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The reliability and validity of the juvenile idiopathic arthritis magnetic resonance scoring system for temporomandibular joints

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ABSTRACT

In children with juvenile idiopathic arthritis (JIA), the temporomandibular joint (TMJ) can be involved. To prevent TMJ damage due to inflammation, early recognition is important, for which contrast-enhanced magnetic resonance imaging (MRI) is the gold standard. In this study, the interobserver reliability and construct validity of the Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints (JAMRIS-TMJ) was assessed. Two radiologists independently examined 38 MRIs using the JAMRIS-TMJ scoring system. Inter-observer reliability was assessed by Cohen's (weighted) kappa (κ), 95% confidence intervals (CIs) and absolute agreement (%). Construct validity was assessed by correlation between the JAMRIS-TMJ items and TMJ involvement, active maximum interincisal mouth opening (AMIO), and anterior maximum voluntary bite force (AMVBF). The interobserver reliability for the JAMRIS-TMJ items varied from poor to good ($\kappa = 0.18-0.61$). Joint enhancement had the highest reliability ($\kappa = 0.61$). Correlations were found between TMJ involvement, AMIO, and the JAMRIS-TMJ items, although variation between radiologists and TMJ side existed. No correlation was found between AMVBF and the JAMRIS-TMJ items for both radiologists. The strongest correlations were found between most of the JAMRIS-TMJ items and AMIO. Our findings support the utility of AMIO as a clinical measure of TMJ status in children with JIA.

1. Introduction

In children with juvenile idiopathic arthritis (JIA) the temporomandibular joint (TMJ) can be involved. TMJ involvement can present symptoms such as pain, a lower health-related quality of life, a limited mouth opening, and facial asymmetry due to growth disturbances (Frid et al., 2017; Stoustrup et al., 2017). To prevent these symptoms, early recognition of TMJ arthritis is important. Different diagnostic methods are available to indicate TMJ abnormalities in children with JIA, such as clinical assessment, ultrasound, cone beam computed tomography (CBCT), panoramic radiographs, or magnetic resonance imaging (MRI).

Overall, contrast-enhanced MRI is advised as the gold standard method to detect TMJ arthritis (Kristensen et al., 2016; Stoustrup et al., 2019). For proper interpretation of MRIs of TMJ in JIA, a standardized scoring system is required (Tolend et al., 2018). In 2018, a structured scoring system was proposed by Outcome Measures in Rheumatology and Clinical Trials (OMERACT) and Juvenile Idiopathic Arthritis MRI (JAMRI) working group (Kellenberger et al., 2015; Tolend et al., 2018). This Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints (JAMRIS-TMJ) contains an inflammatory and a damage-related domain. However, difficulties in interpreting and scoring the MRIs have been described (Malattia et al., 2020;

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Angenete et al., 2021; Tolend et al., 2021). The interobserver reliability of the JAMRIS-TMJ scoring items is reported to vary widely. In the original article of the JAMRIS-TMJ scoring system, the interobserver reliability was reported as sufficiently reliable, with JAMRIS-TMJ items average-measure absolute agreement intraclass correlation coefficients (avICCs) that varied between 0.57 and 0.95 (Tolend et al., 2018), while other more recent studies showed less reliability for items in the JAMRIS-TMJ scoring system (Angenete et al., 2021; Tolend et al., 2021). Therefore, we hypothesized that the reliability of the JAMRIS-TMJ scoring system in our study will demonstrate lower reliability compared to the most recent studies.

Another diagnostic method to detect TMJ involvement in patients with JIA is the clinical examination. Alternative diagnostic methods are desirable because of the disadvantages of MRI, such as the need for contrast infusion, the concern with contrast retention in the (young) human brain, need for sedation in case of anxiety or claustrophobia, high costs, and limited availability and expertise (Elbeshlawi and AbdelBaki 2018; Hechler et al., 2018). Especially in young children, the need for infusion and/or sedation in case of anxiety or claustrophobia can be a burden on the child and the parents. These aspects may contribute to a delay in the recognition of TMJ problems in JIA (Ma et al., 2022). The clinical examination of TMJ is a simple method and is able to detect and monitor TMJ involvement (Stoustrup et al., 2017). For example, the TMJ screening protocol score is a clinical screening tool consisting of history, examination, and inspection domains (Steenks 2015). A TMJ screening protocol score ≥ 2 is operationally defined as TMJ involvement. A limited mouth opening is highly prevalent in children with JIA and TMJ arthritis (Abramowicz et al., 2013; Zwir et al., 2015; Kristensen et al., 2016; Stoustrup et al., 2017; Scolozzi et al., 2022). We hypothesized the strongest correlation between AMIO and the JAMRIS-TMJ scoring system, since mouth opening capacity is one of the most used clinical measurements in TMJ examination in patients with JIA (Stoustrup et al., 2017). A reduced bite force has been reported in children with JIA and TMJ involvement, as well (de Sonnaville et al., 2021). However, no studies focused on bite force and its association with MRI-confirmed TMJ arthritis.

Therefore, the aim of this study is to evaluate the interobserver reliability of the JAMRIS-TMJ scoring system in children with JIA, and to determine the construct validity of the JAMRIS-TMJ scoring system versus TMJ involvement, active maximum interincisal opening (AMIO), and active maximum voluntary bite force (AMVBF).

2. Materials and methods

In this cross-sectional study, children with JIA were recruited and assessed at the outpatient clinic of the Department of Pediatric Immunology and Rheumatology of University Medical Center Utrecht (UMCU), in collaboration with the Department of Oral and Maxillofacial Surgery and Special Dental Care of the UMCU, between January 2018 and May 2020. The inclusion criteria for participation were children with JIA, as classified using the International League of Associations for Rheumatology (ILAR) criteria, and an age between 6 and 18 years old. The exclusion criteria were: 1) a history of mandibular trauma; 2) an additional orofacial condition not related with JIA (e.g., dental pain or a pre-existing jaw or temporomandibular disorder); 3) undergoing orthodontic treatment; 4) a contraindication for MRI; 5) a need for sedation prior to MRI (e.g., as a result of claustrophobia) burden METC; and 6) more than 6 weeks between the clinical examination and MRI, as therapeutic treatment response is expected after 6–8 weeks (Swart et al., 2013; Ferrara et al., 2018). The medical ethical committee of the UMC Utrecht gave approval for this study (ID: NL.METC-17-528/C and NL.METC.17/704/M).

