region of Sweden and included in the 1997 electoral register of the Swedish Sami Parliament or registered as reindeer owners or herders with the Swedish Board of Agriculture. In a previous study, we reported on the prevalence of pain and dysfunction in the jaw–face and head–neck regions in this study population [14]. Based on questionnaire answers, a sample of "cases" and "controls" was selected and invited for a follow-up and clinical examination 2 years after the primary investigation. Those who, at the start, were 21–70 years old, considered themselves Sami, and reported recurrent or consistent symptoms of TMD occurring at least once a week, with an intensity of 5 or more on an 11-point numerical scale (NS) and for a duration of at least 1 year were identified as "cases". A total of 35 females met the inclusion criteria and were selected for clinical examination, free of charge, at a dental clinic in Karesuando, Kiruna, or Gällivare; expenses for travel to and from the clinic were covered. Between the prior [14] and present studies, 10 females had moved, permanently or temporarily, from the region, 6 were not interested in participating, and 1 was severely ill. A total of 18 individuals accepted the offer to participate. In the oldest age group (63–72 years of age), only one subject met the inclusion criteria. To allow this age group to be represented, all those in this age group who reported frequent TMD symptoms of more than 1 year's duration were invited ($n=5$), 4 of whom agreed to participate. Of these, 1 stated the intensity level on the NS to be 8, 2 stated 3, and 1 reported daily pains and feelings of tiredness in the jaws of over 5 years' duration, but did not specify any intensity level. The total cases thus comprised 22 females.

The inclusion criteria for being selected as a "control" were that subjects should consider themselves Sami, have no TMD symptoms, and have no pain in the cervical region. A total of 127 females met these inclusion criteria; these subjects were stratified into 5 age strata based on those of the primary investigation (i.e. 21–30, 31–40, 41–50, 51–60, and 61–70 years old). All subjects in the oldest ($n=28$) and youngest age groups ($n=16$) were selected for clinical examination. In the former sample, 7 did not want to be examined, 4 could not be reached or traced, 3 could not travel to the dental clinic, 1 had recently had a stroke causing complications in locomotion, and 1 was away from home, leaving 12 who agreed to participate. In the youngest sample, 7 could not be reached or traced and another 5 had moved, leaving 4 who agreed to participate. In the other age groups, 16 subjects in each 10-year cluster were randomly selected for examination. Ten subjects could not be reached or traced, 4 did not want to participate, 2 had moved, 1 could not travel to the clinic, and 1 was deceased; a total of 30 subjects thus accepted to participate. The controls comprised 46 females.

### *Methods*

Case histories were collected using the same questionnaire as had been filled out in the previous study [14]. The examiner was blind to both the questionnaire and whether subject affiliation was "control" or "case". The clinical examination included:

1. Number of teeth.
2. Intermaxillary relationships.
   Sagittal: neutro-, dist-, or mesioclusion.
   Frontal: open bite, edge-to-edge bite, normal vertical overbite, and deep bite ($\geq 5$ mm).
   Transversal: cross-bite, scissors-bite, and normal bite.
3. Overjet and overbite to nearest millimeter.
4. Contact pattern in centric relation (unilateral or bilateral).
5. Slide in centric in anterior–posterior, vertical, and lateral directions to the nearest 0.5 mm (0, 0.5, 1.0, 1.5, etc.).
6. The following contact patterns at 3 mm and 9 mm lateral excursion of the mandible were registered: anterior guidance, cuspid guidance, group function, balanced occlusion, laterotrusive side interference, and mediotrusive side interference.

