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ABSTRACT

Introduction: Like any other synovial joint, the temporomandibular joint (TMJ) can be involved in juvenile idiopathic arthritis. TMJ can be affected unilaterally and bilaterally at the beginning or during the course of the disease, and can also often be affected by the first and only affected joint. **Objective:** To describe the clinical case of the patient diagnosed with temporomandibular disorder due to juvenile idiopathic arthritis, emphasizing the need for professionals to address the temporomandibular joint even in the absence of clinical symptoms. **Case Report:** Child, male, 10 years old, diagnosed with juvenile idiopathic arthritis, complained of pain in the region of the temporomandibular joint and limited mouth opening. During the clinical examination, he did not report pain on accessory muscles palpation, masticatory and cervical muscles. However, during palpation of the temporomandibular joint, it was found the presence of bilateral pain and decreased mouth opening, the maximum forced opening, with pain, marked 18 mm. The examination by cone beam computed tomography was requested and images out of the normal range were observed in both heads of the mandible, with the left side being more affected, showing flattening and erosion. After examining the exams, a diagnosis suggestive of type 3 internal derangement was reached. Thus, the proposed treatment consisted of using an occlusal splint and physiotherapeutic follow-up to reduce painful symptoms and increase mouth opening. After two weeks, an increase of 4 mm in the mouth opening was noted, with no effort or pain. After 3 months of proservation, an opening of 32 mm was observed with a stable patient regarding the initial complaints. **Conclusion:** The approach of the temporomandibular joint is extremely important in patients with juvenile idiopathic arthritis, since this association occurs quietly in most cases. Early recognition is essential in favor of favorable conducts and prognoses.

Key-words: Temporomandibular Joint Disorders, Juvenile arthritis, Signs and Symptoms; Therapeutics.

RESUMO

Introdução: Assim como qualquer outra articulação sinovial, a articulação temporomandibular (ATM) pode estar envolvida na artrite idiopática juvenil. A ATM pode ser afetada unilateralmente ou bilateralmente no início ou durante o curso da doença, e também pode ser frequentemente a primeira e única articulação afetada. **Objetivo:** Descrever o caso clínico de um paciente com diagnóstico de desordem temporomandibular decorrente da artrite idiopática juvenil, enfatizando a necessidade dos profissionais de saúde abordarem a articulação temporomandibular mesmo na ausência de sintomas clínicos. **Relato do Caso:** Criança, sexo masculino, 10 anos, diagnosticado com artrite idiopática juvenil, queixava-se de dor na região da articulação temporomandibular e limitação da abertura bucal. Durante o exame clínico, não relatou dor à palpação dos músculos acessórios, mastigatórios e cervicais. Porém, durante a palpação da articulação temporomandibular, constatou-se a presença de dor bilateral e diminuição da abertura bucal, sendo a abertura máxima forçada, com dor, marcada em 18 mm. Foi solicitado o exame por tomografia computadorizada de feixe cônico e observadas imagens fora da normalidade em ambas as cabeças da mandíbula, sendo o lado esquerdo mais acometido, apresentando achatamento e erosão. Após a análise dos exames, chegou-se a um diagnóstico sugestivo de desarranjo interno do tipo 3. Assim, o tratamento proposto consistiu em uso de placa oclusal e acompanhamento fisioterapêutico para redução da sintomatologia dolorosa e aumento da abertura bucal. Após duas semanas, notou-se aumento de 4 mm na abertura bucal, sem esforço ou dor. Após 3 meses de proservação, observou-se abertura de 32 mm com paciente estável em relação às queixas iniciais. **Conclusão:** A abordagem da articulação temporomandibular é de extrema importância em pacientes com artrite idiopática juvenil, visto que essa associação ocorre de forma silenciosa na maioria dos casos. O reconhecimento precoce é fundamental em prol de condutas e prognósticos favoráveis.

Palavras-chave: Transtornos da Articulação Temporomandibular, Artrite Juvenil, Sinais e Sintomas, Terapêutica.