The following data were extracted from the electronic medical records for the included children with JIA: 1) JIA subtype, 2) date of JIA diagnosis, 3) medication, 4) height, 5) weight, 6) gender, 7) age, 8) the presence of antinuclear antibody (ANA) 9) rheumatoid factor (RF), 10)

human leukocyte antigen B27 (HLA-B27), and 11) the clinical Juvenile Arthritis Disease Activity Score (cJADAS) (Consolaro et al., 2014). Data collection was performed using the good clinical practice (GCP) compliant electronic data capture (EDC) system, Research Online, owned by the Julius Center (UMC Utrecht).

Children with JIA were routinely screened at the outpatient clinic for AMIO, AMVBF, and the TMJ screening protocol score (Steenks et al., 2015). The children and their parents were informed about the study in advance. Informed consent was obtained by parents or legal guardians and, depending on age, also by the child prior to this study, and an MRI examination of the TMJs was planned within 6 weeks.

2.1. Magnetic resonance imaging

The MRI was performed on a 3 T system (Signa MR/i Twinspeed scanner, GE Medical Systems, Milwaukee, WI, USA). The total time of investigation was around 30 min. In order to optimally compare the various sequences, the patient was positioned supine in the MRI system, with closed mouth, and asked not to move the head or open the mouth during the examination. After the scout scans, used to identify the joints, fat saturated (FS) T2 weighted (T2W) images (to identify bone marrow edema and joint fluid) and FS T1W images with contrast (to identify synovial enhancement and thickening) were made, according to the TMJ/MRI protocol.

TMJ/MRI protocol: head sense coil, extra surveys coronal (c) and transversal (t) are added to locate the TMJ. Angulations of the sagittal(s) and coronal images are made: 1) multiple stack survey; 2) s T1W spin echo (SE) image whole head; 3) c survey; 4) t survey; 5) s T1W image (3 mm slice thickness, no gap) through both TM joints; 6) s T2W 3 mm short-TI inversion recovery (STIR) image through both TM joints. Injection of Gadolinium (gd) intravenous; 7) s T1W 3 mm spectral pre-saturation with inversion recovery (SPIR) gd through both TM joints; 8) c T1W 3 mm SPIR gd through both TM joints.

2.2. JAMRIS-TMJ scoring system

The MRIs were independently assessed by two radiologists (StH, FN) experienced in pediatric musculoskeletal imaging. They followed the scoring system proposed by the OMERACT and JAMRI working groups (Kellenberger et al., 2015; Tolend et al., 2018). The scoring system construct of the inflammatory domain consists of the items: 1) bone marrow edema (grade 0–1); 2) bone marrow enhancement (grade 0–1); 3) joint effusion (grade 0–2); 4) synovial thickening (grade 0–2); and 5) joint enhancement (grade 0–2). The damage domain consists of the items: 1) condylar flattening (grade 0–2); 2) erosions (grade 0–2); and 3) disk abnormalities (grade 0–1). The first five MRIs were independently assessed by the two radiologists, and afterwards the radiologists compared their results. This procedure was used to discuss some difficulties in the rating process. The MRIs were blinded and displayed in a random order by using the Picture Archiving and Communication System (PACS) software tools (IDS7, Sectra Medical Systems, Linköping, Sweden).

2.3. TMJ screening protocol

The TMJ screening protocol consists of 11 items regarding history, examination, and inspection (Steenks et al., 2015) The history items addressed: 1) problems in chewing; 2) eating slower than others; 3) difficulty in biting hard food; 4) pain while eating; and 5) a limited mouth opening. The clinical examination items of the TMJ screening protocol addressed: 6) limited mouth opening, the cut-off value for which was ≤ 40 mm with exclusion of the overbite; 7) crepitation on mandibular opening and closing; 8) pain on AMIO; and 9) left or right mandibular midline deviation on opening wide. The inspection items of the TMJ screening protocol addressed: 10) facial asymmetry; and 11) retrognathia.

Each positive item of the TMJ screening protocol receives 1 point; negative scoring items receive 0 points. All positive items produced the TMJ screening protocol score. The TMJ screening protocol has been validated against the disease activity score (JADAS-27) (Steenks et al., 2015). A TMJ screening protocol score of at least 2 was found to have the highest sensitivity and specificity to differentiate between a low disease activity score (JADAS-27 of 2.2 or lower) and a high disease activity score (JADAS-27 of 6.4 or higher). Following our protocol, in this study we operationally defined TMJ involvement as a TMJ screening protocol score ≥ 2 .

2.4. Active maximum interincisal mouth opening (AMIO)

The AMIO was measured with a metal ruler to the nearest millimeter. During the AMIO measurement, the participants were asked to open their mouth as wide as possible. The distance between the upper central incisor and the lower central incisor was recorded with a ruler. The overbite was not included in the AMIO values.

2.5. Active maximum voluntary bite force (AMVBF)

The AMVBF was measured using a bite force transducer (de Sonnaville et al., 2021). The transducer was placed between the upper and lower central incisors. The bite force measurement consists of clenching as hard as possible for 10 s at maximum strength. Three attempts were documented. The highest bite force of the three attempts was defined as the AMVBF and is expressed in Newtons (N). A good to excellent reliability of AMVBF measurements was found for children with JIA (de Sonnaville et al., 2023).

2.6. Statistical analysis

Characteristics of the children were presented as numbers and percentages, and as means and standard deviations (SD). Inter-observer reliability was assessed for the items of the inflammatory and damage domains of the JAMRIS-TMJ scoring system and analyzed by Cohen's (weighted) kappa coefficient (κ) and reported with 95% confidence intervals (CIs). Cohen's kappa coefficients were calculated for bone marrow edema, bone marrow enhancement, and disk abnormalities. Weighted kappa coefficients (κ_w) were calculated for joint effusion, synovial thickening, joint enhancement, condylar flattening, and erosions. A kappa score smaller than 0.2 was considered poor, 0.21 to 0.40 as fair, 0.41 to 0.60 as moderate, 0.61 to 0.80 as good, and 0.81 to 1.00 as very good (Cohen 1960). In addition, the absolute agreement (%) was calculated for each item of the inflammatory and damage domains.

