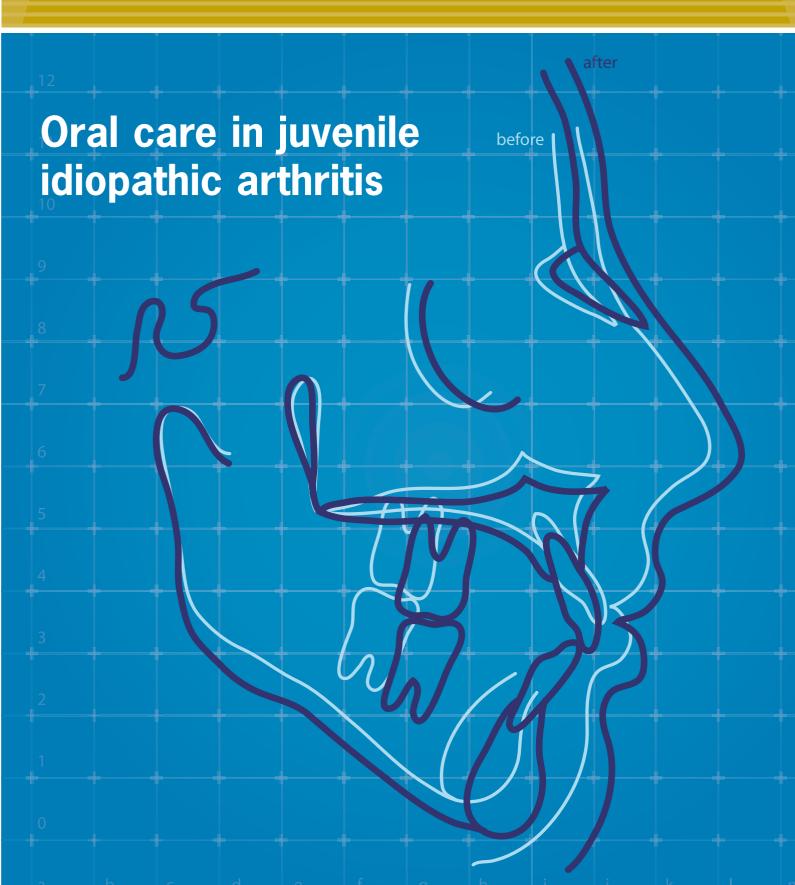
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## Oral health and orthodontic considerations in children with juvenile idiopathic arthritis: review of the literature and report of a case

Abstract: Juvenile idiopathic arthritis (JIA) is a severe disease of childhood, which comprises a diverse group of distinct clinical entities of unclear aetiology. Some abnormality of the immune system is present in all JIA cases. In its most severe clinical form, JIA may show localised and/or systemic complications, including functional impairment of the affected sites. This may result in variable growth and developmental anomalies. In many JIA cases, where the temporomandibular joint (TMJ) is affected, mandibular growth may be restricted, thus leading to the development of mandibular hypoplasia and/or retrognathism. As a result, it is not uncommon for JIA patients to present with skeletal Class II and open bite malocclusions. Furthermore, in JIA cases, early orthodontic intervention facilitates both the skeletal and the occlusal rehabilitation.

Increased prevalence of dental caries and periodontal disease in JIA cases may be attributed to a combination of aetiological factors, including difficulties in executing good oral hygiene, unfavourable dietary practices and side effects from the long-term administration of medication. In addition, an association between periodontal disease and JIA has been reported based on their similar pattern of clinical disregulation of the inflammatory process. This paper presents a brief description of JIA, with special reference to dental health and orthodontic treatment considerations. In addition, a case is presented where the appropriate orthodontic intervention led to the establishment of a normally functioning, as well as an aesthetically pleasing, occlusion.

**KEY WORDS:** juvenile idiopathic arthritis, craniofacial growth, TMJ, oral health, malocclusion, orthodontic treatment.

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# Introduction to juvenile idiopathic arthritis

Juvenile idiopathic arthritis (JIA) is a major connective tissue disease and one of the most common chronic illnesses of childhood.<sup>1</sup> Essentially, it is an early-onset form of arthritis affecting one or more joints. JIA manifests before the age of 16 and remains active for more than six weeks.<sup>2</sup> Its incidence is estimated at around 1-2:10,000 and its prevalence at 1:1,000.<sup>3-6</sup> The diagnostic criteria of JIA are reported in **Table** 1.<sup>7</sup> In general, three major types of JIA are recognised: oligoarthritic; polyarthritic; and, systemic. These three types are briefly outlined in **Table 2.**<sup>2</sup> The exact cause of JIA remains unclear, although it appears to be multifactorial, involving infectious, genetic and endocrine factors.<sup>8-12</sup>

