Long-term treatment outcome for adolescents with temporomandibular pain

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Abstract

Objectives: This study aims to evaluate long-term, self-perceived outcome in adulthood for individuals treated as adolescents for temporomandibular disorder (TMD) pain in two previous randomized controlled trials (RCTs). ***Materials and methods:*** The study included 116 subjects (81% females) treated for frequent TMD pain in two separate RCTs 5–21 (M=14.8, SD=4.9) years previously. Treatment consisted of occlusal appliance (OA) (n=41, 35.3%) or relaxation training (RT) combined with information for the control (Co) group (n=50, 43.1%), both compared to non-responders receiving additional, sequential treatment (ST) in a crossover study (n=25, 21.6%). Participants answered a questionnaire on their experience of frequency and intensity of TMD pain, impaired chewing capacity and daily social activities, help-seeking behavior and treatment, general health, other pain, and depressive symptoms. ***Results:*** Older participants reported lower levels of frequency and intensity of TMD pain, impairment, and depressive symptoms, as well as better general health. Females reported more frequent and more intense TMD pain, and greater impairment, and more often reported "other pain" compared to males. Non-responders receiving ST experienced significantly more TMD and other pain, and higher impairment levels compared to other groups. Those treated with an OA had sought additional treatment significantly less often since the RCTs than ST and RT/Co-treated individuals. ***Conclusion:*** Adolescents treated with OA showed somewhat better sustained improvement over the extended follow-up period than those treated with RT/Co. Non-responders to treatment and females exhibited a poorer outcome. These groups need particular attention and extended or different treatments to achieve a better long-term outcome.

Keywords: Long-term follow-up, adolescence, temporomandibular disorders, occlusal appliance, relaxation training.

Introduction

Temporomandibular disorders (TMDs) are a common cause of chronic orofacial pain comprising pain and dysfunction of the masticatory muscles, temporomandibular joints, and related structures. The overall prevalence rates in the general population of adolescents vary from 3% to 7% [1–3], increasing to an estimated 10–15% in adults [4]. Reports also indicate that TMD pain symptoms among adults may begin during adolescence [5]. While for both genders, the symptoms increase during adolescence and into adulthood, they are more pronounced in females. In a large, cross-sectional, general population-based survey [6], TMD prevalence was also found to vary over the lifetime.

Research evidence has shown comorbidity between TMD pain and other bodily pain conditions, such as tension-type headache or pain in the neck, shoulder, and back [7–12]. Temporomandibular disorder pain conditions are also associated with the experience of stress, anxiety, and depression and other somatic complaints [8, 12–15], as well as impaired normal daily activities [16], and have a negative impact on the quality of life [17].

In a few studies, the course of TMD symptoms in children and adolescents has been investigated longitudinally in both community [18–21] and clinical samples [22]. In an 8-year follow-up study of adolescents, the prevalence of TMD and psychosomatic symptoms was found to be low both during adolescence and in young adulthood [22]. In a 10-year prospective survey of young adults, persistent TMD symptoms were more commonly reported over time in women than in men [18]. Magnusson and collaborators [21], who followed a randomly selected sample over a 20-year period from childhood into adulthood, noted that progression to severe pain and dysfunction, as well as spontaneous recovery from more pronounced symptoms, was rare. In a 5-year follow-up of a mixed sample of community and clinical cases with TMD, Rammelsberg et al. [23] found that 31% of the patients continued to suffer from the condition, while 36% had recurrent TMD pain and 33% were in remission.

The need for systematic treatment is reflected by the fact that a substantial number of untreated TMD patients do not improve spontaneously over time [24]. In general population-based surveys, more than 50% of adolescents reporting frequent TMD pain had a perceived need for professional help [1, 25]. A qualitative study of adolescents who experienced TMD pain reports that, besides seeking professional help, they also reported different strategies to relax and manage the jaws [26]. In a meta-analysis of epidemiological studies, the need for TMD treatment in adults has been found to be approximately 16% [27]. Among predictors, pain intensity and fear of jaw movements influence the decision to seek care, while catastrophizing and the use of pain medication seem to be implicated in continuing help-seeking behavior [28].