Construct validity of the JAMRIS-TMJ scoring was assessed by Spearman's rank correlation coefficient (r_s), because of the ordinal origin of the JAMRIS-TMJ outcomes. The Spearman's correlation was run to determine the concurrent validity by testing the extent of relationship between the outcomes of the JAMRIS-TMJ scoring items (bone marrow edema, bone marrow enhancement, joint effusion, synovial thickening, joint enhancement, condylar flattening, erosions, and disk abnormalities) and TMJ involvement, AMIO, and AMVBF. Correlations smaller than 0.19 were considered as very weak, 0.20 to 0.39 as weak, 0.40 to 0.59 as moderate, 0.60 to 0.79 as strong, and 0.80 to 1.00 as very strong (Swinscow and Campbell 1997). We hypothesized a positive correlation between TMJ involvement and the JAMRIS-TMJ items, with the term TMJ involvement defined as: 'clinical situations in which no contrast-enhanced MRI verification of active TMJ inflammation has occurred but where signs, symptoms, and/or radiological findings suggest the presence of actual or former TMJ arthritis' (Stoustrup et al., 2019). We expected a negative correlation between AMIO and the JAMRIS-TMJ items, as a lower AMIO was correlated with signs of TMJ arthritis on MRI (Zwir et al., 2015). We also hypothesized a negative correlation between AMVBF and the JAMRIS-TMJ items, as a lower AMVBF is found in children with JIA and TMJ involvement compared to children with JIA

without TMJ involvement (de Sonnaville et al., 2021). Since the construct of the JAMRIS-TMJ items is related but dissimilar, a correlation between 0.30 and 0.50 is expected (Mokkink et al., 2018). A p-value of less than 0.05 was considered statistically significant. Statistical analyses were performed using SPSS 25 (IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp).

3. Results

A total of 38 children with JIA were analyzed in this study, 31 with clinically established TMJ involvement (TMJ screening protocol score ≥ 2) and 7 children with a TMJ screening protocol score of 0. There were 31 girls (81.6%) and 7 boys (18.4%), with a mean age of 14.6 years (Table 1). The JIA subtype RF- polyarticular (39.5%) was most common, followed by persistent oligoarticular (21.1%) and extended oligoarticular (18.4%). The cJADAS score was low in 15 children with JIA (39.5%), moderate in 11 (28.9%), and high in nine (23.7%). The cJADAS was missing in three children.

Of the history items of the TMJ protocol score, 24 (77.4%) children

Table 1
Demographics, clinical characteristics.

Variable	N = 38
Gender, n (%)	
Male	7 (18.4)
Female	31 (81.6)
Mean age (years; mean, SD)	14.6 (3.5)
Mean weight (kg; mean, SD)	57.8 (17.8)
Mean height (cm; mean, SD)	161.1 (15.1)
JIA subtype, n (%)	
Systemic	1 (2.6)
Persistent oligoarticular	8 (21.1)
Extended oligoarticular	7 (18.4)
RF- polyarticular	15 (39.5)
RF + polyarticular	7 (18.4)
Enthesitis-related	2 (5.3)
Arthritis Psoriatica	3 (7.9)
Mean disease duration (months)	71.1 (61.7)
cJADAS, n (%)	
0–2 (low)	15 (39.5)
3–7 (moderate)	11 (28.9)
≥ 8 high	9 (23.7)
Missing	3 (7.9)
Current medication use, n (%)	
NSAIDs	16 (42.1)
Corticosteroid	1 (2.6)
csDMARDs	19 (50.0)
bDMARDs	15 (39.5)
No medication	6 (15.8)
csDMARDs, n (%)	
Methotrexate	14 (36.8)
Leflunomide	3 (7.9)
Azathioprine	1 (2.6)
Sulfasalazine	1 (2.6)
No DMARDs	19 (50.0)
bDMARDs, n (%)	
Adalimumab	10 (26.3)
Etanercept	4 (10.5)
Golimumab	1 (2.6)
No bDMARDs	23 (60.5)
TMJ screening protocol score < 2 (n, %)	7 (18.4)
TMJ screening protocol score ≥ 2 (n, %)	31 (81.6)
Mean AMVBF (Newton; mean, SD)	112.6 (54.3)
Mean AMIO (mm; mean, SD)	39.2 (10.2)

AMIO: active maximum interincisal mouth opening; AMVBF: anterior voluntary maximum bite force; bDMARDs: biological disease-modifying anti-rheumatic drugs; cJADAS: clinical juvenile arthritis disease activity score; csDMARDs: conventional synthetic disease-modifying anti-rheumatic drugs; JIA: juvenile idiopathic arthritis; NSAIDs: non-steroidal anti-inflammatory drugs; TMJ: temporomandibular joint.

TMJ involvement was defined as a TMJ screening protocol score ≥ 2 (Steenks et al., 2015).

with JIA had pain while eating, 23 (74.2%) reported biting hard food was difficult, and 20 (64.5%) had problems in chewing (Table 2). Of the examination items, 19 (61.3%) children with JIA had a limited mouth opening, 16 (51.6%) had pain during AMIO, 15 (48.4%) had deviation during AMIO, and nine (29.0%) had crepitation. Of the inspection items, 16 (51.6%) children with JIA had asymmetry and five (16.1%) had retrognathia.

Table 3 presents the items of the JAMRIS-TMJ scoring system, scored by the two independent radiologists for both left and right TMJs. The interobserver reliability of the inflammatory domain of the JAMRIS-TMJ scoring system was poor for bone marrow edema ($\kappa = 0.12$, 95% CI: 0.04–0.20) and bone marrow enhancement ($\kappa = 0.18$, 95%CI: 0.07–0.29), fair for joint effusion ($\kappa_w = 0.40$, 95%CI: 0.18–0.63), moderate for synovial thickening ($\kappa_w = 0.45$, 95%CI: 0.29–0.60), and good for joint enhancement ($\kappa_w = 0.61$, 95%CI: 0.48–0.75) (Table 4). The interobserver reliability of the damage domain items of the JAMRIS-TMJ scoring system was fair for condylar flattening ($\kappa = 0.39$, 95%CI: 0.26–0.53) and disk abnormalities ($\kappa_w = 0.28$, 95%CI: 0.11–0.45), and moderate for erosions ($\kappa = 0.44$, 95%CI: 0.27–0.61). The absolute agreement was lowest for bone marrow edema (40,8%) and highest for joint effusion (69.7%).

