Standardizing the clinical orofacial examination in Juvenile idiopathic arthritis: An interdisciplinary, consensus-based, short screening protocol

Short title: Facial examination in JIA

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Abstract

Objective To develop a consensus-based, standardized, short clinical (<3 minutes) examination protocol to assess the multidimensional, orofacial manifestations of juvenile idiopathic arthritis (JIA).

Methods The study was conducted by a multidisciplinary task force from the Temporomandibular joint juvenile arthritis working (TMJaw) group. The study used an acknowledged sequential approach involving: 1) a global multidisciplinary online questionnaire study, 2) a systematic literature review and consensus-meetings to identify items for inclusion, 3) pilot-testing of included items, 4) test of reliability in 22 subjects with JIA by four examiners, 5) test of construct validity in a case-control study involving 167 subjects, 6) establishment of final recommendations.

Results Six items were recommended for the final examination protocol: 1) clinician assessed pain location, 2) Temporomandibular joint pain on palpation (open and closed mouth), 3) Mandibular deviation at maximal mouth opening (≥3mm), 4) maximal unassisted mouth opening capacity, 5) frontal facial symmetry, 6) facial profile. All recommended items showed acceptable reliability and construct validity. The average mean examination time was two minutes and 42 seconds (SD ± 38.5 seconds).

Conclusion. A consensus-based, short clinical examination protocol was developed. The protocol takes less than 3 minutes to complete and provides information about orofacial symptoms, temporomandibular joint dysfunction, and dentofacial deformity. The standardized examination protocol is applicable to routine clinical care as well as future research studies.

Introduction

Within the past decade, increased attention has been paid to the consequences of temporomandibular joint (TMJ) arthritis in patients with Juvenile Idiopathic Arthritis (JIA). TMJ arthritis is a frequent feature of JIA (1-3).

TMJ involvement may lead to abnormal dentofacial development and significant orofacial disabilities, including chronic orofacial pain and reduced TMJ mobility and masticatory function (4-10). The orofacial manifestations of JIA can have a severe impact on health-related quality of life that may persist into adulthood (8, 11-13). Gadolinium-enhanced magnetic resonance imaging (MRI) is currently the gold standard for diagnosing active TMJ arthritis (3, 7, 14-16).

The clinical orofacial examination constitutes an essential component of the clinical assessment of individuals with JIA, and serves four equally important purposes: 1) the detection of clinical signs of active TMJ arthritis that should prompt further clinical and imaging investigations; 2) the detection of orofacial manifestations caused by previous TMJ arthritis (TMJ involvement); 3) the assessment of dentofacial growth and development in skeletally immature subjects; and 4) the assessment of the longitudinal progression of orofacial symptoms and dysfunction in patients who have already been diagnosed with active TMJ arthritis or TMJ involvement.

Contemporary orofacial examination techniques are based on validated criteria and indices like the Research Diagnostic Criteria for Temporomandibular Disorders (RDC/TMD) and Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) and the Helkimo index (17, 18). However, these tools vary in their complexity and the time required for completion, and do not specifically focus on the orofacial manifestations of JIA. In 2017, general, consensus-based, recommendations were published for the clinical orofacial examination in JIA (19). However, at this point, no JIA-specific, interdisciplinary, consensus-based protocol exists for the clinical orofacial examination in JIA.

The objective of the present study was to develop a consensus-based, standardized short clinical examination protocol to assess aspects of JIA-induced orofacial manifestations to be used routinely in the clinical setting and in future research studies. The examination protocol should be applicable to all health care providers regardless of educational background.

Material and Methods

The study was conducted by a task force from the Temporomandibular Joint Juvenile Arthritis Working Group (TMJaw). TMJaw (formerly known as "EuroTMjoint") is an international, multidisciplinary research network dedicated to studying TMJ arthritis in JIA. The initial task force represented researchers from Europe and North America and consisted of three pediatric rheumatologists, three specially trained orthodontists and two specialists in orofacial pain. Using a sequential-based approach, the present study included the following aspects (20): 1) Conceptual phase and preliminary decision-making, 2) item generation, 3) pilot-testing, 4) test of reliability, 5) test of construct validity, 6) establishment of final recommendations.