7. Number of occluding pair of teeth at light and hard clenching effort.
8. Supporting occlusal zones in accordance with Eichner, without removable dentures.
9. Presence of implant-supported fixed dental prostheses.
10. Degree of tooth wear in accordance with Oilo et al. [15].
11. Mandibular mobility. Unassisted maximal opening, protrusive capacity, and laterotrusive capacity were measured to the nearest millimeter with a ruler.
12. Temporomandibular joint (TMJ) sounds. Auscultation, without a stethoscope, was used to detect the presence of TMJ sounds on opening and closing the jaw; the sounds were classified as sharp clicking, dull clicking, or crepitating.
13. TMJ tenderness. The TMJ was palpated laterally and posteriorly through the auditory meatus; tenderness/pain was registered only if palpation elicited a palpebral reflex in the eye or a protective reflex (withdrawal).
14. TMJ pain on free movement (opening wide, laterotrusion, and protrusion).
15. TMJ loading. The subject was asked to bite hard for 30 s on a double wooden spatula (3 mm) placed in the region of the first molars, each side separately; elicited pain in the contralateral side of the joint was registered as TMJ load pain.
16. TMJ joint play. The examiner's first finger on the right hand was placed on the subject's left molar region; while holding the mandible, the jaw was pulled forwards and backwards with the operator's third finger on the left hand placed over the left TMJ. TMJ movement was registered if restricted, painful, or both. The procedure was repeated on the other side.
17. Muscle tenderness. Presence of tenderness/pain to palpation of muscle sites was registered in the same way as for TMJ. The following sites were palpated: region of the lateral pterygoid muscles, medial pterygoid muscles, anterior and posterior parts of temporal muscles, tendon of the temporal muscles, superficial and deep parts of masseter muscles, sternocleidomastoid muscles, trapezius muscles, neck muscles in the region of the linea nuccae, and the thumb muscles. If the last elicited a pain response, the muscles on the underside of the forearms and the calf muscles were also palpated to measure signs of generalized pain response to palpation.
18. Loading of jaw muscles. The subject was asked to clench her teeth hard in the intercuspal position for 30 s; development of fatigue or pain in the head, face, or jaws during clenching was registered.
19. The perception over the areas innervated by the trigeminal nerve was tested on both sides using von Frey hairs; the following areas were tested on the right and left sides: the forehead, infraorbital region, cheek, mental region, and temple.
20. Weight, height, and calculated body mass index (BMI).

Some of the variables captured in the questionnaire and some registered clinically were used to calculate the anamnestic (Ai) and clinical (Di) dysfunction indexes as formulated by Helkimo [16]. Each subject was classified in accordance with RDC/TMD Axis I [17].

### *Statistical methods*

Data analysis was done using SPSS statistical software v.14.0 (Statistical Package for Social Science, SPSS Inc., Ill., USA). Comparisons of cases and controls with regard to categorical variables were done using the chi-square test or Fisher's exact test if the expected counts were $<5$. Comparisons between groups with regard to continuous variables were done using the *t*-test for independent samples. Comparisons of registrations within cases and controls at the first and second examinations were done using Wilcoxon's signed matched test. Associations were assessed by means of odds ratios (OR) in unconditional logistic regression models. A $p$-value $<0.05$ was considered statistically significant.

## Results

The study population had a mean age of 48.7 (SD 13.1) with no statistically significant difference between cases and controls. The positive predictive value of presenting with signs and frequent symptoms of TMD at follow-up was 0.82; the negative predictive value was 0.87. The calculated annual incidence rate of TMD among controls was 6.5%. There was no statistically significant difference in reported frequency or intensity of symptoms on the NS among the cases between the first and second examinations.

Two subjects, one in each group, were edentulous. The mean number of teeth was 25.2 (SD 7.2) with no statistically significant difference between cases and controls. No statistically significant difference was found between cases and controls for morphologic occlusion (neutroclusion 72%, distoclusion 27%, mesioclusion 1.5%, deep bite 16%, open bite 3%, edge-to-edge bite 8%, cross bite 8%, and scissors bite 3%), contact patterns in centric relation (unilateral contact in 57%), or slide in centric or contact patterns in eccentric positions. Cases had fewer contacting pairs of teeth in centric occlusion (mean 8.2, SD 2.9) than did controls (mean 9.8,