The clinical course of JIA usually involves several sequential long or short periods of flare-ups and remissions.<sup>13-14</sup> It may affect any joint in the body and, in its most severe form, it may be accompanied by systemic complications of the cardiovascular system, the urinary system and the eyes.<sup>14</sup> Systemic or local disturbances of growth may be observed in JIA patients,<sup>13,14</sup> depending on which joints are affected. This may be related to either the disease or to the medication.<sup>15</sup> Most JIA cases require chronic administration of multi-drug medication, including non-

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#### **TABLE 1: Diagnostic criteria and types of juvenile** idiopathic arthritis.<sup>7</sup>

Age at onset younger than 16 years;

- arthritis in one or more joints (defined as swelling or effusion, or the presence of two or more of the following signs: limitation of range of motion; tenderness or pain on motion; and, increased heat);
- duration of disease  $\geq 6$  weeks;
- type of onset of disease during the first six months (polyarthritic when five or more joints are affected; oligoarthritic when less than five joints are affected; systemic when intermittent fever is present and other systems of the body are involved; psoriatic; enthesitis related; and, other), and
- exclusion of other forms of juvenile arthritis.

steroidal anti-inflammatory, steroidal, disease-modifying antirheumatic, and immunosuppressive drugs. The main objective of such treatment is to restore and maintain quality of life for patients, while alleviating pain and controlling inflammation, thereby preventing or minimising joint destruction and deformity.<sup>16</sup>

Despite treatment, an effective permanent change in the clinical course of the disease may not be achieved until adolescence where, in up to 70-85% of cases, a spontaneous remission is observed.<sup>17</sup> Additionally, prolonged administration of medication may inflict severe side effects because of its toxicity, especially with regard to skeletal growth and development. During active phases of the disease, ossification at the epiphyses is accelerated, followed by premature closure of the epiphyseal growth plates in later stages. This may eventually result in retardation of skeletal growth and joint dysfunction.<sup>2</sup> In many JIA cases where the temporomandibular joints (TMJs) are affected by the disease, growth of the mandible is restricted, resulting in severe functional and aesthetic problems of the craniofacial complex.<sup>18-23</sup> Complications due to the disease itself, as well as its treatment, may affect oral health, as evidenced by the reported increased prevalence of caries and periodontal disease in JIA patients.24-26

#### **TMJ involvement in JIA cases**

The reported prevalence of TMJ involvement in patients with JIA varies between 50% and 87%.<sup>27-34</sup> In many such cases, temporomandibular joint dysfunction (TMD) is observed, presenting with restricted mandibular movements, and reduced and/or painful functional or resting activity of the masticatory muscles. Furthermore, the underlying pattern and direction of dentofacial growth may be disturbed.<sup>21,22,27,28,30,35-38</sup> As a result, TMJ involvement in JIA patients is often associated with the development of certain craniofacial and dental features including:

- decreased mandibular length;<sup>18,19,22,39-42</sup>
- Class II division 1 malocclusion, mostly due to mandibular

## TABLE 2: Characteristics of the three major types ofjuvenile rheumatoid arthritis.2

	Polyarthritic	Oligoarthritic	Systemic
Relative frequency	30%	60%	10%
Number of joints			
involved	≥5	≤5	variable
Sex ratio (F:M)	3:1	5:1	1:1
Extra-articular			
involvement	moderate	not present	prominent
Chronic uveitis	5%	20%	rare
Seropositivity –			
rheumatoid factor	15%	rare	rare
Seropositivity –			
antinuclear antibodies	40%	85%	10%
Prognosis	Moderately	Excellent	Moderate
	good		

retrognathism;<sup>19,22,31,39,40,42-44</sup>

- hyperdivergent facial type, increased mandibular plane angle, and decreased posterior facial height;<sup>21,22,33,39,40-42,44,45</sup>
- facial asymmetry in cases with unilateral TMJ involvement,<sup>20</sup> and,
- decreased overbite, open bite or open bite tendency.<sup>18,21,22,29,39,41,43,46,47</sup>

The unfavourable craniofacial growth pattern observed in many JIA patients is usually regarded either as primary or secondary. Primary unfavourable growth pattern is attributed to the disturbed growth of the affected mandibular condyles,<sup>18,19,22,39,46,48,49</sup> while secondary is attributed to the children's impaired oral function<sup>21,45</sup> and/or medication-related somatic growth retardation.<sup>20,40</sup>