Phase 1: Conceptual phase and preliminary decision-making

Initially, the conceptual framework was defined by the task force. In phase 1, a global online questionnaire, asking about JIA management, and approaches to the clinical orofacial examination in JIA was created. In February 2013, members on the mail distribution lists of the *Pediatric Rheumatology Bulletin Board* and TMJaw group were invited to participate in a questionnaire study using the Survey Monkey™ online platform. This assessed: 1) Respondent-related characteristics (professional background, practice setting, geographic location, self-reported expertise in clinical orofacial examination in JIA); 2) maximum amount of time that can be devoted to the clinical orofacial examination during a full-body examination, and 3) ranking of the five most important examination items to include in the clinical orofacial examination of JIA patients. The outcome of the online survey was used to inform item generation.

Phase 2: Item generation

From April 2013 to January 2017 the task force developed general interdisciplinary consensus-based recommendations for the orofacial examination in JIA (19). Following acknowledged steps for the generation of consensus-based guidelines, this project involved a comprehensive systematic literature review and subsequent consensus-meetings. The systematic literature review provided evidence to support inclusion of specific examination items relevant for clinical orofacial examination in JIA. Details about the systematic review and the results are presented in Stoustrup *et al*. 2017 (19). The importance of each of the proposed examination items was assessed during a three-round Delphi study completed by participants on the TMJaw mailing list. During a consensus-meeting in Tampere, Finland in April 2014, the task force used the Delphi study

outcome to identify preliminary examination items for inclusion in a short clinical orofacial examination protocol.

Phase 3: Pilot-testing

The task force created a clinical form which included the preliminary examination items together with detailed instructions on how to perform each clinical examination item. The feasibility and the clinical applicability of the form and instructions were tested at the Section of Orthodontics, Aarhus University, Denmark from April 2014 until December 2016. The ongoing clinical pilot-test led to modification of the examination form and the instructions. The task force decided on a final set of examination items in March 2017.

Phase 4: Test of reliability

In September 2017, subjects with JIA, followed at Section of Orthodontics, Aarhus University, Denmark were randomly selected and invited to participate in a reliability study to assess intra-rater and inter-rater agreement of the proposed examination items. Inclusion criteria were: 1) JIA diagnosis according to the International League of Associations for Rheumatology (ILAR) classification criteria (21); 2) ≥ 7 years and ≤ 18 years; and 3) able to cooperate with the clinical orofacial examination. All subjects were examined by four raters; two pediatric rheumatologists (TH, MT) and two orthodontists (PS, TKP). Subjects were assessed in a random sequence, and were examined twice by all four raters, with a 1-3 hour time lag between the first and the second examination. Prior to the reliability study, a three-hour clinical calibration session, involving five patients, was conducted among the four raters.

Phase 5: Test of construct validity

To assess construct validity, inter-group differences were calculated for each of the examination items between consecutive subjects with JIA and a random group of age-matched non-JIA controls. The JIA group consisted of consecutive subjects seen at the Section of Orthodontics, Aarhus University, Denmark in compliance with the inclusion criteria: 1) JIA diagnosis according to ILAR criteria (21), 2) ≥ 7 years and ≤18 years and compliant with the clinical orofacial examination. The control group consisted of non-JIA subjects followed at the pediatric dental municipal clinics in the districts of Syddjurs and Vesthimmerland, Denmark. Inclusion criteria for

the non-JIA controls were: ≥ 7 years and ≤ 18 years and able to cooperate with the clinical orofacial examination.

Associations between the two groups were assessed following predefined hypotheses (H):

H1: Subjects with JIA have more frequent orofacial pain in comparison to age-matched non-JIA control subjects.

H2: Subjects with JIA demonstrate reduced mandibular function in comparison to non-JIA control subjects.

H3: Subjects with JIA demonstrate more severe dentofacial growth abnormalities in comparison to non-JIA subjects.

Phase 6: Establishment of final recommendations

The results of the field-testing (reliability and construct validity) were used to establish the final recommendations. A consensus-driven approach was used and all authors accepted the final recommendations.

Statistics

Descriptive statistics were computed. Multi-rater Cohen´s kappa was calculated to assess reliability for categorical data. Intra-class correlations coefficient (ICC) was calculated to assess reliability in quantitative data (maximal mouth opening). Construct validity was tested against the predefined hypotheses using chi-square tests for categorical data. The Fisher's exact test was used in outcome variables with less than 5 subjects in either the control group or the JIA group. An unpaired t-test was used for inter-group difference for quantitative data (maximal mouth opening capacity). Construct validity was only accepted if all pre-defined hypotheses were accepted. The level of significance was $p<0.05$.