INTRODUCTION

In view of the need for a common name to include all chronic childhood arthritis of unknown etiology affecting children up to 16 years of age, the International League of Rheumatology Associations (ILRA) defined this set of pathologies characterized by chronic inflammation, which persists for at least 6 weeks, as juvenile idiopathic arthritis (JIA). Through the combination of medical history, clinical presentation and radiological and laboratory changes, it is possible to categorize the affected individuals into seven subtypes: systemic, oligoarthritis, positive rheumatoid factor polyarthritis, negative rheumatoid factor polyarthritis, psoriatic arthritis, enthesitis-related arthritis and undifferentiated arthritis.¹⁻⁴ Stipulated as the most common childhood rheumatic disease,^{1,3,5,6} its cause and pathogenesis are not fully understood.^{3,6-8} It is considered that the individual genetic susceptibility and several environmental factors can lead to an imbalance of the immune system that *posteriori* can result in joint and systemic manifestations.³ In addition, other etiologies may be related, such as the presence of infections, physical trauma, immune disorders and stress.^{4,7}

Like any other synovial joint, the temporomandibular joint (TMJ) can be involved in JIA.⁹ TMJ can be affected unilaterally and bilaterally at the beginning or during the course of the disease, and can also often be the first and only affected joint.^{1,4,10} The literature shows that children affected by this pathology often develop TMJ inflammation with a reported prevalence between 17% and 87%.^{1-3,5,9,10} Patients may present changes in bone metabolism and skeletal growth, which can lead to extra-oral and/or intraoral changes, resulting in craniofacial disorders.¹¹ Such disorders, such as growth disorders, are different from arthritis in other joints due to the special anatomy of the TMJ, with fibrous cartilage and intra-articular condylar growth pattern.¹²

In addition, this joint demonstrates specific organizational-anatomical and developmental differences when compared to other joints.⁸ In this case, the articular and periarticular involvement observed in chronic arthritis leads to inflammatory changes in the synovial membrane, called synovitis. Histologically, this inflammation goes through some phases until the formation of a granulation tissue, which are exudation and cell infiltration. These stages, associated with active disease and subsequent immunopathological events perpetuate the inflammatory reaction and evolve towards the chronicity of the pathology. The destroyed articular tissue will be replaced by granulation tissue which, in turn, has the ability to penetrate the articular cartilage and subchondral bone, forming the pannus.¹¹ This infiltrate and corrodes the articular cartilage and the adjacent bone, which leads to destruction of the

jaw head or complete loss of it.^{3,6} Thus, prolonged inflammation can lead to disturbances in joint formation and damage to osteochondral structures.¹³

Unlike other joints, the mandibular growth zone is located below the surface of the head of the mandible, separated by a thin layer of fibrocartilage, playing an important role in regulating bone formation in the intramembranous and endochondral ossification of the mandible. These characteristics make it particularly vulnerable to damage and these can hinder the development of the growing patient.^{3,5,8,14-17}

In this context, JIA involving this joint potentially leads to degenerative arthritis changes, function restriction and pain, resulting in temporomandibular disorders (TMD).¹⁸⁻²⁰ Thus, all components of the TMJ can be affected, that is, the patient may present micrognathia, retrognathia, malocclusion, limited mouth opening, chewing difficulties, mandibular asymmetry, deviation, deflection, pain when perform the mandibular movements,^{1,3,10,11} joint noise, crackling,¹⁸ crackling, headache,³ palpatory sensitivity of the TMJ or associated muscles,⁶ morning stiffness,²¹ flattening of the condyle, shortening of the mandibular branch, erosive changes in the articular surface and the articular disc can still be observed with perforations and dislocation in long-standing cases.²²

In view of this, this report aims to describe the conduct from the diagnosis of TMD to the preservation of the case with reduction of symptoms in an affected patient by JIA, highlighting the importance of the TMJ approach even in the absence of signs and symptoms.

CASE REPORT

This study was described according to the CARE guidelines: consensus-based clinical case reporting guideline development and approved by the Research Ethics Committee CEP/UFJF, according to the attributions defined in Res. CNS 466/12 and with Operational Norm No. 001/2013, by means of opinion No. 4,255,177 and presents a free consent form and clarified signed and documented.

Child, male, 10 years old, went to the Diagnostic and Orientation Service for Patients with Temporomandibular Disorders, Faculty of Dentistry, Federal University of Juiz de Fora (SERVIÇO ATM-FO/UFJF), accompanied by the mother, with complaints of pain TMJ and difficulty opening the mouth. Anamnesis and physical examination were performed, in which it was found that he had previously been diagnosed with JIA of an oligoarticular character by the Pediatric Rheumatology Sector of the University Hospital (HU-UFJF/Ebserh). This clinical diagnosis was based on criteria that indicated the presence of chronic arthritis due to persistence for more than six weeks and because it was a patient under the age of sixteen who had signs and symptoms of arthritis in the joints of both knees. Added

to these evidences, as a conduct adopted by the medical professional specialized in pediatric rheumatology, during the physical examination, there was an alteration in the mouth opening with a possible cause of arthritis affecting the TMJ. Thus, the professional referred the patient to the Dentistry School for evaluation and therapeutic conduct of orofacial symptoms.

