

# Conservative management of pediatric temporomandibular disc displacement presenting as juvenile idiopathic arthritis: a case report

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*There is limited research regarding management of temporomandibular disorders (TMD) in adolescents with imaging signs of juvenile idiopathic arthritis (JIA). An 11-year-old girl presented to a hospital-based chiropractor for evaluation of a 1.5-year history of unilateral temporomandibular joint (TMJ) pain and trismus. Previously, pediatric rheumatologists diagnosed JIA after contrast-enhanced magnetic resonance imaging revealed edema, effusion, and bilateral anterior disc displacement, and recommended methotrexate, corticosteroid injection, and arthrocentesis. The chiropractor questioned the JIA diagnosis, instead relating symptoms to a mechanical TMD/disc origin.*

*Prise en charge conservatrice, en pédiatrie, du déplacement du disque temporo-mandibulaire se présentant comme une arthrite juvénile idiopathique : à propos d'un cas.*

*Il existe peu de recherches sur la prise en charge des troubles temporo-mandibulaires (TTM) chez les adolescents présentant des signes d'imagerie de l'arthrite juvénile idiopathique (AJI). Une fillette de 11 ans s'est présentée chez un chiropraticien en milieu hospitalier pour l'évaluation d'un antécédent d'un an et demi de douleur unilatérale à l'articulation temporo-mandibulaire (ATM) et de trismus. Auparavant, les rhumatologues pédiatriques diagnostiquaient l'AJI une fois que l'imagerie par résonance magnétique avec contraste aurait révélé un œdème, un épanchement*

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*Manual therapy, TMJ exercises, and acupuncture improved TMJ pain and opening. Invasive medical JIA interventions were avoided without long-term recurrence, further questioning the preceding JIA diagnosis. The success of this case suggests that stepped care, beginning with conservative treatment, has value for adolescents with TMD suspect for JIA. Integration of chiropractors and acupuncturists into healthcare institutions may facilitate this care model by affording nonpharmacologic interventions earlier in patient care.*

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KEY WORDS: acupuncture, chiropractic, differential diagnosis, juvenile arthritis, misdiagnosis, musculoskeletal manipulations, overtreatment, pediatrics, temporomandibular joint

*et un déplacement bilatéral du disque antérieur, et recommandaient le méthotrexate, l'injection de corticostéroïdes et l'arthrocentèse. Le chiropraticien a remis en question le diagnostic d'AJI, associant plutôt les symptômes à une origine mécanique du TTM/disque. La thérapie manuelle, les exercices de l'ATM et l'acupuncture ont amélioré la douleur et l'ouverture de l'ATM. Les interventions médicales invasives d'AJI ont été évitées sans récurrence à long terme, remettant davantage en question le diagnostic d'AJI précédent. Le succès de ce cas suggère que les soins par étapes, en commençant par un traitement conservateur, ont de la valeur pour les adolescents atteints de TTM supposés d'AJI. L'intégration des chiropraticiens et des acupuncteurs dans les établissements de santé peut faciliter ce modèle de soins en permettant des interventions non pharmacologiques plus tôt dans les soins aux patients.*

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MOTS CLÉS : acupuncture, chiropratique, diagnostic différentiel, arthrite juvénile, diagnostic erroné, manipulations musculosquelettiques, surtraitement, pédiatrie, articulation temporo-mandibulaire

## Introduction

Temporomandibular disorders (TMD) are those affecting the masticatory system, which includes the temporomandibular joint (TMJ), masticatory muscles, and associated tissues.<sup>1</sup> The prevalence of TMD is relatively high in adolescents, ranging between 7 and 30%.<sup>2</sup> The etiology of adolescent TMD includes a broad differential diagnosis with the most common causes being myofascial pain and disc displacement<sup>2</sup>, and less common including trauma-associated synovitis<sup>3</sup>, fracture, juvenile idiopathic arthritis (JIA), and idiopathic condylar resorption<sup>4</sup>.