Previous treatment outcome studies of adults have shown that different treatment approaches including the use of occlusal appliances (OAs) and psychological interventions, separately or in combination, produce positive short-term treatment effects on TMD-related pain and functional jaw impairment [29–32]. Long-lasting improvement of TMD pain after treatment has also been reported in the majority of adult patients [33–36]. In a large, epidemiological survey [2], about one-third of adolescents with TMD symptoms had received treatment in dental clinics and the most commonly administered treatment was the use of OAs and information. In two previous RCTs conducted by this group in the same recruitment area and population as in this study, we evaluated the short-term effects of OAs, relaxation training (RT), and both combined with information [37, 38], compared to information alone, in adolescents suffering from frequent TMD [37]. The overall short-term results showed that adolescents with TMD pain had achieved significantly better pain relief after treatment with OA compared to RT, both combined with information [37, 38], or versus information alone [37].

The purpose of the present study was to evaluate self-perceived long-term outcome in adulthood for adolescents treated for frequent TMD pain in two previous RCTs, with regard to: (1) frequency and intensity of TMD pain by gender, age (RCT number), and treatment condition; (2) impairment in chewing capacity, daily and leisure time activities, and at work; and (3) help-seeking behavior and treatment, general health, and experience of other pain and depressive symptoms.

Insert table 1 about here

Materials and methods

Subjects

In our two previous RCTs, 186 adolescents were recruited from a consecutive series of patients referred to the Department of Stomatognathic Physiology, Public Dental Health Service in two Swedish cities, Linköping and Norrköping. The first sample (RCT 1) included 122 subjects (93 girls and 29 boys) recruited between 1996 and 2000 [37]; the second sample (RCT 2) consisted of 64 subjects (61 girls and three boys) participating, between 2003 and 2011, in a trial with a crossover design [38]. A total of 19 patients (10.2%), twelve in RCT 1 and seven in RCT 2 before the crossover phase, dropped out during treatment, leaving 167 subjects to be invited to participate in the present long-term follow-up study, of whom another 51 patients (30.5%) did not respond. The distribution of patients by treatment group in the two RCTs, and group formation in the long-term follow-up is presented in Figure 1. The average follow-up time was 14.8 years (SD=4.9), with a range of 5–21 years.

Insert figure 1 about here

The following treatment inclusion criteria were used: (1) age 12–19 years; (2) a report of pain at least once a week in the face, jaws, TMJs, or temples for at least 3 months; (3) patients

diagnosed according to the Research Diagnostic Criteria for TMD (RDC/TMD) [39]; and (4) wanting treatment. Excluded were subjects with migraine, patients with ongoing orthodontic treatment interfering with an OA, and those with juvenile idiopathic arthritis.

A clinical examination was performed in accordance with RDC/TMD examination guidelines, in which pain site, mandibular movement capacity (mm), and associated pain, presence of joint sounds, and palpatory pain of the temporomandibular muscles and joints were assessed. This procedure allows establishment of the following multiple diagnoses: myofascial pain, disc displacement, and/or arthralgia/arthrosis [39].

Treatment

In RCT 1, patients were randomly assigned to receive treatment with either an OA or four therapist-guided sessions of RT, both combined with information; or information only given in one session as a control treatment (Co) [37]. The second trial (RCT 2) included two phases: In the first phase, patients were randomized to either OA treatment or RT administered during eight therapist-assisted sessions; both groups also received information [38]. This trial included a crossover design in which, in the second phase, non-responders to treatment after phase 1 (those reporting a treatment effect on the Patient Global Impression of Change (PGIC) scale [40, 41] as “Slightly improved;” “No change;” “Slightly worse;” or “Much worse”) were offered the other treatment type, i.e. occlusal appliance or relaxation training (sequential treatment (ST)) (see Figure 1).

Trained and experienced therapists (two dentists and one nurse) administered the treatments. The Co group received standardized information about TMD-related anatomy, TMD pain epidemiology, parafunction, and stress. The OA consisted of a stabilization splint placed in the upper jaw, designed to produce maximum occlusion contact with canine

guidance. These patients were asked to use the splint every night up to the first post-treatment evaluation (at 3 months) and, if needed, to continue its use until the 6-month follow-up.