We hypothesized a positive correlation between TMJ involvement and the JAMRIS-TMJ items. Our results indicate a positive correlation between TMJ involvement and the JAMRIS-TMJ items for radiologist A on left TMJ, except for the items bone marrow edema ($r_s: 0.30$; $p = 0.064$) and disk abnormalities ($r_s: 0.28$; $p = 0.084$). Weak correlations were found for bone marrow enhancement ($r_s: 0.34$; $p = 0.022$), joint enhancement ($r_s: 0.33$; $p = 0.043$), condylar flattening ($r_s: 0.32$; $p = 0.048$), and erosions ($r_s: 0.35$; $p = 0.033$). A moderate correlation was found for synovial thickening ($r_s: 0.43$; $p = 0.007$). The results of radiologist A regarding the right TMJ did not indicate significant correlations. The results of radiologist B showed weak correlations for condylar flattening left TMJ ($r_s: 0.40$; $p = 0.014$), erosions left TMJ ($r_s: 0.33$; $p = 0.046$), disk abnormalities left TMJ ($r_s: 0.42$; $p = 0.009$), bone marrow enhancement right TMJ ($r_s: 0.37$ $p = 0.022$), and joint enhancement right TMJ ($r_s: 0.37$; $p = 0.024$).

As hypothesized, we found negative correlations between AMIO and the JAMRIS-TMJ items for radiologist A (Table 5). Weak correlations for AMIO were found for radiologist A in bone marrow enhancement right TMJ ($r_s: 0.37$; $p = 0.024$), joint effusion ($r_s: 0.35$; $p = 0.031$ and $r_s: 0.33$; $p = 0.042$), joint enhancement left TMJ ($r_s: 0.32$; $p = 0.049$), condylar flattening left TMJ ($r_s: 0.38$; $p = 0.018$), and disk abnormalities left TMJ ($r_s: 0.32$; $p = 0.049$). Moderate correlations for AMIO were found for bone marrow edema left TMJ ($r_s: 0.43$; $p = 0.007$), bone marrow enhancement left TMJ ($r_s: 0.40$; $p = 0.013$), synovial thickening left TMJ ($r_s: 0.46$; $p = 0.004$), joint enhancement right TMJ ($r_s: 0.44$; $p = 0.006$),

condylar flattening right TMJ ($r_s: 0.44$; $p = 0.006$), erosions ($r_s: 0.40$; $p = 0.013$ and $r_s: 0.50$; $p = 0.001$), and disk abnormalities right TMJ ($r_s: 0.43$; $p = 0.008$). The results of radiologist B only showed moderate correlations for joint effusion left TMJ ($r_s: 0.44$; $p = 0.006$) and condylar flattening right TMJ ($r_s: 0.43$; $p = 0.007$), and weak correlations for disk abnormalities ($r_s: 0.38$; $p = 0.380$ and $r_s: 0.38$; $p = 0.018$) and erosions right TMJ ($r_s: 0.37$; $p = 0.024$).

Correlations between AMVBF and the JAMRIS-TMJ items were all non-significant with values ranging from very weak to moderate.

4. Discussion

The reliability of the inflammatory domain items of the JAMRIS-TMJ scoring system varied from poor to good, and between poor to moderate for the damage domain. The highest reliability was found for joint enhancement. Correlations between the JAMRIS-TMJ scoring items and AMIO and TMJ involvement were found, although the results varied per radiologist and TMJ side. Radiologist A found for the items of both the inflammatory and damage domains a negative correlation between -0.30 and -0.50 for AMIO. These findings were not confirmed for radiologist B. Therefore, the validity of the JAMRIS-TMJ items and AMIO is only sufficient for radiologist A. No significant correlation was found between the JAMRIS-TMJ scoring items and AMVBF.

4.1. Comparison with existing literature

In our study, joint enhancement was the only item of the JAMRIS-TMJ scoring system that showed good reliability. The other items of the scoring system showed poor to moderate reliability. The JAMRIS-TMJ scoring system publication reported a moderate interobserver reliability of the inflammatory domain of the JAMRIS-TMJ scoring system and a good reliability of the damage domain (Tolend et al., 2021). In accordance with our study, they found poor reliability for the bone marrow items. Another study found a similar reliability for synovial thickening ($\kappa = 0.44$), compared to our study ($\kappa = 0.45$) (Angete et al., 2021). The items bone marrow edema ($\kappa = 0.54$), joint effusion ($\kappa = 0.71$), joint enhancement ($\kappa = 0.44$), condylar flattening ($\kappa = 0.66$), and disk abnormalities ($\kappa = 0.61$) showed a higher reliability than our results. Some older studies, originating from before the availability of the JAMRIS-TMJ scoring system, only analyzed a selection of the JAMRIS-TMJ scoring items. Moderate reliability ($\kappa_w = 0.51$) was reported for the overall score of the acute scoring items synovial effusion, synovial enhancement, synovial thickening, and bone marrow edema, with a higher interobserver reliability compared to our results (Vaid et al., 2014). One study found an agreement between two radiologists of 98% for joint effusion and 100% for joint enhancement, while we found 69.7% agreement for joint effusion and 67.1% for joint enhancement (Stoll et al., 2018). In another study, an agreement of 75% ($\kappa = 0.38$) was found for joint effusion and 62.5% ($\kappa = 0.33$) for synovial thickening, demonstrating a lower interobserver reliability compared to our results (joint effusion $\kappa = 0.40$ and synovial thickening $\kappa = 0.45$) (Weiss et al., 2008).

In our study, we correlated the clinical items TMJ involvement, AMIO, and AMVBF to each item of the JAMRIS-TMJ scoring system. We operationally defined TMJ involvement as a TMJ screening protocol score ≥ 2 (Steenks et al., 2015). Our definition is in line with the consensus-based definition of the TMJ Jaw Group published in 2019 (Stoustrup et al., 2019). Studies earlier than 2019 may have defined TMJ involvement differently. For example, in the study by Keller, TMJ involvement was defined as signs of inflammation (effusion/increased enhancement) and/or deformation (Keller et al., 2015). One study used a comprehensive clinical examination, like our TMJ screening protocol, and compared these items with MRI findings (Kuseler et al., 2005). The clinical examination consisted of symptoms and clinical findings, and afterwards a scoring of these items was constructed. However, a correlation between the clinical score and MRI was not found. Our

Table 2
Clinical symptoms.