Table I. Conditional regression analysis of factors related to being classified as case or control

| | Prevalence: cases/controls | Crude OR | 95% CI | *p*-value |
|---|---|---|---|---|
| Education: senior high school or university | 0.67/0.67 | 0.97 | 0.3–2.9 | 0.95 |
| Work outside home | 0.55/0.61 | 1.30 | 0.5–3.6 | 0.62 |
| Disease under medical care | 0.41/0.24 | 2.14 | 0.7–6.4 | 0.17 |
| Impaired general state of health | 0.43/0.02 | 29.6 | 3.4–257 | 0.002 |
| On prescribed medication | 0.50/0.24 | 3.09 | 1.0–9.4 | 0.046 |
| Awareness of grinding teeth | 0.44/0.37 | 1.35 | 0.4–4.1 | 0.60 |
| Awareness of clenching teeth | 0.67/0.23 | 6.60 | 2.0–22.1 | 0.002 |
| Awareness on pressing tongue | 0.33/0.09 | 4.88 | 1.1–21.6 | 0.037 |

SD 2.8) ($p=0.034$). There was no significant difference between cases and controls in terms of the distribution of supporting occlusal zones according to the Eichner index.

No statistically significant difference was found between cases and controls for mandibular mobility. The mean maximal jaw opening capacity was 48.4 mm (SD 6.5), the mean laterotrusion to the right side was 9.1 mm (SD 2.5), the mean laterotrusion to the left side was 9.0 mm (SD 2.7), and the mean maximal protrusion was 8.7 mm (SD 2.2). Temporomandibular joint sounds were registered in 50% of cases and 9% of controls ($p<0.001$). Tenderness to palpation over the TMJ (lateral and/or posterior) was noted in 4.5% of cases and 2.2% of controls, with no statistically significant difference between the two groups. Tenderness to palpation over jaw muscle sites was registered in 68% of cases and 20% of controls ($p<0.001$). Forty-five percent of cases developed symptoms during a 30 s clenching task compared to 15% of controls ($p=0.007$). Pain was only elicited from the TMJ region in 26% of cases ($p=0.002$). All those included in the study population had normal perception at all tested regions of the face, as determined using von Frey hairs.

The cases had a statistically significantly ($p=0.027$) higher body mass index (28.2) than did the controls (25.4). Cases had significantly higher odds ratios for reporting impaired general health, taking prescribed medication, and being aware of teeth clenching and tongue pressing (Table I).

The distribution of symptoms (Ai) and clinical signs (Di) of dysfunction is presented in Figure 1. Cases reported frequent TMD symptoms significantly more often ($p<0.001$) than did controls. In total, 91% of cases displayed signs of dysfunction compared to 37% of controls. A diagnosis in accordance with the RDC/TMD classification system was established in 59% of cases. Symptomatic TMJ disk displacement was diagnosed in 46% of cases compared to in 7% of the controls ($p<0.001$). Myofascial pain was diagnosed in 40% of cases as compared to in 2% of controls ($p<0.001$). Diagnoses of TMJ arthritis and TMJ arthrosis were established in 5% and 9%, respectively, of cases, but in no controls; the difference between cases and controls was not statistically significant.

## Discussion

The main findings of this case-control study of a Sami female population were that levels of symptom severity among cases were maintained at the 2-year follow-up and that registered dental occlusion factors did not distinguish cases from controls.