Relaxation therapy included clinic-based training and a manual for home training with taped audio instructions. The importance of regular home practice at least once a day for 15–20 minutes was emphasized. The goal was to provide adolescents with an active coping strategy to be applied in everyday situations at the onset of TMD pain. The RT program has previously been evaluated in several school-based controlled trials of adolescents with recurrent headaches, with positive outcomes [42].

Because minor differences in outcomes between RT and Co treatment were obtained in RCT 1 [37], the two treatment conditions were combined into a RT/Co group in the present analysis.

Procedures

The 167 treatment completers in the two previous RCTs [37, 38] were sent a questionnaire and information about the follow-up study. The questionnaire contained 20 items concerning experience of current frequency and intensity of pain in the face, jaws, joints, or temples, impairment of chewing capacity, daily and leisure time activities and activities at work, number of disability days, help-seeking behavior, and additional treatment, general health, and experience of other pain and depressive symptoms. Those who did not respond after the first mail or a reminder that was sent, were contacted by telephone to arrange a complementary interview.

Assessment

Demographic information included gender and age.

Pain frequency: The participants were asked "Do you have pain in the temples, face, jaws or jaw joints" and to score frequency of pain on a 5-point scale, where 1 = "Never;" 2 = "Once or twice a month;" 3 = "Once a week;" 4 = "Several times a week;" and 5 = "Daily." Using the inclusion criterion of having TMD pain at least once a week as a cutoff point, the post-treatment and follow-up outcome was dichotomized into "less than once a week" vs. "once a week or more often."

Pain intensity: "How would you rate your": (1) Current pain; and, during the past month, (2) Average pain, and (3) Worst pain. Participants rated their experience of intensity on an 11-point numeric rating scale (NRS), with 0 = "No pain" and 10 = "Worst pain imaginable" as end points.

Chewing impairment: "How is your ability to eat (chew, swallow, open wide)"? This was rated on a 0–10 NRS with the end points "No interference" and "Can't do it at all."

Pain-related impairment: "In the past month, how much has TMD pain interfered with your": (1) daily activities; (2) recreation, and social and family activities; and (3) work and house work activities, rated on a 0-10 NRS with the end points 0 = "No interference" and 10 = "Unable to carry on any activities."

Number of disability days: The participants further reported on the number of days that they had been unable to participate in usual activities (e.g., work, school, or housework) because of TMD pain in the last month.

General health: "How is your general health"? This was rated by participants on a 1–5 scale, where 1 = "Very good," 2 = "Good," 3 = "Either good or bad," 4 = "Bad", and 5 = "Very bad."

Other bodily pain: “Have you experienced recurrent or long-lasting pain in other parts of the body”? The response option was, “Yes” or “No”. Participants who answered “Yes” were asked to indicate the localization of pain. They were provided with the following answer alternatives: “Head,” “Neck,” “Shoulders,” “Back,” “Hips,” “Feet,” “Knees,” “Elbows,” and “Hands.”

The Patient Health Questionnaire (PHQ)-9: The participants rated frequency of depressive symptoms during the last 2 weeks on a 0–3 scale using the following response categories: 0 = “Not at all,” 1 = “Several days,” 2 = “More than half of the days,” and 3 = “Nearly every day.” The PHQ-9 consists of nine items and possible scores therefore range from 0 to 27. Scores of 5, 10, and 15 represent cutoff points for mild, moderate, and severe depressive symptom levels. The measure has been validated in patients with a broad range of physical health problems including chronic pain [43, 44].

Help-seeking: Participants were asked “Have you sought help because of TMD pain after completion of the RCT”, and to specify the types of health care providers. The following options were given: “Physician,” “Dentist,” “Physiotherapist,” “Naprapath,” “Chiropractor,” “Occupational therapist,” “Psychologist,” “Psychiatrist,” “Counselor,” and “Other.”

Additional treatment: The subjects were also asked about type of treatment that they had received, out of the following list: “Massage,” “Occlusal splint,” “Surgery,” “Jaw exercises,” “Occlusal adjustment,” “Jaw relaxation,” “Acupuncture,” “Cortisone injection,” “Physical therapy,” “Pharmacological treatment,” “Transcutaneous Electrical Nerve Stimulation (TENS),” “Relaxation/Meditation,” “Cognitive behavioral therapy,” and “Alternative medicine.” In addition, they were asked to specify which treatment was the most effective for TMD pain, and if they would recommend it to a friend with a similar pain condition (Yes/No).