At the point of the first contact with the child, during the anamnesis, the legal guardian for the minor reported that he had had mouth opening restriction since he was nine years old, observing some difficulty in brushing his molars. It was also informed that the patient did not seek any other professional nor did he use any therapy to treat his complaints. Still in the investigation of past history, the parents informed that he did not perform parafunctional habits and for the treatment of JIA, used the following medications: methotrexate, folic acid, prednisone and vitamin D3.

On extra-oral clinical examination, the minor did not report pain during palpation of the masticatory muscles, accessory muscles and cervical muscles. On the other hand, during palpation of the TMJ, he referred to bilateral grade 3 pain, according to Okeson with painful symptoms pain index 10 as recommended by the Subjective/Objective Orofacial Pain Assessment (ASDORF) (figure 1) model employed by the SERVIÇO ATM-FO/UFJF.²³ Still in the facial exam, the absence of asymmetries and joint noises was observed, however, there was an alteration in the mouth opening. This was observed using a Willis compass, in which the forced maximum opening, with pain, marked 18 mm (figure 2a) and slight deflection to the left side. In the intraoral examination, the patient had mixed dentition with erupting permanent lower canines and absence of permanent second molars.

In order to evaluate the joint itself, an image exam by cone beam computed tomography (CBCT) was requested, where the images reported bilateral abnormal patterns, with the left mandibular head being the most affected (figure 3a and 3b). In addition, it presented morphological changes such as the presence of erosions and flattening, resulting in an asymmetry of this structure. Bilateral hypomobility was also observed.

In view of this, from the anamnesis and thorough clinical examination, associated with the complementary image examination, it was possible to arrive at a diagnosis suggestive of type III internal derangement associated

with the presence of JIA.²⁴ Therefore, the proposed therapeutic approach was to use a neuromiorelaxative splint (NMRS), with the objective of regressing painful symptoms and possibly increasing the degree of mouth opening. Initially, this was requested in wax, so that the necessary adjustments in centric relation, along with the contact points and occlusal guides, could be analyzed. *Posteriori*, the acrylic plate was made and adapted with freedom of bilateral and anterior excursion and the minimum possible lifting of the vertical dimension, in addition to the absence of interference in centric relation and occlusion. The patient used it over 24 hours/day for 14 days, removing it only for feeding and brushing.

The boy was instructed to return to the clinical evaluation consultations every 14 days to adjust the device as needed. During the first return, there was an increase of 4 mm in the mouth opening, effortlessly and painlessly, measured in Willis' compass, which was close to 22 mm (figure 2b). Still, there was a significant improvement in painful symptoms, lowering the pain index from 10 to 0, according to the ASDORF model.

In view of the observed, the orientation given to the patient was to reduce the use of the device to only at night, during sleep. After 3 months of preservation, the patient remained stable regarding the complaints initially reported. In this evaluation, the patient presented a mouth opening measuring 32 mm (77.77% increase) without pain and without deflection (figure 2c) and was advised as to the need for joint treatment and monitoring by the Physiotherapy professional.

COVID-19, it was not possible to continue with the return of consultations in the intervals mentioned above and the minor was not assisted by the physiotherapist. On the other hand, the legal guardian for the minor reported that he showed improvement in the systemic symptoms of the disease and, regarding TMD, the information shown in the last return did not change, therefore, he remained without any painful symptoms and kept the 32 mm mouth opening.

DISCUSSION

TMJ involvement in patients affected by JIA is a scientifically proven aspect observed in the literature. This impairment can be presented unilaterally or bilaterally with manifestations at the beginning or during the course of this pathology.^{1,3-5,10} However, due to the



Figure 1: An agreement was made with the patient: he should give score 10 to the pain he was feeling in the moment of the first consultation, regardless of the intensity of the pain in that moment. The authors made clear that in the following meetings, after the beginning of the treatment, he should write down the variation of the score in a scale 0 to 10, being 0 the absence of pain and 10 the pain he was feeling in the very beginning of the treatment.