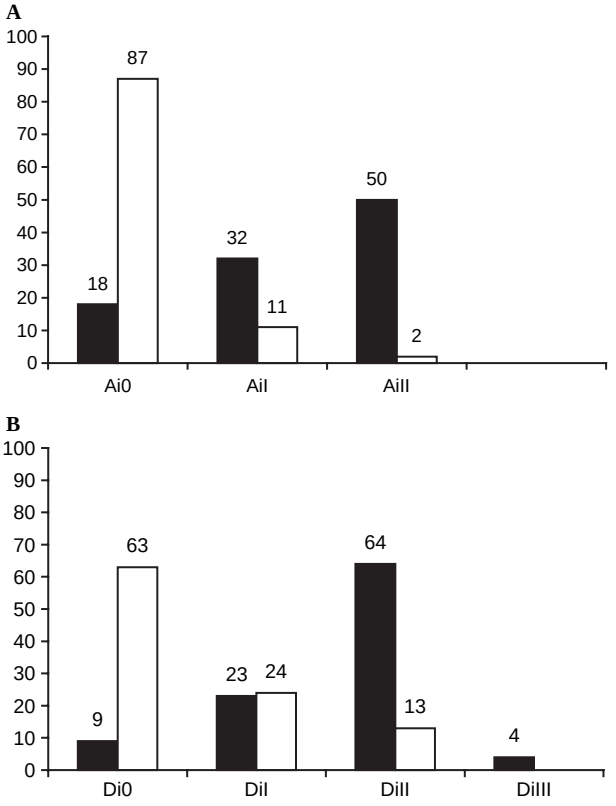


Figure 1. Percentage distribution of symptoms (A) and signs (B) at a 2-year follow-up of TMD among cases (filled bars) and controls (open bars) as identified by questionnaire in a female Sami population in northern Sweden. A. Ai 0 denotes no symptoms, Ai I denotes mild symptoms, and Ai II denotes severe symptoms of dysfunction; the anamnestic dysfunction index was modified in that only symptoms reported at least once a week were included. B. Di 0 denotes no signs, Di I denotes mild signs, Di II denotes moderate signs, and Di III severe signs of dysfunction, in accordance with Helkimo's clinical dysfunction index.

Published longitudinal studies of TMD have demonstrated a fairly consistent pattern of predominantly transient symptoms of pain and dysfunction with time [12,13,18,19]. In a 10-year prospective study, however, females were found to be at a higher risk of reporting long-standing symptoms, even though significant fluctuations in TMD symptoms occurred over the period [20]. This finding was also one of the background reasons for studying this fairly well-defined sample of Sami females. From the clinical perspective, females outnumber males as patients to a greater degree than would be expected from population-based studies. There may be several reasons for this pattern: females may pay more attention to their health, have more sensitive perceptions of the symptoms in question, feel higher levels of disability and impact on social life, and experience the symptoms for longer and at higher intensity levels than males do. Factors related to the female sex or gender may predispose or increase the vulnerability of women to developing long-standing pain and dysfunction in general, as well as in the jaw–face region, which has powerful proprio- and nociception. Polymorphism of catecholamine-O-methyl-transferase [21] and serotonin [22], as well as differences in the haplotypes of β receptors [23], has recently been found to be significantly related to the development of TMD pain. These indications, together with the role of sex steroids and psychosocial factors, have merged to form a recently presented hypothesis of biopsychosocial mechanisms and increased vulnerability of certain subjects to developing pain and dysfunction in the jaw–face region [24]. This hypothesis was supported by a clinical short-term double-blind study in which TMD patients previously treated with positive outcomes and controls received artificial interference in the occlusion. The results indicated that the former sample were more vulnerable to developing symptoms than controls were [25]. Factors that induce increased physical loading on the tissues involved may thus be individually significant and related to the subject's reaction patterns and capacities [26].