The study was approved by the Regional Ethics Committee of the Faculty of Health Sciences at Linköping University (Ref No. 2015/127-31).

Statistics

Descriptive statistics included percentages, means (M) and standard deviations (SDs) for continuous variables. In analyses of bivariate correlations between categorical variables, chi-square test was used. Differences in means between independent groups on continuous variables were examined with Student *t*-test or one-way analysis of variance (ANOVA) with or without covariate (ANCOVA). When significant main effects were obtained, subsequent pairwise comparisons were conducted using Bonferroni test. Linear and logistic regression analyses were also carried out to examine the relative importance of significant explaining factors in bivariate analyses. In all analyses, a significance level of $p<0.05$ was used.

Results

Out of a total of 116 (69.4%) who responded to the survey, the majority were women (81%). The overall mean age of participants was 30.6 (range 19–38) years (Table 1).

Temporomandibular disorder pain. The long-term evaluation showed a significant difference between genders in frequency of pain in the face, jaws, joints, or temples (*less* than once a week vs. once a week or more often). The proportion of participants who reported such pain at least once a week was significantly higher among women (37.2%) than among men (4.5%).

For pain intensity in the same location, a comparison of "Current pain," "Average pain," and "Worst pain" experience between the genders showed that pain intensity was significantly higher among women than among men (means and SDs, see Table 2).

Insert Table 2 about here

The age of the participants who reported a pain frequency of less than once a week at follow-up was significantly higher (M=31.5, SD=4.63) than among those who had not attained this level (M=28.7, SD=4.48), t(114) = 4.15, p<0.001. Similarly, the proportion of subjects who reported this pain frequency was also significantly higher among those who had participated in RCT 1 than among those in RCT 2 (Table 3). The same pattern was observed for "Current pain," "Average pain," and "Worst pain" intensity ratings, which were significantly higher among those who had participated in RCT 2 compared to RCT 1 (means and SDs, see Table 2).

Results of further analyses of pain frequency showed that long-term outcome was significantly worse for individuals who had received ST due to non-response in RCT 2 than for those who had been treated with an OA or RT/Co (Table 3). Regarding pain intensity, no significant difference between treatment groups was found for "Current pain," whereas the outcome was significantly better for those treated with an OA or RT/Co compared to ST regarding "Average pain" and "Worst pain" experience (means and SDs, see Table 2). Subsequent analysis with Bonferroni post hoc test showed no difference between the latter two groups. However, when using follow-up length as a covariate, the results of ANCOVAs showed a significant treatment group effect for "Average " and "Worst" TMD pain intensity, F(2,112) = 3.53, p< .05 and F(2,112(= 3.99, p< .05, respectively. Subsequent Bonferroni tests showed that the OA group had significantly lower scores than the ST group (p< .05), while the same difference for the RT/Co group approached significance (p =.06).

Multivariate analysis. The results of logistic regression analysis showed that there was no significant main effect for any of the explaining factors or interaction effect between trail number and treatment group for frequency of TMD pain (less or one episode per week or more at follow-up). Neither was any difference found between treatment groups for "Current" TMD pain. The results of further analysis with linear regression models revealed significant

effects for gender and treatment condition (OA combined with RT/Co vs ST) on "Average" (standardized beta -0.22 and 0.27, and t-values= -2.45 and 2,52, respectively, both $p< .05$) and "Worst" TMD pain intensity (standardized beta -0.20 and 0.27, and t-values= -2.17 and 2,58, respectively, both $p< .05$).

Impairment. The results showed no difference between gender regarding chewing capacity due to TMD pain, nor regarding the number of disability days. However, women reported significantly higher levels of impairment in daily activities, leisure time activities, and activities at work due to TMD pain compared to men (see Table 2 for mean values and SDs).

Results of further analysis showed that, compared to participants in RCT 2, participants in RCT 1 experienced significantly lower impairment levels on all measures except for number of days with reduced ability to participate in daily activities.

Differences related to treatment condition were significant for all these outcome measures, except for number of days with impairment in common daily activities, which approached significance (p=0.06). In all subsequent post hoc analyses, participants who had received ST showed significantly higher impairment levels compared to those in the other two treatment conditions, which were not significantly different from each other.

