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Standardizing Terminology and Assessment for Orofacial Conditions in Juvenile Idiopathic Arthritis: International, Multidisciplinary Consensus-based Recommendations

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Abstract:

Objectives: To propose multidisciplinary, consensus-based, standardization of operational terminology and method-of-assessment for condition related to temporomandibular joint (TMJ) involvement in juvenile idiopathic arthritis (JIA).

Methods: Using a sequential phased approach, an expert group defined terminology and methods-of-assessment by: 1) establishment of task force, 2) item generation, 3) working-group consensus meeting with drafting of provisional recommendations, 4) content validity testing of provisional recommendations by external experts, and 5) two round Delphi survey to reach final consensus among multidisciplinary group of experts (n=30) representing pediatric rheumatology, maxillofacial surgery, orthodontics, pediatric dentistry, radiology, and orofacial pain.

Results: A total of seven standardized operational terms were defined: TMJ arthritis, TMJ involvement, TMJ arthritis management, dentofacial anomaly, TMJ anomaly, TMJ symptoms, and TMJ dysfunction. All terms received a Delphi survey agreement score >80%. Additionally, the expert group defined methods-of-assessment for each of the terms from a pre-specified list of options.

Conclusions: The definition of seven operational standardized terms provides an optimal platform for communication across health care providers involved in TMJ arthritis management and will serve as reference standard for future research studies of TMJ arthritis in JIA.

Introduction

Temporomandibular joint (TMJ) arthritis is common in juvenile idiopathic arthritis (JIA).[1, 2] TMJ arthritis may impair joint mobility and masticatory function, cause TMJ degeneration, lead to reduced dentofacial growth, create orofacial pain, and impact general quality of life.[3-8] Treatment is complex and multidisciplinary, involving pediatric rheumatologists, maxillofacial surgeons, orthodontists, radiologists, pediatric dentists, occupational and physiotherapists, and orofacial pain specialists.[9] Research in TMJ arthritis has increased exponentially over the last decade. As this field has grown, so has confusion over terminology. Recent systematic reviews have highlighted the need for a standardized set of JIA-associated TMJ arthritis definitions.[6-8] Standardization is critical to enhance research comparability and care provider communication.

The objectives of this study were to use a consensus-based approach to propose: 1) a standardized terminology for JIA-associated TMJ arthritis, and 2) methods of assessment of TMJ arthritis in patients with JIA.

Materials and Methods

This study was conducted using a series of sequential iterations including: 1) establishment of task force and item generation, 2) working-group consensus meeting and drafting of provisional recommendations, 3) content validity testing of provisional recommendations by external experts, 4) Delphi survey to reach final consensus (Figure 1).

Task force assembly, item generation, consensus-meeting

The task force was assembled with members of the TMJ Juvenile Arthritis Working group (TMJaw; formerly known as euroTMjoint research network), an international, multidisciplinary, open group studying TMJ arthritis in JIA, and includes researchers from all specialties involved in JIA-related TMJ

arthritis management. Members of the terminology task force were identified based on clinical experience and scientific contributions. The task force included one pediatric rheumatologist (MT), two oral maxillofacial surgeons (SA, CR) and two orthodontists (TKP, PS), and represents one European and three North American centers.

For item generation, the task force identified TMJ arthritis-related terms used in existing JIA literature.[7, 8] In February 2017, those terms were distributed to all members of TMJaw via email, with an invitation to participate in an online questionnaire. The participants were asked to report their understanding of these terms and the role of different specialties involved in TMJ arthritis management. Based on the results of the online questionnaire, the terminology task force generated six provisional terms for discussion at the TMJaw meeting in Rostock, Germany, March 2017. The provisional terms were adjusted and definitions for each term were established based on consensus from the group.

Test of face validity

In April 2017, 16 external experts were invited to assess the validity of the provisional terms and definitions generated at the Rostock consensus-meeting. The external experts were identified based on TMJ arthritis clinical expertise and scientific merit. The external experts were asked to assess validity, suggest improvements, and address redundancies for the provisional terms. The task force then adjusted the provisional terms and definitions accordingly.

Delphi survey

In September 2017, participants from the Rostock consensus-meeting (n=18) and external experts (n=16) were invited to participate in an online Delphi survey to assess agreement with the provisional terms. Participants were asked to respond to each term and definition with: “agree”, “agree with minor changes”, “do not agree”. Participants were also asked to suggest improvements to the terms and

definitions, and to define methods-of-assessment for each term from the following options: 1) contrast-enhanced magnetic resonance imaging (MRI), 2) MRI without contrast, 3) 3-dimensional (3D) imaging (e.g., computed tomography or cone-beam computed tomography), 4) conventional radiology (e.g., cephalograms and panoramic radiographs), 5) ultrasonography, 6) clinical examination, 7) patient reported outcomes, 8) other (e.g., 3D photographs and scintigraphy). Participants could select multiple options.