There is limited research and testing available to help distinguish between TMJ disc disorders and JIA in adolescents. While limited maximal incisal opening or deviation on jaw opening are predictive of TMJ synovitis in those with a known history of JIA<sup>5</sup>, these findings are also often seen in more common disorders such as TMJ disc displacement<sup>6,7</sup>. In addition, the diagnostic accuracy of

contrast-enhanced magnetic resonance imaging (MRI) for diagnosing JIA of the TMJ has been questioned, as many children without JIA have contrast-enhancing joint fluid.<sup>8</sup> One study comparing the contrast-enhanced MRI features of adolescents with TMJ disc displacement and those having JIA found that both groups had similar rates of joint enhancement, joint effusion, and synovial thickening.<sup>4</sup>

There is also limited evidence to guide the management of pediatric TMD.<sup>2</sup> In adolescents without JIA, there is some evidence that a stabilizing occlusal appliance is superior to advice or relaxation therapy.<sup>2</sup> In adolescents with JIA there is no consensus on TMD treatment, although methotrexate and intra-articular corticosteroid injections are often utilized.<sup>9</sup> In the absence of a standardized management algorithm for adolescent TMD, alternative care models could have value, such as stepped care, which begin with the least invasive treatments.<sup>10</sup>

In adults, occlusal splints are an evidence-based and routinely-used non-invasive treatment for TMD.<sup>1,11</sup> A recent systematic review supported the efficacy of occlusal splints for TMD, which were found to have a positive effect on mandibular movement in several studies.<sup>11</sup> In addition, occlusal splints were found to benefit mouth opening, and TMJ pain, locking, and clicking.<sup>11</sup>

While there is evidence to support less invasive treatments such as manual therapy and acupuncture for TMD, most supporting studies have been conducted with an adult population.<sup>12-14</sup> One systematic review found evidence that manual therapy is effective for medium-term TMD outcomes, and the addition of therapeutic exercises helped maintain these outcomes in the long-term.<sup>12</sup> Another systematic review found that manual therapy or therapeutic exercise were beneficial for patients with TMJ disc displacement without reduction, with limited evidence to suggest that exercises can improve mouth opening.<sup>13</sup> There is limited, lower quality evidence that acupuncture is superior to placebo and equivalent to occlusal splints in treating TMD.<sup>14</sup>

Disorders of the TMJ often co-occur with abnormalities of spinal posture and/or cervical spine dysfunction. Systematic reviews including mostly an adult population have identified moderate evidence of an association between TMD and cervical postural misalignment (i.e. forward head posture)<sup>15</sup>, abnormal global posture<sup>16</sup>, reduced cervical spine range of motion<sup>17</sup>, lower deep cervical extensor muscle endurance<sup>18</sup>, upper and global cervical spine hypomobility<sup>18</sup>, and strong evidence of an association between TMD and self-reported neck disability<sup>18</sup>. There is some evidence that spinal manipulative therapy (SMT) for the cervical spine is beneficial for TMD<sup>19</sup>, while there is less evidence supporting the use of thoracic spine SMT for TMD<sup>20</sup>.

The goal of this case report is to present an adolescent with TMD suspected for JIA that was successfully treated with conservative therapies without recurrence. Our literature search of the Index to Chiropractic Literature, PubMed, and Google Scholar revealed one other pediatric case of TMJ disc displacement that received chiropractic care and was co-managed with arthrocentesis.<sup>21</sup> Otherwise, few cases of chiropractic management of pediatric TMD have been reported.