Health, other pain and depressive symptoms. At the long-term follow-up, 88% of the participants regarded their general health as "Good" or "Very good," while 61.2% reported another pain in the body, and 7.8% reported moderate or severe depressive symptom levels on the PHQ scale (M=4.59, SD=5.65). The difference in reported health was non-significant for gender and treatment condition. However, a higher proportion of older participants from RCT 1 (92.9%) reported "Good" or "Very good health" as compared to those in RCT 2 (80.4%), $\chi 2 (1) = 4.04$, $p< .05$.

While a significantly higher proportion of women (67%) than of men (36.4%) reported another pain localization, $\chi 2$ (1) = 7.06, $p < .01$, no difference was found for trial number or treatment condition. Women also reported significantly more other pain locations compared to men (means and SDs, see Table 2). Results of further analyses showed that number of pain locations was significantly different between treatment groups. Subsequent post hoc test showed that participants who had received ST reported a significantly (p=.05) higher number of pain locations compared to those in OA treatment.

While no difference was found for depressive symptom levels (no or mild vs. moderate to severe) for gender a significant association was found between the two RCTs and depressive symptom levels, $\chi 2$ (1) = 4.10, $p < .05$, in that 23.9% of the younger participants in RCT 2 reported moderate to severe levels as compared to 10% of those in RCT 1. Although depressive symptom levels were higher among those who had received ST (24%) than for those who had been treated with an OA (14.6%) and RT/Co (12%), this difference was non-significant.

Insert table 3 about here

Help seeking and additional treatment. During the follow-up period, 22.4% of participants had sought professional help due to pain in the jaw, face, and joint. Whereas no difference was found related to gender or trial number, a significant relationship was found in regard to treatment condition. The proportion was significantly lower in the OA group compared to the RT/Co and ST groups (Table 3). During follow-up, most patients had sought help from a dentist (16.4%), a chiropractor or naprapath (8.6%), a physician (7.6%), and/or other professionals (5.3%).

During the follow-up period, 22.4% of the participants had received additional treatment due to pain in the jaw, face, or joint. The difference between gender and the RCTs was non-

significant. Although the proportion of participants who had received OA treatment was lower than for those in the RT/Co and ST groups the differences were non-significant (Table 3). Most participants had received additional OA treatment (11.2%) or massage (10.4%), these being rated as the most helpful methods; 4.3% had been treated with jaw movement training or relaxation, 3.5% with meditation/relaxation, 3.4% with acupuncture, and 7.8% with other treatment methods. The received treatments were also recommended by almost all recipients (91.7%) to a friend with similar pain problems.

Discussion

This long-term follow-up study investigated TMD in adolescents previously included in two RCTs and treated for frequent TMD pain with an OA, RT, or information given alone or combined with the two other treatments. The overall majority of participants reported TMD pain less than once a week in the follow-up. Our findings also showed that adolescents treated with an OA showed somewhat better sustained improvement over the extended follow-up period than for those treated with RT/Co, and that non-responders to treatment in the sequential treatment condition and females exhibited a poorer outcome.

Occlusal appliance and other treatment methods such as behavioral approaches including RT, biofeedback, and cognitive behavioral therapy, have in the short-term perspective been found to successively reduce TMD pain in adult patients [29–32]. Several long-term follow-up studies using appliance therapy have reported similar positive outcomes in adult TMD pain patients [33–36]. In the present study, lower levels of frequency and intensity of TMD pain, less impairment and fewer depressive symptoms, and better health were more commonly reported at the follow-up by older individuals (mean age 33.5 years) who had participated in our first trial (RCT 1) than by the younger participants who had been included in RCT 2 (mean age 25.6 years), suggesting that the overall prognosis in the long run also improves into

middle age. Such changes due to the mere passage of time are likely to depend on factors not specifically related to intervention, such as placebo effects, spontaneous remission, or statistical regression effects or shared external environmental factors.