The present study has a weakness, namely, the high drop-out rate and resulting fairly low number of cases. Nothing indicates, however, that the drop-out was related to changes in TMD symptoms. The main reason why certain subjects did not participate was that they could not be reached due to changes in place of residence. There were no statistically significant differences in answers to the first questionnaire between the participating subjects and those who were not accessible. The study also has several strengths. Both cases and controls were drawn from the same population and cases were identified based on severity of symptoms and not on status as consulting patients or by referral. Twice as many controls as cases were included to strengthen the study. In the selection procedure, cases and controls were matched based on age group, sex, culture, and ethnicity. The selection also focused on the extremes at each end of a normal distribution with regard to severity of symptoms in the jaw–face region. The small number of participants, however, increases the risk of making a type-2 error in data interpretation, and accepting a null hypothesis even though it is false. The distributions of most clinical variables examined in the cases and controls were so similar that they would hardly produce statistically significant results even with a significant increase in the number of cases and controls. We are confident that the patterns identified from this sample, although fairly small, are clinically relevant. The examiner (AW) was an experienced specialist in the field of clinical oral physiology and throughout the study was kept blind to subject affiliation. The 2 years that had passed between the first and second examinations meant that the cases really could be defined as severe cases with persistent TMD symptoms. Only those who considered themselves as Sami were included; this was because subsequent thematic interviews were planned, in which one theme was the subjects' lived experience of being Sami females with or without TMD.

One major difference between cases and controls with regard to clinically registered variables was the pain elicited when loading the muscles and the TMJ. One of the first studies to find differences between cases and controls with regard to submaximal clenching was that of Clark et al. [27]. The finding of pain reactions on TMJ loading may indicate that the prevalence of arthralgia was higher than indicated by the RDC/TMD classification schema, in which tenderness to palpation over the joint area is an obligate criterion. The cases also presented a significantly higher proportion of subjects with pain reactions to palpation. These signs are often interpreted as indicating myofascial involvement, although recent research indicates that they should be inferred as indicating a reduced pain threshold to pressure [28]. The significantly higher BMI of the cases may indicate that obesity is related to TMD in females. High levels of estrogen have been related both to higher BMI values [29] and to TMD pain [30]. The link may thus relate to female hormones, physical activity levels, or both, and warrants more research. The present study supplements and confirms the results of previous studies [31,32] demonstrating that subjects with TMD consider their general health status as more impaired than that of controls. The almost 30-times-higher likelihood of reporting impaired health was interesting, since the proportion of those under medical care due to disease did not differ significantly between cases and controls. This result should not be interpreted as indicating direct correlation between the experience of symptoms in the jaw–face region and impaired general health. There may be several confounders of

the results, such as co-morbid pain in other locations with related sleep disturbances [33] and psychological distress [34].

We used von Frey monofilaments to test the tactile thresholds in the trigeminal innervated regions. Persistent pain in the jaw–face region may involve a focal neuropathic pain disorder [35]. Idiopathic pain conditions in the face can be both severe and difficult to manage. It has been hypothesized that neuropathic pain disorders may be related to changed perceptions of tactile stimulation [36]. We had hypothesized that some of these females might display deviating tactile perception, but all responded uniformly to the test in the clinical setting. In the clinic, we have noted that among subjects with severe idiopathic facial pain conditions, signs of both hypoesthesia and dysesthesia of the trigeminal nerve in the painful region can be registered as compared to the non-painful side. This female Sami population of approximately 500 subjects may thus have been too small for such cases to be captured, the prevalence of which may therefore be counted per thousand. The various dental occlusal factors measured did not reveal any major differences between cases and controls. The cause of these long-standing, frequent and severe symptoms in the jaw–face region are thus not likely to be based on different aspects of occluding pairs of teeth. The present results are in line with those of most previous epidemiological studies which demonstrate that a similar variability of dental occlusal factors exists among subjects with and without TMD. The present results also relate to previously found associations between oral parafunctions and TMD [19,32,37], indicating that these jaw motor behavior patterns, especially tooth clenching and tongue pressing, are significant in patients with TMD.

In conclusion, the present study demonstrated that long-standing, severe and frequent TMD symptoms remained essentially unchanged over the 2-year follow-up of females in the Sami population. Few factors examined in this quantitative study were significantly related to the condition of whether or not a subject was a TMD case. Females with severe symptoms of TMD perceived an impaired general state of health and were aware that their jaws were often strained. A subsequent thematic interview of a strategically chosen sub-sample may yield new insight into how these females express, understand, and handle disability in their lived experience of having long-standing severe TMD.

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