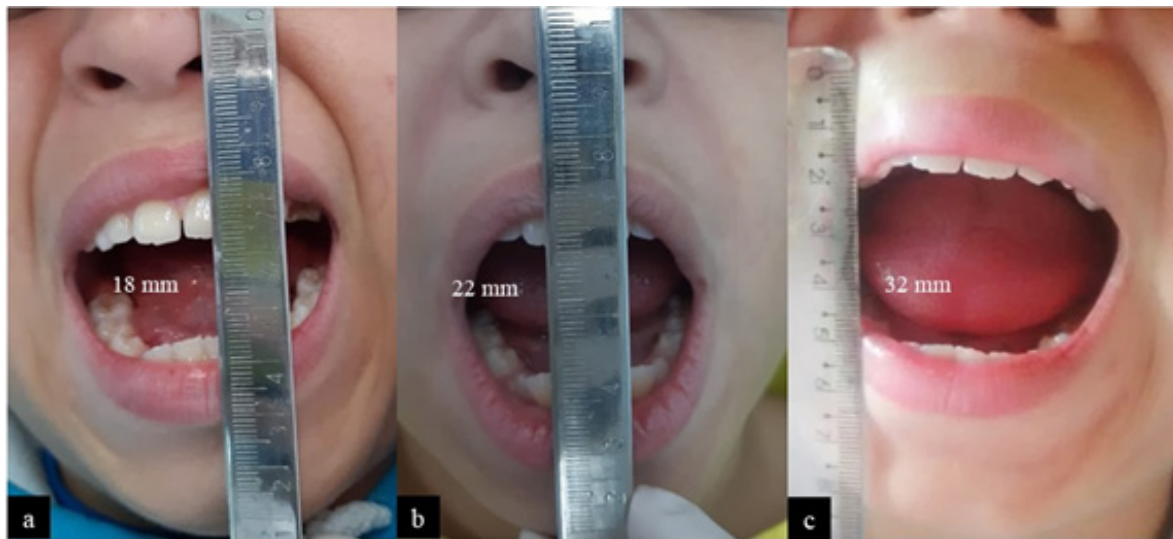


Figure 2: Extra-oral photographic images (a, b and c) showing changes in the mouth opening. Through the Willis' compass, it is possible to observe the limitation of the opening marked at 18 mm (a). After 14 days, there is an increase of 4 mm (b). After 3 months of preservation, a 32 mm mouth opening is observed (c).