Miscellaneous

The study was approved by the Danish Data Protection Agency (1-16-02-16-16 and Aarhus University 20016-051-000001) and conducted in agreement with Danish Health authority regulations on non-interventional studies. Prior to inclusion, informed and signed consent was provided by all participants ≥15 years of age, or by their parents for participants below age 15. All

examination items are approved for use in pediatric patients. The study adheres to TMJaw consensus-based standardized terminology (22).

Results

Conceptual phase and preliminary decision-making

The online questionnaire was completed by 167 health care providers. The majority of the respondents were pediatric rheumatologists (85.6%) and orthodontists (6.6%) (Figure 1a). Respondents represented the following continents: North America (56,6%), South America (6.5%), Europe (35.5%), Australia and Oceania (1.2%). The vast majority were affiliated with academic hospitals (90.5%). The respondents rated their own experience with TMJ and orofacial examination as follows: No experience (1.2%), minimal experience (9.5%), average experience (44%), moderate experience (32.7%), and expert experience (12.5%). Respondents were asked to suggest important clinical examination items and to assess the maximal time needed to complete a clinical orofacial examination during a full-body examination: <1 minute (9.5%), 1-3 minutes (43.5%), 3-5 minutes (24.4%), 5-10 minutes (16.1%), >10 minutes (6.5%) (figure 1b). Based on the results of the questionnaire, the task force decided on a 3-minute time limit for the final examination protocol.

Item generation

The systematic literature review provided evidence to include 12 general items relevant for the clinical orofacial examination in JIA patients (19). The importance of each of the 12 examination items was assessed during a three-round Delphi study by members on the TMJaw mailing list (n=40). Each of the 12 proposed examination items was rated on a 10-point numerical scale (0=Not important, 10=Of utmost importance). Examination items were then subcategorized based on their ratings of importance: “high importance” (score ≥ 8), “moderate importance” (score ≥ 6 and <8), “low importance” (score <6) (19). Based on the Delphi-study results, the task force recommended six examination items for the short clinical examination protocol: 1) clinician assessed pain location, 2) TMJ pain on palpation with open and closed mouth (unilateral, bilateral), 3) Mandibular deviation at maximal mouth opening (≥3mm deviation to the right or left side), 4) maximal unassisted mouth opening capacity measured in millimeters, with the vertical incisal overlap taken into account, 5) frontal facial symmetry (presence of asymmetry), 6) facial profile

(straight, mild convex, moderate convex, micrognathic). To ensure content validity, only items receiving a "high importance" Delphi survey categorization were included in the clinical examination protocol. Specific description of outcomes for examination items are described in Table 1 and Figure 2.

Test of reliability

Twenty-two subjects with JIA were enrolled in this phase of the study. The mean age was 11.6 years (SD±2.5 years) and 55% were girls (n=12). Acceptable intra-rater and inter-rater kappa values were calculated for all examination items ranging from 0.41 to 0.81 (Table 2). According to Landis and Koch a kappa-statistic agreement is "moderate" between 0.41-0.60, "substantial" between 0.61-0.80, and "almost perfect" when >0.80 (23). The average mean examination time across all four raters was two minutes and 42 seconds (SD ± 38.5 seconds, range 90-277 seconds).

Test of construct validity

Two groups with a total of 167 subjects were included in the test of construct validity: JIA group (n = 76, mean age 12.22 years, SD ± 3.0 years) and the control group (n = 91, mean age 13.45, SD ± 2.6 years). The control group was significantly older and included significantly more boys in comparison to the JIA group. Characteristics of included subjects are displayed in Table 3. The results of construct validity testing are displayed in Table 4:

H1 was accepted: A significant larger proportion of JIA subjects (17%) reported orofacial pain within the last two weeks when compared to controls (6%).

H2 was accepted: JIA subjects had a significantly higher frequency of TMJ pain on palpation with open mouth (20% vs. 7%) and mandibular deviation at maximal mouth opening (22% vs 4%). Additionally, maximal mouth opening was significantly reduced between JIA and control groups (difference: -3.17 mm, 95% CI: -4.95 to -1.38 mm).

H3 was accepted: The JIA group displayed a significantly greater proportion of facial asymmetry (65% vs 34%) and presence of micrognathic profiles (7% vs 0%) when compared to controls.