## Case presentation

### *Patient information*

An otherwise healthy 11-year-old female soccer player presented to an integrative hospital-based chiropractic office with an insidious-onset 1.5-year history of left TMJ pain, limited mouth opening, locking, and clicking, and intermittent left temporal headache. Pain was rated 3-5/10 on a numeric rating scale. She had difficulty eating meat and occasionally had difficulty sleeping due to the symptoms. While she had been experiencing TMJ symptoms for over a year, symptoms worsened two months preceding her chiropractic evaluation, when she was hit in the face with a soccer ball that had been kicked towards her, however the exact mechanism of this incident (i.e. direction and location of the hit) was not ascertained. She had prior sports injuries including a left ankle sprain and left forearm fracture but no other significant medical history such as arthralgia or uveitis, and did not have dental braces. She was taking indomethacin 50 mg bid as prescribed by a pediatric rheumatologist. The patient's mother had undergone evaluations and laboratory testing for joint pain via orthopedic and internal medicine specialists, which were not supportive of an inflammatory arthritis, resulting in diagnoses of generalized hypermobility and peripheral joint osteoarthritis. The patient's father had asthma. There was no other significant family history.

Two months prior the patient saw a pediatric otolaryngologist. This provider ordered TMJ radiographs and computed tomography, the findings of which were normal. Based on the patient's symptoms and limited mouth opening on examination this provider considered JIA as the main differential diagnosis, followed by Ehlers-Danlos syndrome, and referred the patient to a pediatric rheumatologist.

The pediatric rheumatologist ordered laboratory tests which were significant for a positive HLA-B27, however the other tests were normal including: erythrocyte sedimentation rate (4 mm/hr), c-reactive protein (0.058 mg/dL), antinuclear antibodies, rheumatoid factor, anti-cyclic citrullinated peptide antibodies, complete blood count with differential, tissue transglutaminase antibodies, Gliadin peptide IgA, Varicella Zoster IgG, tuberculosis (TB) spot, reticulin antibody IgA, endomysial IgA <1:10, Westergren ESR 4 mm/hr, CRP 0.058 mg/dL, comprehensive metabolic panel, gamma glutamyl trans-

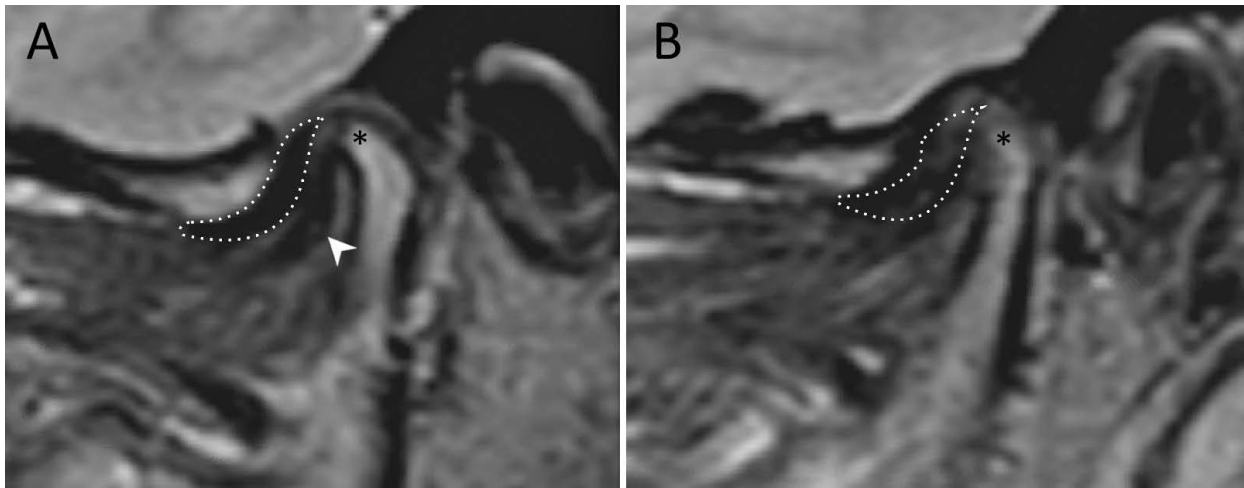


Figure 1.

*Oblique sagittal T2-weighted MRIs of the left (A) and right (B) TMJ with the mouth closed. The articular disc (dotted line) is displaced anteriorly from its normal position near the top of the mandibular condyle (\*). Thickening of the lateral pterygoid muscle tendon (arrowhead) is seen parallel and subjacent to the disc, more clearly seen in image A, producing the “double disc” sign.*

ferase, phosphorus, lactate dehydrogenase, and uric acid. Diclofenac 50 mg bid was prescribed.