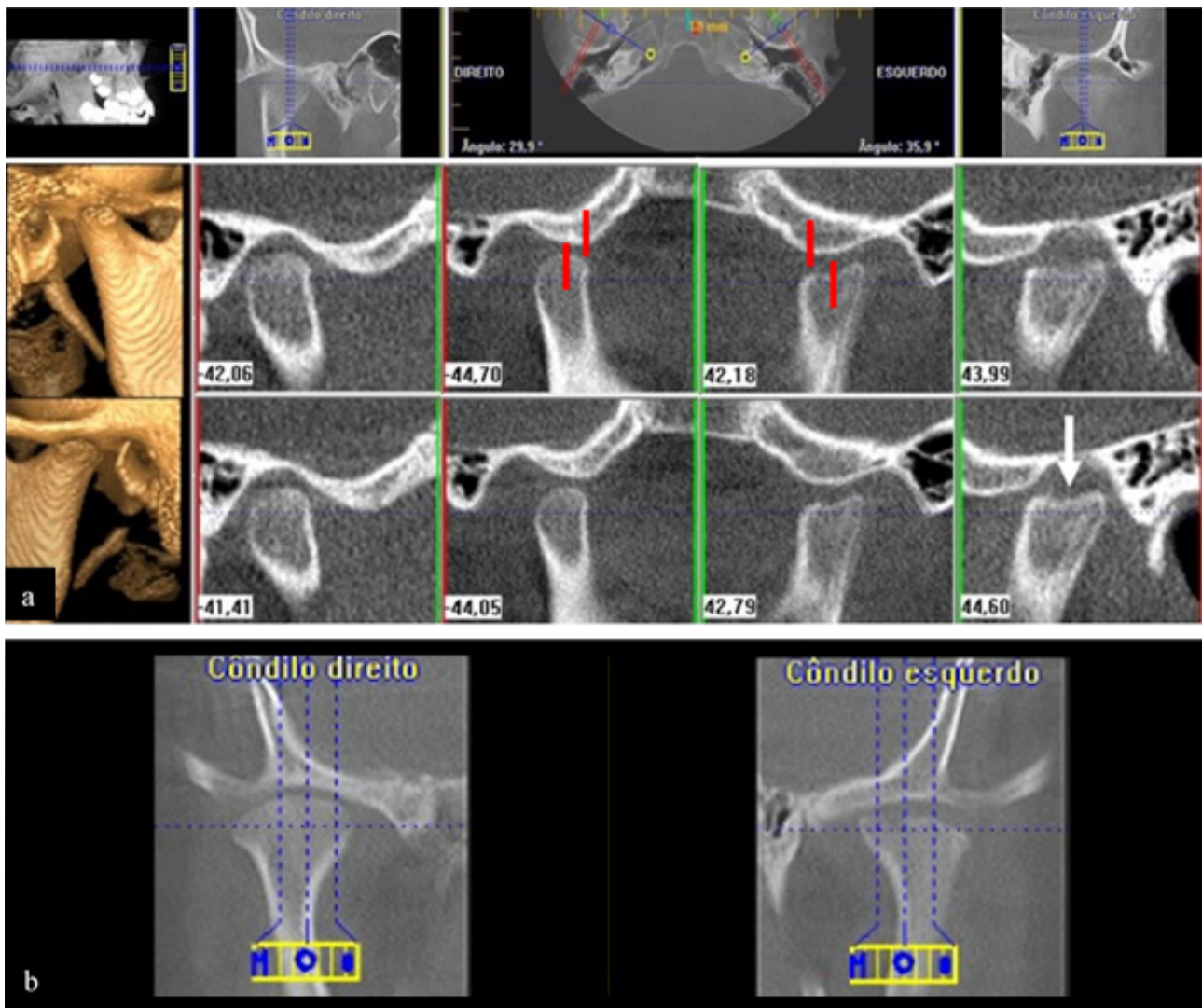


Figure 3: CTFC image at maximum habitual intercuspatation and at maximum mouth opening, exposing morphological changes in both heads of mandible (a) and coronal section showing details of the conditions (b). It is observed on the left side (arrow) the presence of erosions and planing and it is possible to notice hypomobility bilaterally (dashes).

absence of visible joint edema and the lack of symptoms at the beginning or throughout the pathogenesis, this joint becomes extremely challenging during clinical evaluation, which can provide serious structural changes in its form and function.^{3,9,18,19} In the clinical case in question, these structural changes were evident, given the consistent erosive process observed in the left jaw head and the mandibular hypomobility with consequent restriction of mouth opening.

The data collected during the anamnesis reported that the patient described has JIA of the oligoarticular type, the most common among the subtypes, represented in 50% of those affected by this pathology.²¹ According to the literature, the JIA subtype strongly influences the severity orofacial changes. Thus, studies have pointed out the polyarticular subtype as the most likely to present these variations.^{1,8,9,11,12} On the other hand, related to TMJ involvement, studies mention the oligoarticular subtype as the most implicated,^{5,19} since children with major condylar disorders belonged to the oligoarticular category in 53% of cases.²

As symptomatology identified in the patient, TMJ pain was present on both sides, limited mouth opening, morphological changes such as flattening and erosion on the surfaces of the jaw head and bilateral hypomobility. This report corroborates the research carried out by Urtane et al¹⁴, which aimed to assess the relationship between clinical signs and radiological characteristics of TMJ in CBCT of patients with JIA in which pain was the most frequently reported sign in 58 of the 65 observed patients (89.2%) and mouth opening was limited to 33.8% of the studied sample. In addition, this study also mentions the presence of flattening, erosion and osteophytes in 62 (95.4%), 48 (73.8%) and 30 (46.2%) patients, respectively, (14) thus associating the frequent presence of these signs and symptoms in groups with JIA with TMJ manifestation. In addition, when compared to the group without TMJ involvement, they commonly have crackles,⁴ functional limitations, especially in food and oral hygiene, functional disability, lower health-related quality of life and longer disease duration,^{2,6,9,12,25} which matches the patient's history and clinical conditions.

Data on the time of treatment for JIA and the use of medications for systemic control in improving the clinical symptoms of TMD showed that the time of treatment did not prevent the development of these disorders and the symptoms presented can be concealed in view of the proposed therapies.^{10,4} On the other hand, despite these findings, studies describe that TMJ can respond to systemic immunosuppressive treatment, but there is a lack of information on how this return is performed.^{3,18,22} In these terms, it is essential to be concerned with the clinical and images due to the lack of signs and symptoms possibly covered by supportive therapies.

