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Agreement between child- and parent-reported orofacial symptoms in patients with juvenile idiopathic arthritis

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Objective: To assess the agreement between child- and parent-reported orofacial symptoms in the Danish version of the patient questionnaire Assessment of Orofacial Symptoms in Juvenile Idiopathic Arthritis.

Method: This cross-sectional study was conducted at Aarhus University in March 2023. Eligible candidates were consecutive subjects with juvenile idiopathic arthritis (JIA) and temporomandibular joint involvement accompanied by a parental proxy for examination in the Craniofacial Clinic. After obtaining written informed consent, the questionnaire was completed individually and separately by the child and the parent without any communication between them. The level of agreement was analysed using Cohen's (weighted) kappa for nominal and ordinal outcome variables (orofacial pain frequency, pain location, jaw function, orofacial symptoms, and changes since last visit) and the intraclass correlation coefficient for linear outcome variables (orofacial pain intensity and functional disability of the jaw).

Results: The 34 included dyads had an overall 'poor' to 'moderate' child-proxy reporting agreement on the questionnaire for the assessment of JIA-related orofacial symptoms. After dividing the children into two age groups, < 13 and ≥ 13 years old, we found substantial agreement on pain frequency and moderate to excellent agreement on pain intensity for the older group. The child-proxy agreement for children aged < 13 years was slight on pain frequency and poor to moderate on pain intensity.

Conclusion: The child-proxy reporting agreement on JIA-related orofacial symptoms is inconsistent. We suggest collecting information from both children and parents, especially when assessing orofacial pain and symptoms in children < 13 years of age.

Juvenile idiopathic arthritis (JIA) is the umbrella term for arthritis of unknown origin in children, with an onset before the age of 16 years (1). The temporomandibular joint (TMJ) is frequently involved (30–49%), leading to craniofacial growth disturbances and orofacial dysfunction, with impacts on quality of life (2–7). The presence of orofacial

signs and symptoms is more prevalent in JIA than in the background non-JIA population, and the orofacial manifestations of TMJ involvement can continue into adulthood, with the risk of a pronounced impact on patients' overall health (8, 9). Diagnosis and management of TMJ arthritis and the related orofacial manifestations can be challenging, calling for interdisciplinary efforts (7, 9–11). In addition to the clinical examination, the patient's orofacial symptoms should be assessed in a standardized and regular manner, as an important part of monitoring the TMJs and orofacial health (12).

Historically, physicians have based their overall assessment of children's symptoms mainly on parental

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proxy reports. However, over the past 30 years, increased attention has been paid to the importance of letting children articulate their own perceived health status, communicate their own experiences of symptoms, and participate in health decisions (13). A child's capability to communicate these feelings is closely tied to their cognitive growth. It is therefore essential to employ tools suitable for the child's age when collecting this self-reported information (14). Research concerning the consensus between children and their parents in health-related matters shows moderate to poor consensus on the reporting of symptoms (14, 15). Two Scandinavian studies on childhood non-JIA-related pain revealed that parental proxy reporting may be associated with insecurity of the actual pain experiences of the child (16, 17). In adolescents with JIA, Shaw et al found a wide variation in agreement between the adolescent and parental proxy reports, according to pain and general well-being (18).

A consensus-based patient questionnaire was recently developed and validated by the Temporomandibular Joint Juvenile Arthritis Working Group (TMJaw) for the assessment of orofacial symptoms in subjects with JIA aged 10–18 years (19). In the process of adapting this standardized questionnaire to a younger age group, the current study aims to assess the agreement between parental proxy-reported orofacial symptoms and pain and children's self-assessment in the Danish version of the patient questionnaire Assessment of Orofacial Symptoms in Juvenile Idiopathic Arthritis.

Method

Study design and setting

This cross-sectional study was conducted at the Section of Orthodontics, Department of Dentistry and Oral Health, Aarhus University, Denmark, in March 2023. The terminology adheres to JIA-TMJaw consensus-based standardized terminology (20). We identified eligible candidates from consecutive subjects with JIA and TMJ involvement attending an appointment at the Regional Craniofacial Clinic, Section of Orthodontics, accompanied by at least one of their parental proxies. Those candidates were diagnosed by a paediatric rheumatologist at the Aarhus University Hospital according to the International League of Associations for Rheumatology (ILAR) JIA criteria, with TMJ involvement previously confirmed by either magnetic resonance imaging or cone beam computed tomography. Eligible subjects and their parent(s) were invited to participate in this study. The child and the parental proxy completed the questionnaire on orofacial symptoms in JIA separately, without any communication between them during the completion (19).