	JIA (N = 38)
Items of the TMJ screening protocol score	
History:	
Problems in chewing (n, %)	20 (64.5)
Eating more slowly than others (n, %)	13 (41.9)
Biting hard food difficult (n, %)	23 (74.2)
Pain while eating (n, %)	24 (77.4)
Limited mouth opening (n, %)	15 (48.4)
Examination:	
Limited mouth opening (n, %)	19 (61.3)
Crepitation (audible) (n, %)	9 (29.0)
Pain AMIO (n, %)	16 (51.6)
Deviation AMIO (>2 mm) (n, %)	15 (48.4)
Inspection:	
Asymmetry (n, %)	16 (51.6)
Retrognathia (n, %)	5 (16.1)

AMIO: active maximum interincisal mouth opening; JIA: juvenile idiopathic arthritis; TMJ: temporomandibular joint.

Table 3
Scored items of the JAMRIS-TMJ scoring system.

Radiologist	Left TMJ						Right TMJ					
	A			B			A			B		
	Score 0	Score 1	Score 2	Score 0	Score 1	Score 2	Score 0	Score 1	Score 2	Score 0	Score 1	Score 2
Bone marrow edema (n, %)	27 (71.1)	11 (28.9)		5 (13.2)	33 (86.8)		31 (81.6)	7 (18.4)		8 (21.1)	30 (78.9)	
Bone marrow enhancement (n, %)	25 (65.8)	13 (34.2)		7 (18.4)	31 (81.6)		32 (84.2)	6 (15.8)		13 (34.2)	25 (65.8)	
Joint effusion (n, %)	27 (71.1)	8 (21.1)	3 (7.9)	21 (55.3)	11 (28.9)	6 (15.8)	28 (73.7)	8 (21.1)	2 (5.3)	23 (60.5)	10 (26.3)	5 (13.2)
Synovial thickening (n, %)	20 (52.6)	9 (23.7)	9 (23.7)	15 (39.5)	12 (31.6)	11 (28.9)	27 (71.1)	6 (15.8)	5 (13.2)	16 (42.1)	18 (47.4)	4 (10.5)
Joint enhancement (n, %)	16 (42.1)	11 (28.9)	11 (28.9)	11 (28.9)	15 (39.5)	11 (31.6)	22 (57.9)	11 (28.9)	5 (13.2)	18 (47.4)	14 (36.8)	6 (15.8)
Condylar flattening (n, %)	21 (55.3)	5 (13.2)	12 (31.6)	9 (23.7)	12 (31.6)	17 (44.7)	26 (68.4)	2 (5.3)	10 (26.3)	7 (18.4)	15 (39.5)	16 (42.1)
Erosions (n, %)	20 (52.6)	5 (13.2)	13 (34.2)	12 (31.6)	9 (23.7)	17 (44.7)	23 (60.5)	5 (13.2)	10 (26.3)	15 (39.5)	8 (21.1)	15 (39.5)
Disk abnormalities (n, %)	28 (73.7)	10 (26.3)		16 (42.1)	22 (57.9)		28 (73.7)	10 (26.3)		17 (44.7)	21 (55.3)	

JAMRIS-TMJ: Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints; TMJ: temporomandibular joint. The scoring system consists of an inflammatory and a damage domain. The inflammatory domain includes the items: 1) bone marrow edema (grade 0–1); 2) bone marrow enhancement (grade 0–1); 3) joint effusion (grade 0–2); 4) synovial thickening (grade 0–2); and 5) joint enhancement (grade 0–2). The damage domain contains the items: 1) condylar flattening (grade 0–2); 2) erosions (grade 0–2); and 3) disk abnormalities (grade 0–1) (Kellenberger et al., 2018).

Table 4
Interobserver Cohen’s (weighted) kappa coefficient and 95% confidence interval for JAMRIS-TMJ scoring items.

	Interobserver Cohen’s (weighted) kappa coefficient (95%CI)	Interobserver % absolute agreement
Inflammation domain		
Bone marrow edema	0.120 (0.04–0.20) ^a	40.8%
Bone marrow enhancement	0.179 (0.07–0.29) ^a	48.7%
Joint effusion	0.404 (0.18–0.63) ^b	69.7%
Synovial thickening	0.446 (0.29–0.60) ^b	65.2%
Joint enhancement	0.612 (0.48–0.75) ^b	67.1%
Damage domain		
Condylar flattening	0.393 (0.26–0.53) ^b	43.4%
Erosions	0.440 (0.27–0.61) ^b	60.5%
Disk abnormalities	0.282 (0.11–0.45) ^a	61.8%

CI: confidence interval; JAMRIS-TMJ: Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints; TMJ: temporomandibular joint.

^a Cohen’s kappa.

^b Cohen’s weighted kappa.

operationally defined TMJ protocol score of ≥ 2 has not been used in other studies relating signs and symptoms to MRI findings. Mostly single clinical variables are compared to MRI outcomes. However, in the study by Koos et al., a combination of clinical variables, such as the TMJ screening protocol, demonstrated a higher sensitivity for TMJ arthritis than each clinical variable separately (Koos et al., 2014).

A limited mouth opening is a common finding in children with JIA and TMJ arthritis (Abramowicz et al., 2013; Zwir et al., 2015; Kristensen et al., 2016; Stoustrup et al., 2017; Scolozzi et al., 2022). Moreover, a limited AMIO is associated with TMJ arthritis on MRI (Müller et al., 2009; Mohammed et al., 2012; Abramowicz et al., 2013; Zwir et al., 2015; Scolozzi et al., 2022). However, other studies did not find correlations between AMIO and TMJ arthritis (Pedersen et al., 2008; Koos et al., 2014; Keller et al., 2015). In one study, TMJ arthritis was defined as the presence of joint enhancement (Keller et al., 2015). Significant correlations existed between a limited mouth opening and TMJ arthritis

in this study, however, when patients with TMJ damage were excluded, significant correlation between a limited mouth opening and TMJ arthritis was non-existent. In another study, with the focus on TMJ damage and clinical findings, an association between TMJ damage and limited mouth opening was found (Rongo et al., 2019)

4.2. Clinical implications

In our study, the findings of radiologists A and B varied. Radiologist A had stronger correlations than radiologist B, and therefore more often significant correlations with AMIO and TMJ involvement. AMIO is found to be correlated with signs of TMJ arthritis on MRI in some studies (Müller et al., 2009; Mohammed et al., 2012; Abramowicz et al., 2013; Zwir et al., 2015; Scolozzi et al., 2022). Radiologist A was more experienced in evaluating TMJ MRIs, while radiologist B had less experience. In the original study of the JAMRIS-TMJ scoring system, all readers attended a video tutorial. Their training session may have improved the interobserver reliability of the scoring system, as they found a higher reliability than our study in which training was limited to assessment and comparison of the first five MRIs. An atlas and tutorial did not improve agreement within radiologists (Tolend et al., 2021), although, an atlas and tutorial improved the agreement between radiologists and non-radiologists. Our results suggest that experience and/or training is a factor influencing the results of radiologists using the JAMRIS-TMJ scoring system. Therefore, based on these results, training on the use of the JAMRIS-TMJ scoring system is recommended for all raters.