During the extra-oral physical examination, it

was observed that the patient in question had limited mouth opening, deflection to the left side, sensitivity to palpation of the TMJ and no pain on muscle palpation. These reports are in agreement with other studies, in which it was observed that JIA when it affects the TMJ causes anatomical changes in the components of this joint, significantly reducing the maximum mouth opening.^{5,9,19,22} These clinical characteristics were exhibited in the patient of this clinical case. In addition, similarly to these findings and the patient described, this same symptom was observed in another case report, in which there was an alteration during mouth opening and closing with a 22 mm mouth opening limitation, deflection to the right side and TMJ pain during palpation on the same side, without accusation of symptoms on muscle palpation.²⁰

These clinical findings identified in the literature and observed in this clinical case show the need for a complementary examination to obtain a diagnosis of excellence and the taking of an effective therapeutic approach, since the clinical symptoms associated with the results of the physical examination itself, they are just not good markers of TMJ involvement.^{3-5,19} In this regard, CBCT and magnetic resonance imaging (MRI) can be used, the time periods are different and define which exam will be recommended. Synovial inflammation appears early to rigid tissue deformities.¹⁶ Thus, while CBCT provides a more detailed assessment of the anatomy of TMJ bone components with 3D images, high diagnostic quality and without obstruction and overlapping of anatomical structures,^{7,8,10,14,19,22} MRI allows the assessment of soft tissues, the identification of acute joint inflammation and the differentiation between pannus and synovial hypertrophy.^{7,22}

In this context, MRI has become the gold standard for assessing TMJ in children with JIA, due to the assessment of both inflammation and joint damage.^{8,13-15,19} Unlike the exposed technique, the use of CBCT is not able to examine soft tissues and provides information only on arthritic sequelae resulting from a late diagnosis.^{8,10,14,18,21,22} Thus, it was the complementary exam used in the patient described, since he did not present an early diagnosis and bone alterations were already suspected. In addition, the high cost of MRI made it impossible for this examination to be carried out.

After analyzing the CBCT images, it was possible to observe morphological changes in both heads of the mandible with a high degree of bone deformation on the left side. This occurrence solidifies the professional's necessary attention during clinical and imaging examinations in the early detection of TMJ involvement before significant craniofacial deformities and problems in occlusion appear.^{6,9-11,14} Thus, the rheumatologist when detecting such changes early you need to refer the patient to a team that should be, preferably, multidisciplinary, obligatorily involving the specialist

dentist in TMD.^{3,11,21} The team approach is crucial to assess the different active points of the disease and provide a more comprehensive view, where the coexistence of systemic disorders is considered and provides better possibilities for restoring patients' function and quality of life.^{20,21} In the patient described, there was already complexity in feeding caused by difficulty in chewing and compromised oral hygiene due to the limited degree of mouth opening.

The literature describes numerous therapies that can be used to manage TMJ complications from JIA. A combination of pharmacological therapies, physiotherapy, occlusal devices, orthodontics, surgery and counseling are included.^{8,21} In view of a pathology of such complex symptoms, it is recommended that the initial therapy instituted is reversible and non-invasive.⁸ In the case of described clinician, the NMRS and physiotherapeutic follow-up were chosen in view of the complaints reported by the patient. Physiotherapy in this scenario, has good effects in the face of decompensating masticatory conditions and in cervical muscles in the absence of active inflammatory disease.³ The results obtained in this study corroborate those observed in another report, which, after using the plate for six months and physiotherapy with compresses and stretching, the patient showed a significant increase in mouth opening from 22 mm to 33 mm, with control of the aforementioned pains.²⁰ These findings contrast data exposed in the literature on therapeutic intervention through NMRS, in which indicated It should be noted that this should only be started during the early stages of TMD and that it usually requires many years of adherence.^{4,18} What was not observed in the clinical case described and, in the case, mentioned above, (20) the results obtained in patient were proportionally better (77.7%) in the first weeks of adherence and even after the late diagnosis of this pathology.

CONCLUSION

In view of this, this clinical case emphasizes the importance of approaching the TMJ in patients affected by JIA, due to the presence of numerous neglected and late diagnosed cases. Furthermore, this study highlights the need for professionals to investigate TMJ even in the absence of clinical signs and symptoms, since the pathologies that affect this region often occur in an asymptomatic and silent manner. The pediatric patient affected by this association is exposed to high levels of disability, longer disease duration and decreased quality of life. Thus, the early recognition of this involvement becomes essential in the proper approach and treatment.

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