Inclusion criteria were subjects (i) with a diagnosis of JIA according to the ILAR criteria (1); (ii) with

involvement of one or both TMJs, in agreement with the consensus-based definition of 'TMJ involvement' (20); (iii) 10–18 years of age; and (iv) accompanied by at least one parental proxy at the agreed appointment.

Exclusion criteria were dyads with a child or parental proxy who was (i) not fluent in the Danish language or (ii) unable to understand the context of the questionnaire questions; or (iii) dyads that were observed interacting while completing the questionnaire. The absence of orofacial symptoms was not an exclusion criterion as we found it important to also include subjects without orofacial symptoms.

Questionnaire

We used the Danish version of the TMJaw questionnaire, which is standardized and has been cross-culturally validated with the English version (19). The questionnaire relates to the multidimensional aspects of JIA-related symptoms within the past 2 weeks. The questionnaire contains seven items, where subjects are asked to report on orofacial pain frequency, intensity, pain location, functional disability, presence of specific symptoms related to TMJ involvement, and changes in pain and functioning since the last visit (Table 1).

The English version of the full questionnaire is available at <https://www.jrheum.org/content/50/5/676.tab-supplemental> as open access (19).

Ethical approval

The project was conducted in accordance with Danish ethical guidelines for questionnaire surveys. Patients and parents provided written informed consent before inclusion in the study. For patients below the age of 15 years, written consent was granted by the parents. The project was registered with the Danish Data Protection Agency through Aarhus University's internal reporting system (no 2022-0367531.3144).

Statistical analysis

Statistical analyses were conducted using STATA version 18 software (STATA Corp, College Station, TX, USA). For nominal and ordinal outcome variables, the level of agreement between the child and proxy reports was estimated using Cohen's kappa and weighted Cohen's kappa coefficients, respectively, with benchmark agreements of < 0.00 poor, 0.00–0.20 slight, 0.21–0.40 fair, 0.41–0.60 moderate, 0.61–0.80 substantial, and 0.81–1.00 almost perfect (21). To evaluate the child-proxy agreement on the visual analogue scale (VAS) for orofacial pain and functional disability, we used the intraclass correlation coefficient (ICC) estimates and their 95% confidence intervals (CIs), based

Table 1. Overview of the items in the questionnaire.

Questionnaire item	Assessment for outcome
1. Pain frequency?	Five ordinal outcomes: 0: Never 1: Less than once a week 2: Several times a week 3: Several times a day 4: All the time
2. Pain intensity?	VAS 0–100 mm (0 = no pain, 100 = worst possible pain)
3. Pain location?	Patient identification of pain locations on face map
4. Jaw function?	VAS 0–100 mm (0 = not affected, 100 = severely affected)
5. Presence of specific symptoms?	Seven questions (dichotomous outcomes, 'yes'/'no'): 1: 'I felt pain when I chewed' 2: 'I avoided hard or chewy foods because it hurt my face or jaw' 3: 'I felt pain when I opened my mouth wide (e.g. yawning)' 4: 'I felt stiffness in my jaw muscles in the morning' 5: 'I felt that my jaw got stuck in the open or closed position' 6: 'I felt pain in my jaw when I talked for a long time' 7: 'I felt clicking or popping from my jaw when I opened my mouth'
6. Changes in face and jaw pain?	Four nominal outcomes: 1: No change 2: Improved (less pain) 3: Worse (more pain) 4: Cannot remember, not applicable
7. Changes in jaw function?	Four nominal outcomes: 1: No change 2: Improved (less pain) 3: Worse (more pain) 4: Cannot remember, not applicable

VAS, visual analogue scale.

on a mean-rating ($k = 2$), absolute-agreement, one-way random-effects model. The reliability, based on the 95% CI of the ICC estimate, was considered to be poor < 0.5 , moderate 0.5–0.75, good 0.75–0.9, and excellent > 0.9 (22). Additional Bland–Altman plots were applied to the VAS scores to explore the concordance between children and their parental proxies. The difference between the child and proxy ratings for VAS on the y-axis was plotted against the mean of the child and proxy VAS rating on the x-axis (23).