Clinical examination may not be considered as an accurate method of diagnosing TMJ involvement in JIA patients.<sup>18,19,30,47,49,50</sup> Therefore, the application of an appropriate TMJ imaging method is often indicated.<sup>51-53</sup> Techniques used include panoramic radiography (PR), tomography, arthrography, fluoroscopy, computerised tomography, magnetic resonance imaging (MRI), and radionuclide imaging.<sup>54-56</sup> There is almost universal agreement that the examination of the osseous surfaces of the TMJ is facilitated by the application of tomography rather than plain film techniques.<sup>57-61</sup> Fine bone details can be visualised without projection limitations, while the real shape and size of anatomical structures is displayed.<sup>62,63</sup> In contrast, imaging of soft tissues of the TMJ is investigated in most cases by the use of MRI, which is non-invasive and does not result in the patient being exposed to ionising radiation.<sup>64,65</sup>

#### Dental health considerations in JIA patients

Although TMJ involvement in JIA is well described, only a few studies report its potential adverse effects on dental health. Several factors related to JIA may unfavourably affect oral health. TMJ-related dentofacial abnormalities may be considered as contributing factors in the aetiology of oral diseases. TMD accompanied by impaired masticatory function and functional impairment of upper limbs may affect toothbrushing competence.<sup>66-68</sup> Dietary practices such as the consumption of softer, more sugary foods in frequent small amounts, sweets given as consolation, the use of oral medication containing sugar<sup>43,69-71</sup> and factors related to psychological issues delaying optimal dental care,<sup>26,72</sup> all adversely affect oral health.

In JIA cases, an increased prevalence of dental caries<sup>24,25,73,74</sup> and a higher risk for periodontal disease have been reported.75,76 Interestingly, some common clinical and pathogenic features of periodontitis and rheumatoid arthritis have been recognised.<sup>77,78</sup> This implies a possible association between the two diseases that may share a common underlying disregulation of the inflammatory response.<sup>77</sup> Alternatively, the reported higher prevalence of periodontal disease may be considered as secondary to the overall higher plaque accumulation recorded in JIA patients,<sup>79</sup> or to the longterm administration of medication resulting in immunosuppression, xerostomia, stomatitis and gingival overgrowth.<sup>80</sup> Whatever its cause, poor oral health is considered to be potentially detrimental for the systemic condition of JIA cases because untreated dental caries and/or periodontal disease, combined with poor oral hygiene, may increase the risk for systemic infection, especially if the patient is taking immunosuppressive drugs such as methotrexate.<sup>26</sup>

In all JIA cases, intensive prophylactic and therapeutic measures are required to prevent or reduce the potential damage to the dental and periodontal tissues. Regular dental and orthodontic examination, case-specific oral hygiene instructions, topical and systemic use of fluoride, appropriate dietary modification, and prescription of sugar-free medicines are of prime importance for maintaining optimal dental health in patients with JIA.<sup>26,72</sup> As immunosuppression is a common side effect of several anti-rheumatic drugs, it may be wise to apply antibiotic prophylaxis before dental management of certain JIA patients.

In addition to advice on prevention, and treatment for dental diseases, dentists should also provide information to the patient and their family regarding JIA's possible consequences for oral health. It is not uncommon for lay people parenting JIA patients to be totally unaware of the effects of JIA on oral health.<sup>81</sup>

#### **Orthodontic treatment considerations in JIA patients**

There is extensive documentation regarding the impact that TMJ involvement may have on the pattern of craniofacial growth in JIA patients, resulting in the development of certain malocclusions.<sup>19-22,31,29</sup> Accordingly, case-specific objectives of orthodontic treatment in JIA patients should be assessed, always keeping in mind that the principal objectives in the management of the systemic disease are relief of pain and discomfort, avoidance of permanent joint damage and preserving an acceptable level of quality of life.<sup>23</sup> In any JIA case, the indicated orthodontic treatment can be applied on the condition that TMJ inflammation caused by the systemic disease is controlled by proper medical care.<sup>82,83</sup> In cases where limitation of any mandibular movement is observed, the application of a properly designed occlusal splint is indicated to alleviate TMD symptomatology and, while unloading the joints, to re-establish normal masticatory physiology.<sup>28</sup>

addressed by orthodontic/orthopaedic treatment with functional appliances.  $^{\rm 22}$ 

It is not ubiquitously accepted that orthodontic treatment should be instigated early in all cases presenting malocclusions warranting prompt intervention.<sup>22,84</sup> The reluctance of many practitioners to treat skeletal Class II malocclusions of JIA patients with functional appliances while the disease remains active is due to the supported risk of flareup in the articular surfaces of the TMJs, resulting in a net bone loss in the condylar growth centre.<sup>85</sup> Whether justified or not, such delayed intervention would result in excluding any orthopaedic effect from orthodontic treatment, since full remission of JIA does not occur until adolescence. Thus, in the majority of cases, the only choices remaining for such cases would be treating them in late adolescence, orthodontically-induced occlusal camouflaging of the underlying skeletal discrepancy or, even, in extreme cases, the application of a combination of orthodontic treatment and orthognathic surgery.<sup>21,86,86-89</sup> In the latter cases, costochondral grafting may be applied to serve as replacement for the completely destroyed condylar head.<sup>90,91</sup>