The pediatric rheumatologist also ordered a gadolinium-enhanced MRI of the TMJ (Figures 1 and 2) which identified bilateral anterior disc displacement, and additional findings in the left TMJ of synovial enhancement, marrow edema, and enhancement of the mandibular condyle without discrete erosion. The radiologist noted that the findings were “concerning for early inflammatory arthritis in the proper clinical setting.”

The pediatric rheumatologist considered the patient to have JIA and for the purpose of treating this suspected arthritis changed the patient’s medication to nabumetone 1000 mg, followed by indomethacin 50 mg bid. Because none of these therapies alleviated the patient’s symptoms, the specialist then prescribed oral methotrexate 20 mg weekly (15 mg/m<sup>2</sup>), along with a folic acid supplement (1 mg/day), with the goal of eventually transitioning to adalimumab (Humira®). The patient and her family sought a second opinion from a pediatric rheumatologist at another health care organization, who concurred with the JIA diagnosis, and in addition, recommended an intra-articular corticosteroid injection.

The family then sought the opinion of two maxillofacial surgeons. One surgeon recommended arthrocentesis and

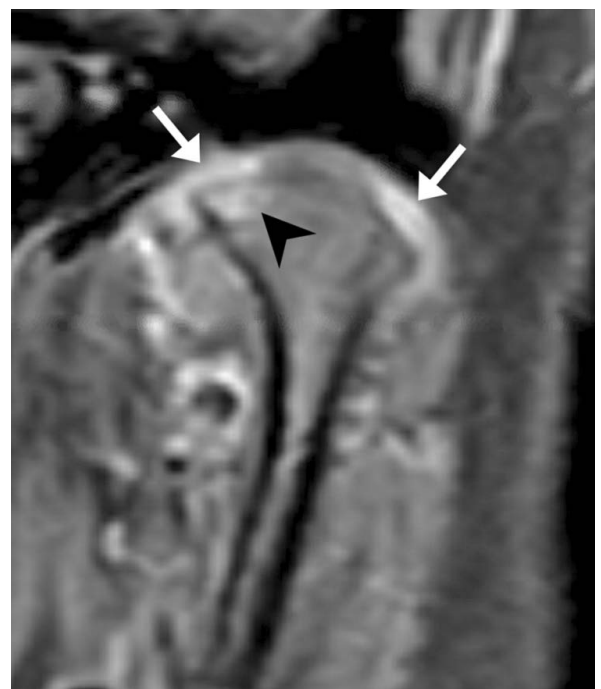


Figure 2.

*Coronal fat-saturated T1-weighted MRI with contrast, at the left mid-mandibular condyle shows synovial enhancement (arrows), focal condylar marrow enhancement (arrowhead).*