Results

We invited 35 consecutive child–proxy dyads to participate in the study, and all dyads agreed to join. One dyad was excluded because the child collaborated with their parental proxy while completing the questionnaire. The characteristics of the 34 included dyads are presented in Table 2. None of the participants was on systemic steroids or had recently received intra-articular steroid injections in the TMJs.

Orofacial pain

Pain in the child's face or jaw in the past 2 weeks was reported by 61.8% of children and 41.2% of the parental

proxies. There was moderate agreement between children and proxies (Cohen's weighted kappa $\kappa = 0.42$, $p = 0.002$). After dividing the cohort into age groups, we found a slight reporting agreement for children aged < 13 years and their proxies ($n = 16$, weighted $\kappa = 0.16$, $p = 0.465$). There was substantial agreement for the group of children aged ≥ 13 years ($n = 18$, weighted $\kappa = 0.63$, $p < 0.001$) (Table 3).

Pain intensity

Pain intensity was reported by 34 children and 33 parental proxies, and the dyad with a missing proxy score was excluded from the ICC analysis. On average, children reported a marginally significantly higher pain intensity compared to the assessment provided by the parental proxies ($p = 0.005$). The ICC for pain intensity (ICC = 0.604; 95% CI 0.205–0.804) showed poor to good agreement for the total group of children and their proxies (Table 4). After dividing subjects into subgroups according to age, we found a poor to moderate agreement between children < 13 years of age and their parental proxies, and moderate to excellent agreement for children aged ≥ 13 years and their proxies. The Bland–Altman plots showed a higher average bias and a wider range between the upper and lower limit of agreement for younger children and their proxies

Table 2. Distribution of characteristics for the 34 participants divided by age.

Characteristic	All subjects (N = 34)	< 13 years (n = 16)	≥ 13 years (n = 18)
Age at visit (years)	13 (11–14)	11 (10.5–12)	14 (14–16)
Gender of child, female	27 (79.4)	13 (81.3)	14 (77.8)
Gender of parental proxy, female	23 (67.6)	11 (68.8)	12 (66.7)
Age at JIA onset (years)	5.5 (2–10)	5 (2–6.5)	9.5 (3–12)
Disease duration (years)	6 (3–10)	6 (4–9.5)	5.5 (3–12)
JIA category			
Oligoarthritis persistent	17 (50.0)	7 (43.8)	10 (55.6)
Oligoarthritis extended	7 (20.6)	4 (25.0)	3 (16.7)
Polyarthritis RF ⁻	7 (20.6)	4 (25.0)	3 (16.7)
Psoriatic arthritis	2 (5.9)	1 (6.2)	1 (5.5)
Enthesitis-related arthritis	1 (2.9)	0	1 (5.5)
No DMARDs	8 (23.5)	4 (25.0)	4 (22.2)
sDMARDs only	4 (11.8)	2 (12.5)	2 (11.1)
bDMARDs*	22 (64.7)	10 (62.5)	12 (66.7)

Data are shown as median (interquartile range) or n (%).

*bDMARDs alone or in combination with sDMARDs.

JIA, juvenile idiopathic arthritis; RF⁻, rheumatoid factor negative; DMARD, disease-modifying anti-rheumatic drug; sDMARD, synthetic disease-modifying anti-rheumatic drug; bDMARD, biological disease-modifying anti-rheumatic drug.

Table 3. Orofacial pain frequency according to rater (child, proxy) and age group.

Pain frequency	All subjects (N = 34)		< 13 years (n = 16)		≥ 13 years (n = 18)	
	Child	Proxy	Child	Proxy	Child	Proxy
Never	13 (38.2)	20 (58.8)	6 (37.5)	9 (56.3)	7 (38.9)	11 (61.1)
Less than once a week	10 (29.4)	6 (17.7)	6 (37.5)	3 (18.7)	4 (22.2)	3 (16.6)
Several times a week	6 (17.7)	5 (14.7)	1 (6.3)	3 (18.7)	5 (27.8)	2 (11.1)
Several times a day	5 (14.7)	2 (5.9)	3 (18.7)	1 (6.3)	2 (11.1)	1 (5.6)
All the time		1 (2.9)	0	0	0	1 (5.6)
Agreement						
Weighted κ-value	0.42		0.16		0.63	
p-Value	0.002		0.465		< 0.001	
95% CI	0.16 to 0.68		-0.30 to 0.62		0.35 to 0.92	

Data are shown as n (%).