Establishment of final recommendations

The results were presented to members of the task force and consensus of the final recommendations was created through email correspondence. The clinical examination protocol and specific instructions for each item are found in the online supplemental material.

Discussion

This project proposes a consensus-based, short, clinical examination protocol for routine use in clinical care and research settings in subjects with JIA. The screening protocol consists of six unique items, which encompass features of TMJ symptoms, TMJ dysfunction, and dentofacial deformity. Detailed instruction for each of the items have been developed to support clinical training and enhance reliability across health care providers (Online supplement material). The items show acceptable inter-rater reliability and construct validity, and represent some of the most consistently used outcome variables in the existing literature dealing with the orofacial examination in JIA (19). Most included items originate from traditional orofacial examination methods like the Helkimo index and the DC-TMD criteria (17, 18). However, unique to this project, we have identified a group of "traditional" items that are specifically relevant to JIA-related orofacial manifestations and combined those with additional items to assess dentofacial growth and development.

Identification of TMJ involvement in JIA patients is the first step to appropriate management. The protocol can be completed in less than 3 minutes. This meets the optimal time limit determined by the online questionnaire with respect to the maximal amount of time the pediatric rheumatologist can devote to a dentofacial examination. The short completion time makes this protocol a valuable addition to the routine full body assessment of JIA patients. The standardized clinical examination provides a first-line, non-invasive, solid foundation for the dentofacial evaluation when conducted in combination with contemporary imaging and radiological examination standards (24).

According to the recent consensus-based recommendations on TMJ arthritis-related terminology, TMJ arthritis is defined as active inflammation in the TMJ, whereas TMJ involvement is defined as abnormalities presumed to be the result of TMJ arthritis (22). In general, the absence of orofacial symptoms is not a valid predictor for the absence of TMJ inflammation and vice versa (7). Standardized TMJ MRI examinations were not available for participants in the JIA group. It is

therefore unclear whether the orofacial symptoms and dysfunctions in the JIA group is due to active TMJ arthritis or TMJ involvement.

Across the literature, assessment of mouth opening capacity is the most frequently deployed clinical orofacial examination item in JIA (7, 19). In this study, the maximal mouth opening capacity in the JIA group was significantly reduced. Cross-sectional studies have shown a limited diagnostic sensitivity of reduced maximal mouth opening capacity of <40 mm in subjects with TMJ arthritis (1, 9, 16). Abramowicz et al. have reported that patients with a limited mouth opening capacity of two standard deviations below age-related normative values were 6.7 times more likely to have TMJ arthritis (14). Furthermore, Abramowicz et al. have also demonstrated that limited mouth opening capacity in combination with mandibular deviation at maximal mouth opening was associated with a predictive value of 1.00 for the presence of MRI-verified TMJ synovitis (14). Recent systematic reviews have shown that the presence of mandibular deviation at maximal mouth opening is one of the most sensitive predictors for the presence of TMJ inflammation in JIA (7, 19).

Assessment of mouth opening capacity is also the most frequently used outcome variable in TMJ arthritis follow-up studies. Changes in mouth opening capacity have been used as an indirect measure of TMJ functional status; post-interventional increase in opening capacity has been regarded as a sign of TMJ functional improvement. Commercial products exist to assist assessment of maximal mouth opening. In addition, methods like the “3 finger assessment method”, and standardized cut-off values for assessment of mouth opening capacity have been proposed (25, 26). We recommend including the vertical incisal overlap when measuring maximal mouth opening capacity in JIA in relation to age-related normative values. This takes into account the substantial change of mouth opening capacity with growth and development, during transition between primary and permanent dentition (27-29).

TMJ arthritis is a subcategory within the general term temporomandibular disorders (TMD) (18). TMD diagnoses encompass both dysfunctional and autoimmune etiologies as well as pain conditions . The conditions vary from mild, temporary, non-symptomatic disc issues to severe conditions like TMJ degeneration, myalgia, and chronic orofacial pain conditions (18). The reported prevalence of TMD is 10-16 percent in the non-JIA adolescent population, which is greater than the prevalence of four to seven percent in the control group of present study (30, 31).

This substantial difference is explained by differences in methodology. In the current protocol, we decided to exclude assessment of TMJ noise (clicking and crepitation) due to low diagnostic sensitivity for TMJ arthritis in JIA (1). A meta-analysis performed by Da Silva et al. demonstrated that the most prevalent clinical finding of TMD in the non-JIA population was asymptomatic TMJ noises (30).