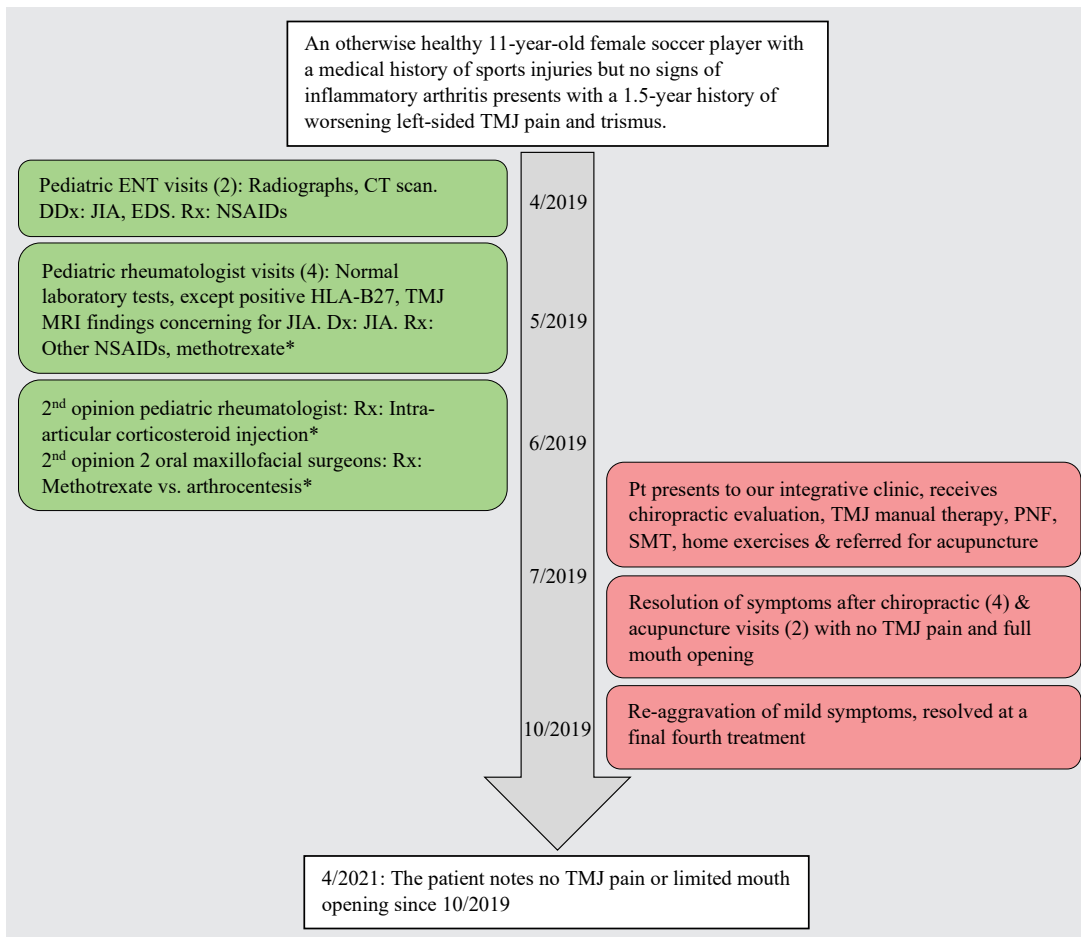


Figure 3.

*Timeline of events. Otolaryngologist (ENT), computed tomography (CT), juvenile idiopathic arthritis (JIA), prescription (Rx), non-steroidal anti-inflammatories (NSAIDs), magnetic resonance imaging (MRI). The green boxes on the left side show the patient's management prior to presenting to our clinic for chiropractic care (red boxes on right).*

*Treatments with an asterisk (\*) were not utilized by the patient. Timeline created according to Scientific Writing in Health and Medicine (SWIHM) and CARE guidelines.*

discussed the possibility of eventual arthroplasty. The other concurred with the use of methotrexate. This surgeon also stacked tongue depressors between the front teeth to maximally stretch the TMJ, and recommended the patient incrementally add tongue depressors each day. The family deferred medical treatment and sought chiropractic care based on recommendations from family and colleagues (see Figure 3 for the case timeline).

### Clinical findings

Physical examination of the patient revealed a thin, athlet-

ic adolescent of 1.52 meters and 38.6 kg, with a forward head posture and slight thoracic hyperkyphosis. The patient had significant tenderness to palpation of the left TMJ and a limited mouth opening of 20 millimeters (mm) with slight deviation of the mandible to the left during opening (measurements obtained using a TheraBite Range of Motion Scale®). The patient was unable to open the TMJ without moderate to severe pain. The cranial nerve examination was normal, there were no neurological deficits noted in the upper extremities, gait was normal and spinal range of motion was full. There were no abnormalities de-

tected in the lumbopelvic region with regards to posture or palpation findings.

### *Diagnostic assessment*

The chiropractor considered the patient to have a mechanical issue of the TMJ rather than a primary diagnosis of JIA, in view of her getting hit in the face with a soccer ball, displacement of the TMJ discs seen on MRI, and the absence of typical clinical, symptomatic, and laboratory features of JIA. A trial of manual therapy, home exercises, and acupuncture was recommended.