κ, Cohen's kappa; CI, confidence interval.

Table 4. Pain intensity according to age group and rater (child, proxy).

	Child		Proxy		ICC [†]	95% CI [†]		Sig
	n	Median (IQR)*	n	Median (IQR)*		Lower bound	Upper bound	
All	34	14.5 (0–28)	33	0 (0–20)	0.604 [‡]	0.205	0.804	0.005
< 13 years	16	15.5 (0–39)	16	2.5 (0–23.5)	0.250	-0.353	0.582	0.287
≥ 13 years	18	14 (0–28)	17	0 (0–20)	0.887 [‡]	0.695	0.959	0.000

*Pain intensity score on the visual analogue scale (0–100).

[†]ICC estimates and 95% CIs based on mean-rating (k = 2), absolute-agreement, one-way random-effects model.

[‡]The dyad with a missing proxy report for pain intensity was excluded from the ICC analysis.

ICC, intraclass correlation coefficient; CI, confidence interval; Sig, significance; IQR, interquartile range.

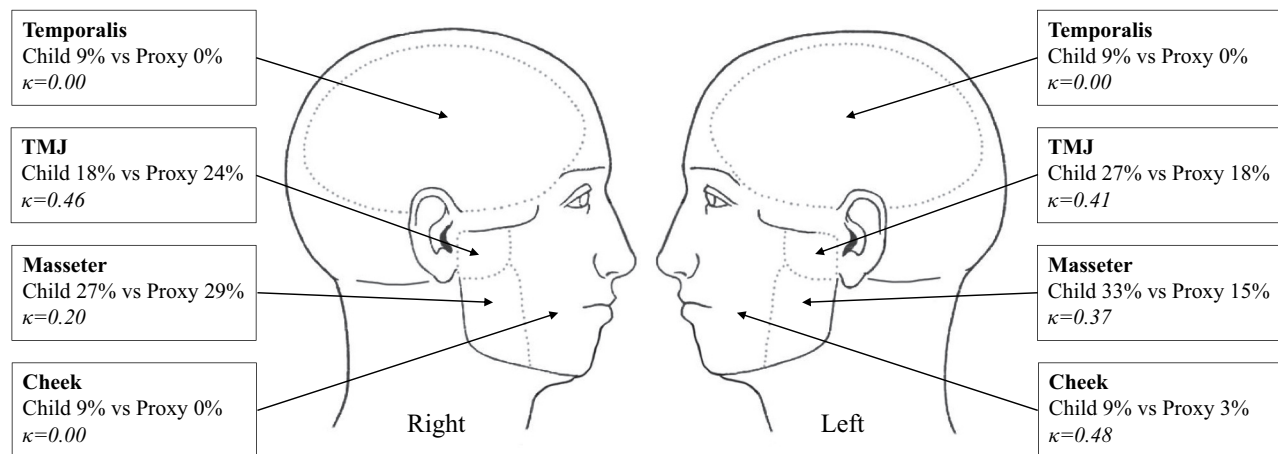


Figure 1. Pain location according to children and parental proxies in the 34 dyads, κ , Cohen's kappa; TMJ, temporomandibular joint.

compared to the older group (online supplemental material, Additional file 1, Figure 1).

Pain locations

The pain locations are presented in Figure 1. According to both the children and the proxy reports, the most frequently reported site of pain was the masseter area, followed by the TMJ. The best agreement for pain locations was 'moderate' agreement, obtained for the left cheek ($\kappa = 0.48$, $p < 0.001$) and the TMJs (right TMJ $\kappa = 0.46$, $p = 0.003$ and left TMJ $\kappa = 0.41$, $p = 0.007$).

Jaw function

JIA-related TMJ dysfunction within the past 2 weeks was reported by 29.4% of the children and 26.5% of the parental proxies (Supplemental material, Additional file 2, Table 1). In six of the dyads, both the child and the proxy reported impaired jaw function

($\kappa = 0.49$, $p = 0.007$) (online supplemental material, Additional file 2, Table 1). The ICCs for functional disability in the jaws had wide 95% CIs with lower bounds beneath 0.5 (online supplemental material, Additional file 2, Table 2).