It is noteworthy that the symptoms and clinical findings of arthritis-induced dysfunction are comparable to those encountered in other TMDs (18, 30). Differential TMD should be considered in patients with JIA who present orofacial dysfunction or dentofacial deformities during the clinical examination. Such findings may not exclusively be caused by active TMJ arthritis from JIA. This is illustrated by the fact, that dentofacial asymmetry was found in 34 percent of the control group in the present study. This is consistent with research by Liukkonen et al., who reported dentofacial asymmetry to be a common clinical finding in the background population (32).

A standardized orofacial examination will provide complex information about dentofacial function, growth, and development. Regardless of etiology, abnormal clinical findings are a red flag, and should prompt increased attention during follow-up visits, and referral for appropriate imaging when indicated. Follow-up imaging should be guided by recent consensus-based protocols for TMJ magnetic resonance imaging (MRI) (15, 33) and 3D assessment of TMJ deformity and dentofacial deformities in JIA (10, 15). The various TMJ imaging techniques come with their own benefits, drawbacks and limitations (34).

Attention to dentofacial growth and development is an important examination item to help detect dentofacial deformities. Economou et al. demonstrated a significant correlation between dentofacial hard-tissue and soft-tissue asymmetries in JIA where even minor mandibular asymmetries were detected by visual inspection during the clinical examination (35). Findings by Ikavalko et al. also support the valid use of profile assessment to identify subjects with micrognathic mandibles (36). This study demonstrates that moderate dentofacial convexity may be found in JIA as well as in the background population. In contrast, micrognathia was only identified in JIA. Management of arthritis-induced dentofacial deformity can be guided by recent recommendations (37).

Longitudinal, interventional studies have documented a poor association between the fluctuation of orofacial symptoms/dysfunction and post-interventional MRI findings (38-40). From a clinical standpoint, this highlights the important contribution of both clinical and MRI examinations and underlines the relevance of both modalities in the dentofacial health assessment in JIA. Previous research has focused on the ability to predict the presence of TMJ inflammation based on items from the clinical examination. Less attention has been devoted to studying the implications of dentofacial signs and symptoms on long-term outcomes regardless TMJ status. Recent data from a Danish cohort study revealed that 56 percent of the cohort presented with at least one clinical sign of dentofacial dysfunction and 35 percent were diagnosed with an arthritis-induced dentofacial deformity within the first 5 years after JIA diagnosis (41). These findings underscore the importance of routine, standardized orofacial examination in JIA patients.

There are certain limitations to this study that warrant further consideration: 1) Not all of the proposed examination items reached above a "moderate" agreement level (r=0.41-0.60) during assessment of intra-rater and inter-rater reliability. 2) Although this protocol consists of the most widely used outcome variables for assessment of TMJ arthritis in interventional studies (19), its ability to detect changes in orofacial dysfunction (responsiveness) still needs to be evaluated in future studies. 3) The significant difference in age and gender between the JIA group and the control group in the construct validity test: Inter-group differences in age and sex are potential sources of biases to the test of construct validity since general TMD is most often found in pubertal girls (30, 31). 4) Also, the absence of routine MRI examination for assessment of TMJ arthritis/involvement in all JIA group subjects is considered a limitation to the present study.

Significant strengths of the study: The protocol was meticulously developed by using established sequential-phased approach in an interdisciplinary setting. In phase 1, the global online questionnaire strengthened the clinical usability of the proposed examination protocol. Since treatment of TMJ arthritis involves an interdisciplinary approach, a primary goal of our recommendations was to create a protocol that can be used by healthcare providers without specialized training in the TMJ and dentofacial examination. Another strength is the detailed instructions provided with each item found in the online supplements. An important future focus is to produce educational video material to ensure reliability and validity among health care providers who are less experienced with orofacial examination.

In summary, we have developed a consensus-based short clinical examination protocol showing acceptable construct validity and test-retest reliability. This protocol takes less than 3 minutes to complete and will generate essential information about TMJ symptoms, TMJ dysfunction, and dentofacial deformity. It is our hope that this screening protocol will be integrated into standard clinical care and will be incorporated in future research studies.

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Legends

Figure 1. Conceptual phase and preliminary decision-making: a) Professional background of responders (n=167) to online survey dealing with orofacial examination in JIA. b) Response to the question: “What is the maximal time that can be devoted to clinical orofacial examination during a full-body examination of subjects with JIA?".