### *Therapeutic intervention*

Chiropractic treatment included gentle extra-oral myofascial release of the TMJ muscles including the masseter, temporalis, and pterygoids bilaterally. Intra-oral therapy was not performed as the patient could not open her mouth sufficiently and was in significant pain. Proprioceptive neuromuscular facilitation (PNF) was performed for TMJ opening, closing, and lateral deviation using light to moderate resistance and three to five repetitions for each movement, to patient tolerance.

Jaw opening was resisted isometrically using external pressure at the inferior aspect of the chin (Figure 4). After each five-second contraction of resisted opening,

the patient was asked to relax while the chiropractor performed a gentle passive stretch to open the TMJ for an additional five seconds, using inferiorly-directed pressure at the mentolabial sulcus. The goal of this exercise was to reciprocally inhibit the hypertonic jaw-closing muscles (e.g. masseter, temporalis) and facilitate a greater stretch to improve TMJ opening range of motion.

Jaw closing was resisted isometrically using external pressure at the mentolabial sulcus, for three to five seconds, followed by relaxation, and a passive stretch at end-range was not performed (Figure 5). For lateral deviation, gentle isotonic resistance was applied against the left side of the mandible to prevent aberrant ipsilateral deviation during mouth opening, and counteract hyperactivity of the contralateral (right) lateral pterygoid. A passive stretch was not applied after this maneuver.

Chiropractic spinal manipulation was performed using high-velocity, low-amplitude manipulations of restrictions at the cervical spine (C0, C1, C5 and C6) and thoracic spine (T2, T3, T4 and T7). Ischemic compression was performed at the cervical and thoracic paraspinal muscles, upper trapezius, levator scapulae, and suboccipital muscles.

The patient was given a home “jaw opener” exercise in which the patient slowly and actively opened the jaw, as



Figure 4.  
*Demonstration of resisted jaw opening.*



Figure 5.  
*Demonstration of resisted jaw closing*

far as possible without provoking moderate or severe pain. This was performed for multiple repetitions at least once per day for about a minute. The patient was instructed in-office on how to perform this exercise while looking into a mirror to ensure that the mandible was tracking properly, without lateral deviation.

The patient was also given postural home exercises. A stretch to improve thoracic extension and target the pectoralis muscles was to be performed for two to three minutes lying supine, with a pillow in the mid-thoracic region, arms abducted to 90°, and elbows slightly flexed. Chin tucks were to be performed at least once daily for 10 repetitions by retracting the head and neck with a brief hold at end-range.

The chiropractor also referred the patient to an in-office Licensed Acupuncturist. The patient attended sessions which took place later in the same day of the first and third chiropractic visits. At these appointments, 0.14x30 mm monofilament needles were utilized to stimulate the Stomach 7, Stomach 36, and Large Intestine 11 acupoints bilaterally. This treatment was well-tolerated.

#### *Follow-up and outcome*

The patient noted immediate relief with each treatment and a similar treatment protocol was performed at each visit for a total of four chiropractic and two acupuncture sessions. The first three chiropractic and two acupuncture visits were over the span of one month. At the third chiropractic visit, the patient presented with full mouth opening without pain of over 40 mm, and no locking or crepitus. Intermittent mild TMJ pain 1-2/10 NRS returned two months later which prompted the fourth and final chiropractic visit three months later which led to full symptom resolution.

Follow-up 18 months later revealed that the patient had no TMJ pain, locking, or crepitus, or difficulty eating since the final treatment, and had no other significant health issues aside from a gluteal muscle injury that developed when slipping while running. She never filled her methotrexate prescription, was not prescribed adalimumab, and did not have steroid injections, arthrocentesis or other medical procedures for the TMJ. As the patient was a minor, informed consent for treatment was obtained from the patient's parent. In addition, the parent provided written informed consent for this case report to be published.