The multidisciplinary group of experts of the JAMRIS-TMJ scoring system published a study that weighted joint enhancement and synovial thickening as more important inflammatory items of the JAMRIS-TMJ scoring system than the bone marrow items (Tolend et al., 2022). In addition, synovial enhancement has been reported as the most common early finding in children with JIA (Müller et al., 2009). In most clinical studies, TMJ arthritis is defined as joint effusion, synovial thickening, and/or joint enhancement (Pedersen et al., 2008; Weiss et al., 2008; Müller et al., 2009; Mohammed et al., 2012; Koos et al., 2014; Keller et al., 2015; Zwir et al., 2015; Scolozzi et al., 2022). Therefore, these inflammatory items of the JAMRIS-TMJ scoring system seem to be more important than bone marrow edema and bone marrow enhancement. In addition, our results showed a higher reliability for scoring these items than for bone marrow edema and bone marrow enhancement.

Table 5
Correlation between JAMRIS-TMJ scoring items and TMJ involvement, AMIO, and AMVBF.

TMJ involvement				
Radiologist	Left TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	0.303	0.064	0.217	0.191
Bone marrow enhancement	0.343	0.022	0.301	0.066
Joint effusion	0.327	0.164	0.300	0.067
Synovial thickening	0.429	0.007	0.099	0.555
Joint enhancement	0.330	0.043	0.280	0.089
Condylar flattening	0.322	0.048	0.396	0.014
Erosions	0.347	0.033	0.326	0.046
Disk abnormalities	0.284	0.084	0.420	0.009
Radiologist	Right TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	0.226	0.173	0.254	0.124
Bone marrow enhancement	0.206	0.215	0.371	0.022
Joint effusion	0.137	0.412	0.256	0.121
Synovial thickening	0.305	0.171	0.178	0.285
Joint enhancement	0.161	0.334	0.365	0.024
Condylar flattening	0.107	0.524	0.254	0.124
Erosions	0.174	0.296	0.306	0.062
Disk abnormalities	0.284	0.084	0.255	0.122
AMIO				
Radiologist	Left TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	-0.432	0.007	-0.092	0.581
Bone marrow enhancement	-0.398	0.013	-0.299	0.068
Joint effusion	-0.351	0.031	-0.438	0.006
Synovial thickening	-0.457	0.004	-0.122	0.467
Joint enhancement	-0.321	0.049	-0.154	0.356
Condylar flattening	-0.380	0.018	-0.297	0.070
Erosions	-0.401	0.013	-0.179	0.282
Disk abnormalities	-0.322	0.049	-0.380	0.019
Radiologist	Right TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	-0.313	0.056	-0.127	0.448
Bone marrow enhancement	-0.366	0.024	-0.227	0.171
Joint effusion	-0.331	0.042	-0.299	0.068
Synovial thickening	-0.296	0.071	-0.157	0.347
Joint enhancement	-0.442	0.006	-0.242	0.143
Condylar flattening	-0.440	0.006	-0.429	0.007
Erosions	-0.498	0.001	-0.365	0.024
Disk abnormalities	-0.426	0.008	-0.382	0.018
AMVBF				
Radiologist	Left TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value

Table 5 (continued)

AMVBF				
Radiologist	Left TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	-0.308	0.064	-0.155	0.358
Bone marrow enhancement	-0.308	0.063	-0.221	0.189
Joint effusion	-0.155	0.359	-0.165	0.330
Synovial thickening	-0.300	0.072	0.136	0.421
Joint enhancement	-0.189	0.262	0.056	0.743
Condylar flattening	0.019	0.910	-0.027	0.875
Erosions	-0.095	0.576	-0.020	0.904
Disk abnormalities	-0.112	0.509	0.026	0.881
Radiologist	Right TMJ			
	A		B	
	Correlation coefficient (r_s)	p-value	Correlation coefficient (r_s)	p-value
Bone marrow edema	-0.268	0.109	-0.092	0.587
Bone marrow enhancement	-0.170	0.314	-0.116	0.494
Joint effusion	-0.293	0.079	-0.130	0.442
Synovial thickening	-0.198	0.241	-0.042	0.806
Joint enhancement	-0.182	0.282	-0.148	0.383
Condylar flattening	-0.250	0.135	-0.193	0.253
Erosions	-0.238	0.156	-0.270	0.106
Disk abnormalities	-0.239	0.154	-0.147	0.384

AMIO: active maximum interincisal mouth opening; AMVBF: anterior maximum voluntary bite force; JAMRIS-TMJ: Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints; JIA: juvenile idiopathic arthritis; r_s : Spearman's rank correlation coefficient; TMJ: temporomandibular joint.

4.3. Strengths and limitations

The strengths of this study are the comprehensive clinical screening and the use of the JAMRIS-TMJ scoring system. The limitations of this study are the low level of training of the JAMRIS-TMJ scoring system for the radiologists and a selected group of children. Our study consisted of 31 children with TMJ involvement and 7 children with JIA without any signs or symptoms. The reason for the small number of children without TMJ involvement is explained in an earlier study (de Sonnaville et al., 2022). Unfortunately, we were not able to reach our sample size, partly because of participants not willing to be included and partly because of delay between the rheumatologist visit and the presentation to the researcher.

Another limitation of our study is that the TMJ screening protocol cannot distinguish clinical signs of the left and right TMJ and the outcomes of MRI. When one side is involved, as a consequence the mouth opening and bite force will probably be affected as well. Our clinical approach using the TMJ screening protocol is unable to distinguish between the affected side(s) of TMJ. In perspective of treatment, systemic medication affects both TMJs.