Figure 2

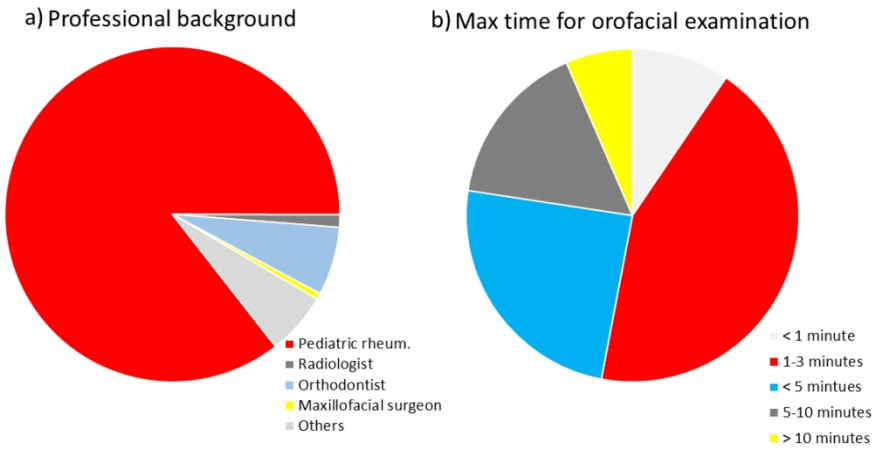
Figure 2. Clinical examination items in the short screening protocol. a) Temporomandibular joint palpation with closed mouth. b) Temporomandibular joint palpation with open mouth. c) Maximal mouth opening capacity. Please see online supplemental material for instructions on how to account for the vertical incisal overlap. d) Mandibular deviation at maximal mouth opening. “X” indicate the chin-point. e) Assessment of facial symmetry. f) Assessment of facial profile (e.g. Convexity)

Table 1. Description of items included in the short clinical examination protocol. TMJ, Temporomandibular joint. * During maximal mouth opening (including incisal overlap).

Table 2. Test of reliability. Mean prevalence of findings among the four raters, 95% confidence interval in brackets. Intra-rater and inter-rater reliability by Cohen´s kappa. Twenty-two subjects with juvenile idiopathic arthritis examined by four raters. *The prevalence is calculated as an average value of the findings of the four raters. **continuous data and calculated as an intra-class correlation coefficient. CI: Confidence interval; SD: Standard deviation.

Table 3. Cohort characteristics for test of construct validity

Table 4. Test of construct validity. Inter-group proportional difference for examination items included in the clinical examination protocol. Maximal mouth opening capacity is presented as continuous data. Frequencies are reported on patient-level. * At maximal mouth opening position, **Fisher’s exact test used since n< 5 subjects in control group. *** Comparison of means with unpaired *t*-test. CI: Confidence interval; SD: Standard deviation.



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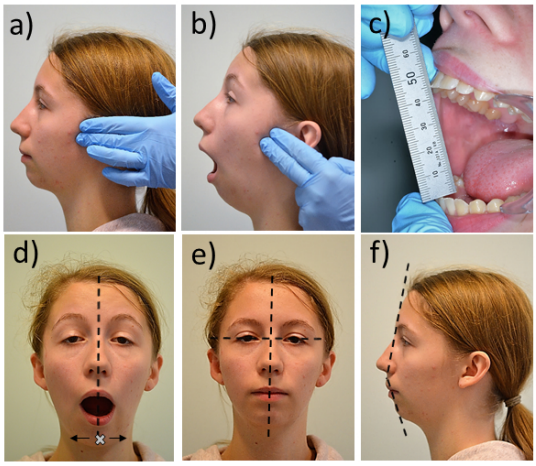
338x190mm (96 x 96 DPI)



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288x150mm (96 x 96 DPI)

Table 1

Examination item	Outcome measure	Assessment of outcome
Clinician assessed pain location	TMJ symptoms	Pain areas are marked on face-map
TMJ pain on palpation		
-Closed mouth	TMJ symptoms,	Four outcomes: No pain, unilateral right-sided TMJ pain, unilateral left-sided TMJ pain, bilateral TMJ pain
-Open mouth	TMJ symptoms	Four outcomes: No pain, unilateral right-sided TMJ pain, unilateral left-sided TMJ pain, bilateral TMJ pain
Mandibular deviation (≥3mm)*	TMJ dysfunction	Three outcomes; No deviation, right-sided deviation, left-sided deviation
Maximal mouth opening	TMJ dysfunction	Absolute measure in millimeters
Frontal facial asymmetry	Dentofacial anomaly	Three outcomes: No asymmetry, right-sided asymmetry, left-sided asymmetry
Facial profile	Dentofacial anomaly	Four outcomes: Straight, mild convex, moderate convex, micrognathic

Table 1. Description of items included in the short clinical examination protocol. TMJ, Temporomandibular joint. * During maximal mouth opening (including incisal overlap).