The provisional terms and definitions were adjusted based on the results of this Delphi survey. These results were provided to participants before initiation of the next iteration in December 2017. In this final Delphi round, participants could “agree” or “disagree” with each term and definition. Only terms and definitions that received “agreement” by >80% of participants were included in the final recommendations. The results were summarized, and final consensus was reached in February 2018.

Results

Thirty experts participated in the final Delphi survey (Table 1). The following specialties were represented: pediatric rheumatology (n=10), maxillofacial surgery (n=5), orthodontics (n=8), pediatric dentistry (n=3), radiology (n=3), orofacial pain (n=1). Based on results of the first round of the Delphi survey, the number of terms was expanded from six to seven; the term “TMJ disability” was divided into “TMJ symptoms” and “TMJ dysfunction”. All seven provisional terms received a Delphi survey agreement score >80%.

TMJ arthritis

Arthritis is defined as “inflammation in a joint”. The term “TMJ arthritis”, therefore, is intended to indicate the presence of TMJ inflammation, and is independent of signs and symptoms. To add the qualifier “active” to the definition was considered redundant by some, but this word was ultimately included to highlight the strict nature of the term, which refers only to inflammation and not joint

damage per se. The term “chronic” TMJ arthritis has been used in literature, but the task force recommends avoiding this term due to imprecision.

Contrast-enhanced MRI is the current method for assessment of active inflammation in TMJ arthritis.[7]

Other methods (e.g., clinical examination and patient reporting) may suggest the presence of TMJ arthritis, but cannot confirm the presence of inflammation. Contrast-enhanced MRI was the only method-of-assessment that received a recommendation score >80%. A recently published MRI scoring system is recommended.[10, 11]

TMJ involvement and TMJ arthritis management

“TMJ involvement” is defined as “abnormalities presumed to be the result of TMJ arthritis”. This term is less restrictive than “TMJ arthritis”. The presence of active TMJ inflammation (“TMJ arthritis”) is not a prerequisite for “TMJ involvement”, but “TMJ arthritis” implies the presence of “TMJ involvement”. The term “TMJ involvement” is intended for: 1) 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; 2) patients with no current MRI evidence of active TMJ inflammation (“TMJ arthritis”) but with abnormalities indicating previous TMJ arthritis. Once the TMJ has been inflamed, it is prospectively considered “involved” regardless of the current inflammatory state.

By definition, “TMJ arthritis management” embraces diagnosis, treatment, and monitoring of TMJ arthritis and involvement. Methods-of-assessment include contrast-enhanced MRI, 3D imaging, clinical examination, and patient-reported outcomes.

Dentofacial anomaly and TMJ anomaly

“Dentofacial anomaly” refers to growth deviation that occurs as a result of TMJ arthritis in patients with JIA. Growth deviation may affect the morphology and position of the mandible, maxilla, and/or dental occlusion.[12, 13] The recommended assessment of dentofacial anomaly includes 3D imaging, conventional radiography, clinical examination and photographs, and recent recommendations are available.[13]

“TMJ anomaly” indicates arthritis-related alteration of the anatomy of the TMJ. This term does not cover signs and symptoms (which are considered in a future term, “TMJ dysfunction”), but rather is limited to anatomic anomalies. 3D imaging and conventional radiographs are used to assess osseous TMJ anomalies, with the caveat that soft tissue changes, which are often present in TMJ anomalies, will not be visible with these imaging techniques but would require MRI evaluation.

TMJ symptoms and TMJ dysfunction

“TMJ symptoms” refers to patient/parent reported measures. In contrast, the term “TMJ dysfunction” addresses clinical examination signs of abnormal mandibular function believed to be related to TMJ involvement. Recent recommendations for clinical orofacial examination in JIA are available.[8] The final recommendations for standardized terminology and definitions are presented in Table 1. The final recommendations for methods-of-assessment are shown in Table 2.

Discussion

In this study, the TMJaw group has described and defined the most common terms used in TMJ arthritis research and is uniquely positioned to provide these recommendations for standardization of terminology representing multiple specialties and many North American and European TMJ arthritis research centers. Consensus-based standardization of terminology provides an optimal platform for communication across health care providers involved in research and management of TMJ arthritis in

JIA. An important qualifier is that these terms only apply to JIA patients. It must be noted that TMJ symptoms and/or TMJ dysfunction may not be directly attributable to JIA, as in a patient with myofascial pain disorder. At this point, no reliable diagnostic method exists to distinguish between JIA-related orofacial symptoms/dysfunction and similar findings due to other etiologies. This project did not intend to define terms applicable to other temporomandibular dysfunction; further studies are required. Additionally, the methods-of-assessment that received recommendation scores >80% represent the most frequent diagnostic measures used to assess each term, and serve as a guide. Methods-of-assessment with a recommendation score <80% should not be discounted, however, as they may still be useful in special clinical scenarios.

The unequal numbers of participating experts representing different specialties may be a limitation to this study, however we are convinced that it strengthens the recommendations that so many different experts agree on the proposed standardized terminology.