#### **Discussion**

The clinical picture in this case was suggestive but not diagnostic of JIA. Although an insidious onset of chronic TMJ pain in an adolescent female can be consistent with JIA, and some cases of JIA present with TMD as the first sign<sup>22</sup>, the imaging findings of joint effusion and bone marrow edema could also be explained by disc displacement<sup>4</sup>. While the laboratory testing was not suggestive of JIA, it also could not rule it out, as these tests may be normal in oligoarticular JIA.<sup>23</sup> However, one test result actually reduced the likelihood of JIA, as HLA-B27 positivity is associated with a reduced risk of TMJ involvement in JIA.<sup>24</sup>

Given that the clinical features in the current case were equivocal, a trial of conservative manual therapy and acupuncture was the deciding factor in distinguishing the etiology of the patient's TMD. Because the patient's response was rapid and sustained without utilization of anti-rheumatic or anti-inflammatory medications, a diagnosis of JIA no longer fit. In addition, the patient had no joint symptoms over 18 months' follow-up, without taking medications. Such improvement would be rare if the patient truly had JIA, as only 7% of patients will achieve remission without medications during this time frame.<sup>25</sup>

The medical treatments recommended in the current case did not match the patient and her family's preference and have certain risks. Methotrexate, the most commonly used disease-modifying anti-rheumatic drug used in JIA, is associated with adverse effects in nearly half of patients.<sup>26</sup> Most adverse effects are non-serious, for example nausea, vomiting, abdominal pain, restlessness, and anxiety<sup>26</sup>, and may be mitigated by folic acid<sup>23</sup>. Intra-articular steroids also have adverse effects, as repeated injections can inhibit mandibular growth<sup>27</sup> and promote heterotopic bone formation<sup>28</sup>.

In the absence of a clear management algorithm, the stepped care model, which begins with less-invasive treatments<sup>10</sup>, could be an option for patients inconclusive for JIA affecting the TMJ. In this model, patients progress to more complex interventions if they do not respond to care.<sup>10</sup> Other models such as matched care (individualized interventions) or stratified care (categorization and treatment according to risk factors) may not be as optimal for adolescent TMJ, as diagnostic testing can be inconclusive.

Prior research has discussed the value of early identification and treatment of TMD in JIA in preventing on-

going damage to the orofacial structures.<sup>4,24</sup> While we do not contradict the value of this strategy, in the current case it appears a sense of urgency to recommend medical treatments for JIA led to conservative measures being overlooked. Because the diagnosis of JIA was not certain, it could have been more appropriate to begin treatment with a brief trial of conservative care including manual therapies, as in the stepped care model. This would provide the patient, family, and medical team with greater clinical insight regarding the patient response to care, and inform the strategy to recommend more complex interventions as needed.

In the current case report, the patient and her family only sought out conservative care after being recommended but deferring more invasive medical interventions. If the model of stepped care had been applied to this case, then the order of these treatments would have been different such that the manual therapy and/or acupuncture would have been recommended before the methotrexate, corticosteroid injection, and arthrocentesis.

Manual therapy in the current case targeted both the TMJ and the spine. Although these are separate regions, they are interdependent and may have been equally important in helping resolve the patient's TMD. Treatment of the TMJ may have inhibited hypertonic musculature, facilitated repositioning of the articular discs, and restored normal TMJ mobility. Spinal manipulative therapy may have been beneficial in addressing forward head posture, which is associated with hyperlordosis of the cervical spine, hyperextension at the occipital-cervical junction, retrusion of the mandible, alteration in masticatory muscle activity<sup>29</sup>, and reduced masticatory muscle pressure pain thresholds<sup>30</sup>. We suspect that treating the cervico-thoracic spine in tandem with the TMJ was beneficial in improving static posture and reducing compensatory TMJ dysfunction.