Orofacial symptoms

The frequency of orofacial symptoms and the child-proxy agreement are presented in Table 5. Clicking or popping from the jaw was the most frequent proxy- and self-reported symptom, with fair agreement between children < 13 years of age and their parental proxies ($\kappa = 0.26$, $p = 0.124$) and 'almost perfect' agreement for children ≥ 13 years of age ($\kappa = 1.00$, $p < 0.001$).

Changes since the last visit

Most parental proxies (78.1%) and less than half of the children (45.2%) reported no change in the child's face and jaw pain since the last visit (online supplemental

Table 5. Orofacial symptoms according to rater (child, proxy) and separated by age group.

Symptom	All subjects			< 13 years			≥ 13 years		
	Child (N = 34)	Proxy (N = 33)	κ^*	Child (n = 16)	Proxy (n = 16)	κ	Child (n = 18)	Proxy (n = 17)	κ^*
Pain chewing	7 (20.6)	4 (12.1)	0.46	5 (31)	3 (19)	0.35	2 (11)	1 (6)	0.64
Avoiding hard/chewy foods	4 (11.8)	2 (6.1)	0.27	1 (6)	1 (6)	-0.07	3 (17)	1 (6)	0.45
Pain opening wide	10 (29.4)	8 (24.2)	0.39	3 (19)	3 (19)	0.18	7 (39)	5 (29)	0.49
Morning stiffness jaw	8 (23.4)	1 (3.0)	0.18	2 (13)	0	0.00	6 (33)	1 (6)	0.21
Jaw getting stuck	3 (8.8)	0	0.00	1 (6)	0	0.00	2 (11)	0	0.00
Pain talking	4 (11.8)	2 (6.1)	0.27	2 (13)	1 (6)	-0.09	2 (11)	1 (6)	0.64
Clicking or popping	12 (35.3)	9 (27.3)	0.65	6 (38)	3 (19)	0.26	6 (33)	6 (35)	1.00

Data are shown as n (%).

*The dyad with a missing proxy report for symptoms was excluded from the kappa analysis.

κ , Cohen's kappa.

material, Additional file 3, Table 1). In total, 45.2% of the children reported less pain than at the last visit and 9.6% of the children reported more pain. According to the proxies, 12.5% and 9.4% of the children had less or more pain, respectively.

Regarding jaw function, 70.0% of the children and 80.7% of the parental proxies reported no changes in jaw function since the last visit, while 30.0% of the children and 12.9% of the proxies reported improvement (online supplemental material, Additional file 3, Table 2). In two dyads (6.4%), the parental proxies rated jaw function as worsened.

There was slight agreement on changes in jaw pain (weighted $\kappa = 0.11$, $p = 0.337$) and function (weighted $\kappa = 0.12$, $p = 0.490$) (online supplemental material, Additional file 3, Tables 1 and 2).

Discussion

Orofacial symptoms are a prevalent finding in subjects with JIA (8, 24). Regular assessment of these symptoms is an important part of the general monitoring of these patients (12). Our study found that the overall agreement between child self-report and parent-proxy report of orofacial symptoms was 'poor' to 'moderate' for the 34 included subjects and their parental proxies. After subgrouping by age, children ≥ 13 years of age had better agreement with their parents than the younger subgroup < 13 years of age. Children reported pain and functional disability of the jaw more often than their parental proxies, and reported higher pain intensity and higher dysfunction of the jaw than their proxies. This indicates that the child self-report and parental proxy report are not easily interchangeable.

To our knowledge, this is the first study to compare the agreement between child self-report and parent-proxy report on JIA-related orofacial symptoms in children. Several studies have assessed the child–parent agreement on general pain and well-being in children and adolescents with JIA (25–28). Similarly to our findings on TMJ pain frequency, Palermo et al found 'fair' child–parent reporting agreement on general pain frequency in a study on 63 children 8–16 years of age with JIA (25). Garcia-Munitis et al studied the child–parent reporting agreement in 94 5–18-year-old children with JIA and found a 'moderate' agreement on general pain intensity, with lower pain ratings in children compared to the rating of their parents (26). This contrasts with our findings on TMJ pain intensity, where both children (< 13 years) and adolescents (≥ 13 years) reported higher levels of pain than their parental proxies. A tendency for the parents to underestimate their children's general pain intensity was also found by Lal et al in a study on 204 adolescents with JIA, with 'moderate' agreement on pain intensity (27). Vanoni et al found substantial agreement between children's

and parents' median scores for the quantitative items of the Juvenile Arthritis Multidimensional Assessment Report, but with a low agreement for the individual dyads; however, they had a substantially higher number of participants than in our study (28).