We encourage investigators and clinicians to use the recommended terms and definitions in future publications as a reference standard. We suggest to include the following text: “terminology adheres to TMJaw consensus-based standardized terminology.”

Collaborators

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Figure 1. Flowchart of consensus process. *Response rate.

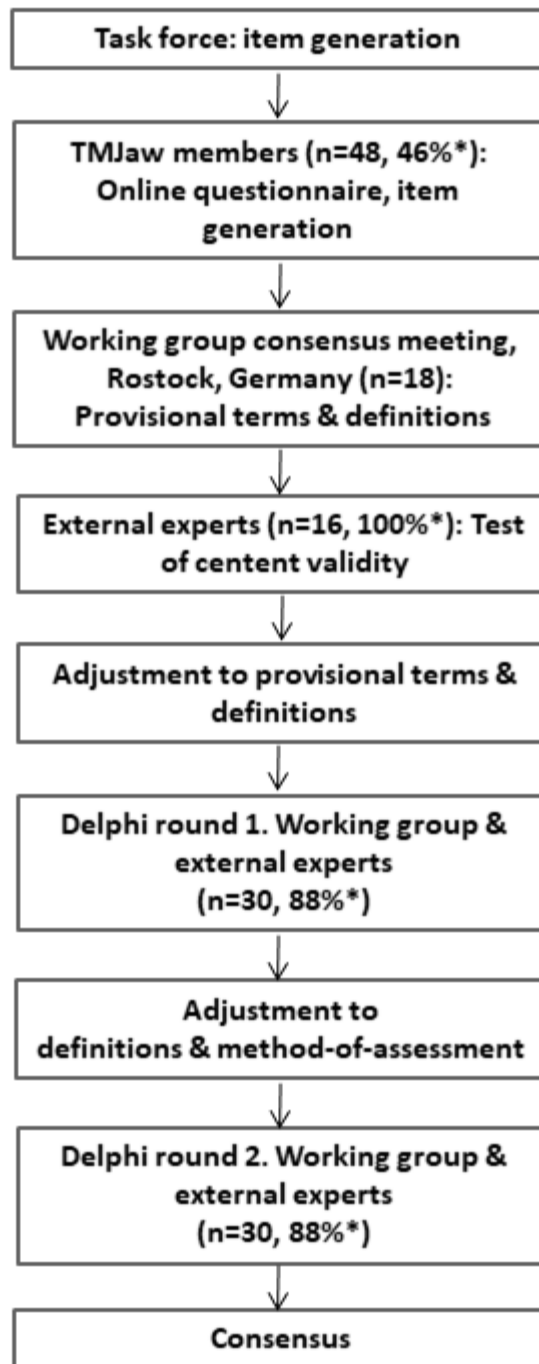


Table 1. Recommended standardized operational terminology, definitions and agreement with definitions. The terminology applies to subjects with a diagnosis of juvenile idiopathic arthritis. TMJ, temporomandibular joint.

Term	Definition	Agreement with definition
TMJ arthritis	Active inflammation in the TMJ	100%
TMJ involvement	Abnormalities presumed to be the result of TMJ arthritis	90%
TMJ arthritis management	Diagnosis, treatment and monitoring of TMJ arthritis and involvement	100%
Dentofacial anomaly	Abnormality in growth, development, structure and/or alignment of the facial bones and dentition	93%
TMJ anomaly	Abnormality in growth, development or structure of the osseous and/or soft-tissue components of the TMJ	90%
TMJ symptoms	Patient or parent-reported conditions related to TMJ arthritis or involvement	93%
TMJ dysfunction	Physician-reported functional examination abnormalities related to TMJ arthritis or involvement	90%

Table 2. Standardized terminology and method-of-assessment. Specific method of assessment are recommended when expert assessment $\geq 80\%$. Method-of-assessment with a recommendation score $< 80\%$ should not be discounted as they may still be important in special situations. * “Others” represent 3D photography and clinical photos. **Scintigraphy was included as an option to assess “TMJ anomaly” in the second Delphi-round and was recommended by 28% of the participants. TMJ, temporomandibular joint.

Term	Method of assessment							
	MRI with contrast	MRI without contrast	3D scans	Conventional radiology	Ultrasound	Clinical examination	Patient reported outcome	Others
TMJ arthritis	100%	7%	0%	3%	13%	29%	23%	0%
TMJ involvement	87%	43%	83%	67%	20%	83%	57%	13%
TMJ arthritis management	93%	35%	90%	69%	41%	93%	90%	17%
Dentofacial anomaly	35%	31%	97%	90%	14%	97%	28%	86%*
TMJ anomaly	62%	59%	100%	86%	28%	83%	24%	7%**
TMJ symptoms	18%	14%	14%	7%	7%	59%	100%	7%
TMJ dysfunction	14%	24%	7%	7%	4%	100%	31%	7%

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