Table 2

Examination item	Mean prevalence of subjects with finding n=22 subjects. (95% CI)*	Intra-rater reliability	Inter-rater reliability
Clinician assessed pain location	25% (17.1-35.0)	0.81	0.57
TMJ pain on palpation			
Closed mouth	10% (5.3-18.5)	0.41	0.52
Open mouth	24% (16.1-33.8)	0.77	0.66
Mandibular deviation	30% (21-39.8)	0.60	0.47
Maximal mouth opening	50.5 mm (SD 5.7mm)	0.88 (95%-CI: 0.82-0.92)**	0.77 (95%-CI: 0.61-0.89)*
Frontal facial asymmetry	71% (60.2-79)	0.76	0.44
Facial profile	-	0.47	0.46
moderate convex or micrognathic	21% (13.3-30.1)	-	-

Table 2. Test of reliability. Mean prevalence of findings among the four raters, 95% confidence interval in brackets. Intra-rater and inter-rater reliability by Cohen´s kappa. Twenty-two subjects with juvenile idiopathic arthritis examined by four raters. *The prevalence is calculated as an average value of the findings of the four raters. **continuous data and calculated as an intra-class correlation coefficient. CI: Confidence interval; SD: Standard deviation.

Table 3.

Cohort characteristics	JIA group	Control group
Number	76	91
Females	53 (69.7%)	39 (42.9%)
Mean age at baseline, years (SD)	12.22 (3.0)	13.45 (2.6)
JIA subcategories, number		
Oligoarticular	46 (61%)	-
Polyarticular	26 (34%)	-
Systemic	3 (4%)	-
Psoriatic	-	-
Enthesitis related arthritis	1 (1%)	-
Unknown	-	-
Medical treatment at time of orofacial examination		
No medication	34 (45%)	-
NSAID	10 (13%)	-
Methotrexate	24 (32%)	-
Leflunomide	6 (8%)	-
Systemic steroid	-	-
Anti-TNF	26 (34%)	-
Anti-IL6	3 (4%)	-
Single drug	22 (29%)	-
Combination of two drugs	16 (21%)	-
Combination of three drugs	3 (4%)	-

Table 3. Cohort characteristics for test of construct validity

Table 4

Examination item	JIA group prevalence n=76 (n, 95% CI)	Control group prevalence n=91 (n, 95% CI)	Inter-group difference
Orofacial pain in past two weeks	17% (n=13, 10.1-27.2)	7% (n=6, 0.3-14)	p=0.033
TMJ pain on palpation			
Closed mouth	11% (n=8, 5.2-19.7))	6% (n=5.2-12.5)	n.s.
Open mouth	20% (n=15, 12.2-30.2)	7% (n=6, 2.8-13.9)	p=0.011
Mandibular deviation*/**	22% (n=17, 14.4-33.0)	4% (n=4, 1.4-11.1)	p= 0.001
Maximal mouth opening***	49.0 mm (SD 6.2 mm)	52.2 mm (SD 5.5 mm)	p< 0.001
Frontal facial asymmetry	65% (n=49, 53.2-74.3)	34% (n=31, 25.1-44.3)	p=0.001
Facial profile: Moderate convex, or micrognathic	17% (n=13, 10.1-27.2)	11% (n=10, 5.9-19.2)	n.s.
Facial profile: Micrognathic**	7% (n=5, 2.5-14.8)	0% (n=0)	p=0.018

Table 4. Test of construct validity. Inter-group proportional difference for examination items included in the clinical examination protocol. Maximal mouth opening capacity is presented as continuous data. Frequencies are reported on patient-level. * At maximal mouth opening position, **Fisher's exact test used since n< 5 subjects in control group. *** Comparison of means with unpaired *t*-test. CI: Confidence interval; SD: Standard deviation.