A study by Schwartz et al on mother–child concordance in a paediatric chronic non-JIA pain sample found 'moderate to better' agreement for about half of the areas assessed using a standardized body map, with 'moderate' agreement for the head area (29). In our study, children and parents marked the location of pain on a figure of the head, and the highest achieved agreement was 'moderate' agreement for the left cheek and the TMJs. For the temporalis region, there was no agreement between the parents and the children. Children reported more pain regions than the parental proxies; therefore, our results indicate that the assessment of the pain location based only on parental proxy reports would lead to an underreporting of pain locations.

In a review that assessed the child–parent agreement in studies on children with health issues or functional limitations, Hemmingsson et al found better agreement on observable, external symptoms than on non-observable symptoms such as feelings and emotions (15). This may explain the findings in our study, with better agreement on symptoms that the parents can hear or observe during a shared meal, such as clicking or popping of the TMJ, pain chewing, and avoiding hard/chewy foods, in contrast to symptoms that are not as easily observable, such as morning stiffness in the jaw or the subjective feeling of the jaw getting stuck. However, in the group of younger children, the agreement on clicking and popping of the jaw was only fair and not statistically significant. Owing to the small sample size of the present study, we only have a few observations on specific symptoms, which makes it difficult to generalize our findings in this aspect.

The agreement on changes in orofacial pain and jaw function since the last visit was poor and the Z-statistic was not significant, indicating no better agreement than by chance alone. From a clinical point of view, this illustrates the importance of applying caution when asking about changes in orofacial symptoms since last time. The time interval since the last visit and the parent following the child to the last visit was not assessed and the number of participants was low, making it difficult to draw valid conclusions regarding agreement on changes in pain and function.

The results of our study suggest that parental proxy reports of orofacial symptoms in JIA alone, especially in children < 13 years of age, may not be sufficient when collecting comprehensive reports for clinical settings or research studies. We therefore suggest letting children < 13 years of age complete

the questionnaire together with their parental proxy. This may not only provide the practitioner with a more comprehensive symptom depiction but also give parents a better understanding of their child's current orofacial symptoms. The best child–parent agreement for younger children was a 'fair' agreement on pain when chewing. In older children, the best child–parent agreement was almost perfect agreement on clicking or popping and substantial agreement on pain chewing and talking.

There are important strengths and limitations to this study that need consideration. Limitations to the present study are (i) the small number of participants, which makes it difficult to subgroup some items for analysis; and (ii) missing information on participants' disease activity in terms of the 71- or 27-joint Juvenile Arthritis Disease Activity Score (JADAS-71 or JADAS-27), and other disease descriptors such as the presence of antinuclear antibodies.

Important strengths of the present study are that (i) the analysis of the agreement between children and proxies was conducted in a group of consecutive patients; (ii) the dyads were first informed about the study on the day of the invitation and participation, leaving no time for the child to discuss or agree on orofacial symptoms with the parental proxy beforehand; and (iii) the questionnaire was completed under supervision, preventing interaction between child and proxy, and exclusion of dyads where interaction took place.

Conclusion

There is an overall 'poor to moderate' child–proxy reporting agreement on the questionnaire for assessment of JIA-related orofacial symptoms. After subgrouping, we found a higher child–proxy agreement in adolescents ≥ 13 years of age compared to children < 13 years of age. Collecting information only from the parental proxy could lead to an underestimation of the reported JIA-related orofacial symptoms. To collect a comprehensive report, we suggest including child ratings when assessing orofacial pain and symptoms in children aged < 13 years.

Authors' contributions

All authors were involved in revising the article critically and approved the final version to be published. PS and MT contributed to the design and conception of the study. JMH, TKP, and PS were responsible for the acquisition of data. JMH performed the analyses and wrote the manuscript together with PS and EBN. JMH, TKP, EBN, MT, and PS carried out this study on behalf of the Temporomandibular Joint Juvenile Arthritis Working Group (TMJaw).

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Disclosure statement

No potential conflict of interest was reported by the author(s).

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Data availability statement

The data sets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

Supplementary material

Supplemental data for this article can be accessed online at <https://doi.org/10.1080/03009742.2024.2412459>

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