4.4. Future research

In future research, it would be interesting to study a group of children with JIA with a mixed presentation of clinical signs and symptoms. TMJ arthritis without any clinical signs or symptoms, known as the silent joint, has been described (Weiss et al., 2008). The silent joint is mostly mentioned in studies comparing only a few clinical TMJ

variables to MRI outcomes (Koos et al., 2014; Kristensen et al., 2016; Stoustrup et al., 2017). It would be interesting to examine children with JIA with a comprehensive TMJ screening, such as the TMJ screening protocol, and compare the clinical findings with contrast-enhanced MRI findings. In order to include enough patients without any clinical TMJ signs or symptoms, a multicenter design is probably mandatory (de Sonnaville et al., 2022). Another suggestion for further research is to study the effect of training on the reliability of the JAMRIS-TMJ items in a pilot study.

5. Conclusion

The results of our study showed an interobserver reliability of the JAMRIS-TMJ items ranging from poor to good. Joint enhancement had the highest reliability. Correlations were found between clinically established TMJ involvement, AMIO, and the JAMRIS-TMJ items, although this varied per radiologist and TMJ side. Experience level of the radiologist seems to influence the scoring of the JAMRIS-TMJ items. The strongest correlations were found between most of the JAMRIS-TMJ items and AMIO. Our findings support the utility of AMIO as a clinical measure of TMJ status in children with JIA.

Declarations of interest

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References

- Abramowicz, S., Susarla, H.K., Kim, S., Kaban, L.B., 2013. Physical findings associated with active temporomandibular joint inflammation in children with juvenile idiopathic arthritis. *J. Oral Maxillofac. Surg.* 71 (10), 1683–1687.
- Angenete, O.W., Augdal, T.A., Rygg, M., Rosendahl, K., 2021. MRI in the assessment of TMJ-arthritis in children with JIA; Repeatability of a newly devised scoring system. *Acad. Radiol.* 1–16. <https://doi.org/10.1016/j.acra.2021.09.024> [Internet].
- Cohen, J., 1960. Weighted kappa: nominal scale agreement provision for scaled agreement or partial credit. *Psychol. Bull.* 70, 213–220.
- Consolaro, A., Negro, G., Chiara Gallo, M., Bracciolini, G., Ferrari, C., Schiappapietra, B., et al., 2014. Defining criteria for disease activity states in nonsystemic juvenile idiopathic arthritis based on a three-variable Juvenile Arthritis Disease Activity Score. *Arthritis Care Res.* 66 (11), 1703–1709.

- Elbeshlawi, I., AbdelBaki, M.S., 2018. Safety of gadolinium administration in children. *Pediatr. Neurol.* 86, 27–32. <https://doi.org/10.1016/j.pediatrneurol.2018.07.010> [Internet].
- Ferrara, G., Mastrangelo, G., Barone, P., La Torre, F., Martino, S., Pappagallo, G., et al., 2018. Methotrexate in juvenile idiopathic arthritis: Advice and recommendations from the MARAJIA expert consensus meeting. *Pediatr. Rheumatol.* 16 (1), 1–14.
- Frid, P., Nordal, E., Bovis, F., Giancane, G., Larheim, T.A., Rygg, M., et al., 2017. Temporomandibular joint involvement in association with quality of life, disability, and high disease activity in juvenile idiopathic arthritis. *Arthritis Care Res.* 69 (5), 677–686.
- Hechler, B.L., Phero, J.A., Van Mater, H., Matthews, N.S., 2018. Ultrasound versus magnetic resonance imaging of the temporomandibular joint in juvenile idiopathic arthritis: a systematic review. *Int. J. Oral Maxillofac. Surg.* 47 (1), 83–89.
- Kellenberger, C.J., Arvidsson, L.Z., Larheim, T.A., 2015. Magnetic resonance imaging of temporomandibular joints in juvenile idiopathic arthritis. *Semin. Orthod.* 21 (2), 111–120.
- Kellenberger, C.J., Junhasavasdikul, T., Tolend, M., Doria, A.S., 2018. Temporomandibular joint atlas for detection and grading of juvenile idiopathic arthritis involvement by magnetic resonance imaging. *Pediatr. Radiol.* 48 (3), 411–426.
- Keller, H., Müller, L.M., Markic, G., Schraner, T., Kellenberger, C.J., Saurenmann, R.K., 2015. Is early TMJ involvement in children with juvenile idiopathic arthritis clinically detectable? Clinical examination of the TMJ in comparison with contrast enhanced MRI in patients with juvenile idiopathic arthritis. *Pediatr. Rheumatol.* 13 (1), 1–10.
- Koos, B., Twilt, M., Kyank, U., Fischer-Brandies, H., Gassling, V., Tzaribachev, N., 2014. Reliability of clinical symptoms in diagnosing temporomandibular joint arthritis in juvenile idiopathic arthritis. *J. Rheumatol.* 41 (9), 1871–1877.
- Kristensen, K.D., Stoustrup, P., Küsel, A., Pedersen, T.K., Twilt, M., Herlin, T., 2016. Clinical predictors of temporomandibular joint arthritis in juvenile idiopathic arthritis: a systematic literature review. *Semin. Arthritis Rheum.* 45 (6), 717–732. <https://doi.org/10.1016/j.semarthrit.2015.11.006> [Internet].
- Kuseler, A., Pedersen, T., Gelineck, J., Herlin, T., 2005. A 2-year follow-up study of enhanced magnetic resonance imaging and clinical examination of the temporomandibular joint in children with juvenile idiopathic arthritis. *J. Rheumatol.* 32 (1), 162–169.
- Ma, K.S.K., Thota, E., Huang, J.Y., Wei, J.C.C., Resnick, C.M., 2022. Increased risk of temporomandibular joint disorders and craniofacial deformities in patients with juvenile idiopathic arthritis: a population-based cohort study. *Int. J. Oral Maxillofac. Surg.* 51 (11), 1482–1487.
- Malattia, C., Tolend, M., Mazzoni, M., Panwar, J., Zlotnik, M., Otobo, T., et al., 2020. Current status of MR imaging of juvenile idiopathic arthritis. *Best Pract. Res. Clin. Rheumatol.* 34 (6), 101629.
- Mohammed, Y., Saeed, O., Zaghloul, N., Samer, S., Mahmud, S., Abdulah, A., 2012. Juvenile idiopathic arthritis and the temporomandibular joint. *Alexandria Journal of Medicine* 48 (2), 123–129.
- Mokkink, L.B., Prinsen, C.A., Patrick, D.L., Alonso, J., Bouter, L.M., de Vet, H.C., et al., 2018. COSMIN Methodology for Systematic Reviews of Patient-Reported Outcome Measures (PROMs). COSMIN manual for systematic reviews of PROMs COSMIN, pp. 1–78 (February).
- Müller, L., Kellenberger, C.J., Cannizzaro, E., Ettlin, D., Schraner, T., Bolt, I.B., et al., 2009. Early diagnosis of temporomandibular joint involvement in juvenile idiopathic arthritis: a pilot study comparing clinical examination and ultrasound to magnetic resonance imaging. *Rheumatology* 48 (6), 680–685.
- Pedersen, T.K., Kuseler, A., Gelineck, J., Herlin, T., 2008. A prospective study of magnetic resonance and radiographic imaging in relation to symptoms and clinical findings of the temporomandibular joint in children with juvenile idiopathic arthritis. *J. Rheumatol.* 35 (8), 1668–1675.
- Rongo, R., Alstergren, P., Ammendola, L., Bucci, R., Alessio, M., D'Antò, V., et al., 2019. Temporomandibular joint damage in juvenile idiopathic arthritis: diagnostic validity of diagnostic criteria for temporomandibular disorders. *J. Oral Rehabil.* 46 (5), 450–459.
- Scolozzi, P., Rabuffetti, A., Hanquinet, S., Hofer, M., Courvoisier, D.S., Antonarakis, G.S., 2022. A clinical and MRI retrospective cohort study of patients with juvenile idiopathic arthritis (JIA) to determine if initial temporomandibular joint (TMJ) examination findings are associated with severity of TMJ arthritis. *J. Cranio-Maxillofacial Surg.* 50 (4), 328–335.
- de Sonnaville, W., Speksnijder, C., Zuithoff, N., Verkouteren, D., Wulffraat, N., Steenks, M., et al., 2021. Maximum bite force in children with juvenile idiopathic arthritis with and without clinical established temporomandibular joint involvement and in healthy children; a cross-sectional study. *J. Oral Rehabil.* 48 (7), 774–784.
- de Sonnaville, W.F.C., Steenks, M.H., Speksnijder, C.M., Wulffraat, N.M., Rosenberg, A.J.W.P., 2022. Challenging the silent temporomandibular joint paradigm in children with juvenile idiopathic arthritis. *Pediatr. Rheumatol.* 20 (1), 1–2.
- de Sonnaville, W.F.C., Steenks, M.H., Zuithoff, N.P.A., Wulffraat, M., Rosenberg, A.J.W.P., Speksnijder, C.M., 2023. Reliability and Measurement Error of Anterior Maximum Voluntary Bite Force in Children with Juvenile Idiopathic Arthritis and Healthy Children. *PLoS One*, pp. 1–13. <https://doi.org/10.1371/journal.pone.0280763> [Internet].
- Steenks, M.H., Giancane, G., de Leeuw, R.R.J., Bronkhorst, E.M., van Es, R.J.J., Koole, R., et al., 2015. Temporomandibular joint involvement in juvenile idiopathic arthritis: reliability and validity of a screening protocol for the rheumatologist. *Pediatr. Rheumatol.* 13 (1), 1–8.
- Stoll, M.L., Guleria, S., Mannion, M.L., Young, D.W., Royal, S.A., Cron, R.Q., et al., 2018. Defining the normal appearance of the temporomandibular joints by magnetic