There may have been a synergistic effect of adding acupuncture to the patient's care. Acupuncture may provide benefits to overall health quality<sup>31</sup>, yet in addition, address local myofascial sources of TMJ pain<sup>14,31</sup>. Stomach 7 is one of the most common acupoints used in treatment of TMD, and due to its alignment with the masseter muscle, may help relax this muscle when stimulated.<sup>31</sup> The remaining acupoints were used as part of an overall acupuncture diagnosis and treatment, despite having no direct anatomical link to the TMJ. Stomach 36 was used

to facilitate the effects of Stomach 6, being in the same meridian, while Large Intestine 11 was used for clearing heat (TMJ-related inflammation).

The model of stepped care including conservative treatment for equivocal TMD in JIA may function particularly well in a health care organization offering chiropractic, acupuncture, and other integrative therapies. In this type of healthcare setting chiropractors work alongside medical specialists and have access to the same electronic medical records and laboratory and imaging results. In the current case, the chiropractor was employed by the same health care organization and was able to review the medical specialists' assessments and prior imaging, which informed the treatment approach.

We propose that the success of this case is partially related to the structural integration of chiropractic services, along with the manual therapies these providers utilize, across University Hospitals. Integrative medical interventions (e.g. chiropractic care, manual therapies, acupuncture, massage therapy, integrative medicine consultations) within health care organizations can facilitate a coordinated care model in which less invasive, but therapeutic, treatments are provided earlier in a patients' care pathway.<sup>32</sup>

### *Limitations*

As this is a case report, results may not be generalizable to a larger population. The exact relationship between the soccer ball hit and etiology of the patient's TMD was unclear. The patient had a history of less severe TMJ pain and limited opening leading up to this event, yet there was no prior TMJ examination or imaging to compare to. It is plausible but inconclusive that the soccer ball incident was responsible for acutely restricting the patient's mouth opening, triggering TMJ disc displacement, and causing TMJ edema as seen on MRI. Repeat imaging could have determined if the TMJ disc was successfully recaptured with conservative care, and if inflammatory signs were reduced. Repeat imaging was unnecessary as the patient had no further symptoms, and gadolinium contrast has risks. A TMJ-specific outcome assessment questionnaire was not utilized in this case which could have helped track the patient's progress over time. As a range of treatments were used (acupuncture, manual therapies, home exercise), it is inconclusive which treatment helped the most or if the patient's improvement was due to a combination



of these therapies. In addition, it is possible the patient's TMJ disorder could have self-resolved regardless of the interventions used.

### Summary

This case highlights the lack of a standardized treatment algorithm and the shortcomings of diagnostic imaging for adolescent TMD. The combination of these factors can allow for possible misdiagnosis and escalation of treatment, such as in the current case when advanced imaging findings suggestive of JIA prompted recommendations for methotrexate, corticosteroid injection, and arthrocentesis. However, the success of chiropractor-led management in this case highlights the potential utility of a stepped care model in which early conservative therapies are provided as both a diagnostic and therapeutic trial for those with TMD and an uncertain diagnosis of JIA. We suggest that this care pathway can inform the clinical decision-making prior to the use of higher levels of care such as disease-modifying antirheumatic drugs and/or injections. Chiropractors, acupuncturists, and medical specialists have an opportunity to collaborate within integrative settings to optimize the treatment for adolescents with TMD.

### Patient perspective

I felt that during all my doctor appointments I was not part of the decision of what was going to happen to me. I felt like the doctors did not think it mattered what I thought, but when I met [the chiropractor], he told me exactly what he was going to do before he did it and asked me how I was feeling the whole time. I felt like I was actually part of the conversation with [the chiropractor]. Before I saw [the chiropractor] we had been looking for about a year for someone who could tell me what was wrong with my jaw, and what we could do to fix it. Lots of doctors told me to go on Humira, but at the age of 12 my mom and I did not think that would be a smart choice. We then found [the chiropractor] who realized that my jaw was out of place and none of the other doctors had been able to see that. We had three appointments with [the chiropractor] that consisted of massaging my jaw and readjusting it. Now two years later my jaw feels normal again, all because of the acupressure and the help of [the chiropractor].

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