Differential long-term effects were also observed in regard to treatment and gender as did findings in a short-term perspective for participants in our two trials in which girls had a poorer outcome than boys [45]. Noteworthy is also that the long-term outcome was significantly worse for adolescents who did not respond to treatment in the first period of our cross-over study despite receiving additional treatment in the subsequent period. In this treatment group, only 40% had achieved a TMD frequency of less than once a week, compared to most of the participants (73–80%) in the OA and RT/Co groups. Multiple pain sites, levels of depressive symptoms, somatization, and catastrophizing have been proposed to predict a poorer outcome in adult patients treated for TMD [46]. Similar findings in the present study, indicating a poorer long-term prognosis, were also evident among non-responders to treatment as they reported more TMD and other pain, higher levels of depressive symptoms, and social impairment compared to those treated with an OA and RT/Co.

In a 1-year prospective study of TMD pain patients primarily consisting of women (83%), the report of several previous health care visits and comorbid pain, with high pain intensity and disability, indicated a poorer TMD prognosis [47]. These findings also concur with the results of the present study of treated adolescents, in which more females than males reported more severe TMD pain, with an impact on their everyday social function, and also experienced more other bodily pain symptoms. These findings indicate a particular, and increased, risk for prolonged TMD pain in adult women compared to men.

In a general population-based survey investigating care-seeking behavior because of orofacial pain, the authors reported that the likelihood of seeking professional health care increased with age, number of pain symptoms, frequency and intensity of pain, duration of pain episodes, and disability due to pain, as well as a decreased ability to control and reduce the pain [48]. The care-seeking behavior because of TMD pain has also been reported to be more common in women than in men [4]. In a community sample of individuals suffering from different bodily pain, such as back pain, headache, chest pain, abdominal pain, or TMD pain, the authors found that when men and women reported similar pain levels they also sought care at similar rates [49]. By contrast, in the present clinical sample of individuals previously treated in adolescence, we found no differences with regard to gender and help-seeking behavior for TMD pain in adulthood even though pain intensity was significantly higher among the women than among the men.

In an 8-year follow-up of treatment with appliance therapy (occlusal or non-occluding appliances) administered to an adult sample, the majority of patients (63%) had received additional treatment mostly consisting of use of another OA [36]. In the present long-term follow-up, the proportion of individuals who had sought additional treatment during the extended follow-up was substantially lower (22.4%), and their treatment had mainly consisted of OA and massage. What we find noteworthy is that the proportion of help-seekers was even lower (9.8%) among those previously treated with the most effective treatment method, i.e., an OA. This seems to suggest that the various types of treatments administered to the adolescents and decreasing their TMD pain complaints also contributed to a lower need for additional treatment during the extended follow-up period.

Limitations and strengths

Some limitations of the present study need to be highlighted. One weakness is the somewhat limited response rate in our study (69.4%); however, given the extended length of follow-up it appears to be acceptable. Our response rate is also comparable to the rate in other, similar long-term follow-up studies in adults (69–76%) [35–36]. A second limitation is that the design of the study comprised two RCTs with clinical samples of adolescents also including an arm of non-responding participants in our second RCT (RCT 2) in which crossover treatment was administered in a sequential and non-randomized fashion. The relatively few responders in the first treatment phase in RCT 2 were here included together with responders and non-responders in the OA and RT/Co groups in the other RCT (RCT 1), thus slightly overestimating the effects of these treatments. Another potential confounder of outcome is that less than a quarter of the participants also had received additional treatment during the long-term follow-up. However, the proportion of having received such help was lowest among those having received OA (about 10%) contrasting to about one quarter or one third among participants in the RT/Co and ST groups. Finally, all information at the follow-up was based on questionnaire data and did not include clinical assessment.

The strengths of our study are that the same inclusion TMD criteria were applied for inclusion of adolescents in two controlled trials and that they were recruited from the same catchment areas in one county and assessed using the same measures and procedures. The wide range of follow-up time in the two RCTs has further provided an opportunity to evaluate whether the time length of follow-up after treatment had a differential impact on outcome.

Conclusion

Differential treatment effects were found in our extended follow-up study in that adolescents treated with an OA showed somewhat better, sustained improvement, with lower health care use over the extended follow-up period compared to those treated with the alternate method,

RT/Co. However, the most striking findings were that non-responders to these treatments as well as females clearly exhibited a poorer outcome. These outcomes underline that the latter groups need particular attention and should be offered extended treatments or different treatments to achieve a better outcome. Further treatment outcome research is needed to investigate which treatment components are effective for adolescents suffering from frequent TMD pain and comorbid complaints in a longer perspective.

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