- resonance imaging with contrast : a comparative study of children with and without juvenile idiopathic arthritis. *Pediatr. Rheumatol.* 16 (8), 1–7.
- Stoustrup, P., Resnick, C.M., Pedersen, T.K., Abramowicz, S., Küsel, A., Verna, C., et al., 2019. Standardizing terminology and assessment for orofacial conditions in juvenile idiopathic arthritis : International, multidisciplinary consensus-based recommendations. *J. Rheumatol.* 46, 518–522.
- Stoustrup, P., Twilt, M., Spiegel, L., Kristensen, K.D., Koos, B., Pedersen, T.K., et al., 2017. Clinical orofacial examination in juvenile idiopathic arthritis: International consensus-based recommendations for monitoring patients in clinical practice and research studies. *J. Rheumatol.* 44 (3), 326–333.
- Swart, J.F., de Roock, S., Wulffraat, N.M., 2013. What are the immunological consequences of long-term use of biological therapies for juvenile idiopathic arthritis? *Arthritis Res. Ther.* 15 (3).
- Swinscow, T., Campbell, M., 1997. *Statistics at Square One*, ninth ed. University of Southampton BMJ Publishing Group, London, UK, p. 22.
- Tolend, M., Doria, A.S., Meyers, A.B., Larheim, T.A., Abramowicz, S., Aguet, J., et al., 2021. Assessing the Reliability of the OMERACT Juvenile Idiopathic Arthritis Magnetic Resonance Scoring System for Temporomandibular Joints (JAMRIS-TMJ). Tolend, M., Junhasavasdikul, T., Cron, R.Q., Inarejos Clemente, E.J., von Kalle, T., Kellenberger, C.J., et al., 2022. Discrete choice experiment on a magnetic resonance imaging scoring system for temporomandibular joints in juvenile idiopathic arthritis. *Arthritis Care Res.* 74 (2), 308–316.
- Tolend, M.A., Twilt, M., Cron, R.Q., Tzaribachev, N., Guleria, S., von Kalle, T., et al., 2018. Toward establishing a standardized magnetic resonance imaging scoring system for temporomandibular joints in juvenile idiopathic arthritis. *Arthritis Care Res.* 70 (5), 758–767.
- Vaid, Y.N., Dunnivant, F.D., Royal, S.A., Beukelman, T., Stoll, M.L., Cron, R.Q., 2014. Imaging of the temporomandibular joint in juvenile idiopathic arthritis. *Arthritis Care Res.* 66 (1), 47–54. <https://doi.org/10.1002/acr.22177> [Internet].
- Weiss, P.F., Arabshahi, B., Johnson, A., Bilaniuk, L.T., Zarnow, D., Cahill, A.M., et al., 2008. High prevalence of temporomandibular joint arthritis at disease onset in children with juvenile idiopathic arthritis, as detected by magnetic resonance imaging but not by ultrasound. *Arthritis Rheum.* 58 (4), 1189–1196.
- Zwir, L.M.L.F., Terreri, M.T.R.A., Sousa, S.A., Fernandes, A.R.C., Guimarães, A.S., Hilário, M.O.E., 2015. Are temporomandibular joint signs and symptoms associated with magnetic resonance imaging findings in juvenile idiopathic arthritis patients? A longitudinal study. *Clin. Rheumatol.* 34 (12), 2057–2063.