In contrast to postponing treatment until cessation of growth, early treatment with functional orthodontic appliances aims to achieve and maintain occlusal balance, and rehabilitate and preserve TMJ function, while allowing for uninhibited mandibular growth.<sup>28,32,43,67</sup> There is evidence that if functional conditions are created, growth has the potential to normalise.92 Close monitoring of the reaction of the condylar growth centre to functional stimuli is of utmost importance to decrease the risk of possible undesirable side effects. After the normalisation of craniofacial growth pattern, the application of fixed orthodontic appliances facilitates in finishing the orthodontically induced occlusal rehabilitation. In JIA cases presenting non-skeletal malocclusions, timely application of fixed orthodontic appliances only is indicated. In any JIA case warranting some kind of orthodontic treatment, it must be noted that the administered medication may interfere with bone physiology, adversely affecting bone turnover, thereby restraining orthodontic tooth movements. In all cases, light orthodontic forces should be applied to eliminate the risk of side effects such as apical root resorption, while facilitating optimal tooth movement.<sup>23</sup>

#### Case report

A female patient, aged 10 years and six months, presenting polyarthritic JIA (involving at its onset both knees and several joints of the hands), diagnosed at the age of two at the Paedo-Rheumatological Department of the Paediatric Clinic "Aglaia Kyriakou" of Athens, was referred for orthodontic consultation with special emphasis placed towards a comprehensive evaluation of the TMJ physiology. At the patient's initial admittance, the disease was in remission using medication that included methotrexate in combination with certain corticosteroids. Her medical history revealed a penicillin allergy. The physical examination did not show any clinically significant aberrations from normal height and weight percentiles. Her extra-oral examination showed symmetrical craniofacial features along with normal proportions of her anterior



FIGURE 1: The anterior facial view of the patient at rest and smiling at her initial admittance.



FIGURE 2: The posteroanterior cephalometric radiography did not reveal any significant asymmetry of any craniofacial structure.

lower facial height (Figure 1). These were also evidenced by the evaluation of the posteroanterior cephalometric radiograph (Figure 2). In contrast, her profile view was concave; however, at rest position some interference of the lips was evident in addition to some tension of the genial muscle (Figure 3). Overall, her prominent ears affected her facial aesthetics unfavourably and the aesthetics of her smile were impaired by the malaligned anterior teeth (Figure 1).

The intra-oral examination revealed a late mixed dentition stage. She had a Class II division 1 malocclusion with bilateral posterior crossbite, severe anterior crowding of both arches, a reduced overbite and an increased overjet (Figure 4). No dental pathology was observed with the exception of some plaque-induced gingival inflammation restricted mostly to the crowded areas. The clinical TMJ evaluation did





FIGURE 3: The profile view of the patient and the lateral cephalometric radiography at her initial admittance.



FIGURE 4: The anterior and lateral view of the patient's occlusion before the initiation of orthodontic treatment.

not show any signs and symptoms of TMJ dysfunction and no restriction was observed in her range of movements. The examination of the panoramic and the corrected sagittal tomography revealed some flattening and erosive lesions on both condylar articular surfaces (Figures 5 and 6). The study of the lateral cephalometric radiograph showed a hyperdivergent facial growth type presenting skeletal Class II division 1 malocclusion, mostly due to the posteriorly and downward rotated direction of mandibular growth. An increase of the lower to the total anterior facial height ratio was noted, in addition to a clinically significant decrease of the posterior to anterior facial height ratio. These observations implied that mandibular growth was impaired, probably as a result of the condylar growth centres having been affected by JIA.

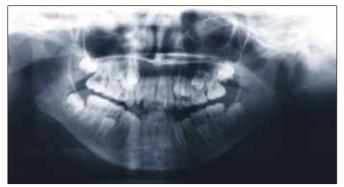


FIGURE 5: The initial panoramic radiograph of the patient.

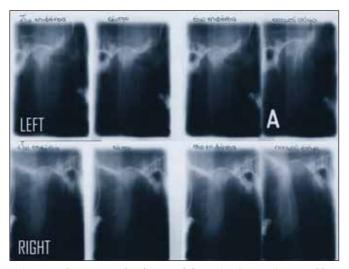


FIGURE 6: The tomographical views of the patient's TMJs (corrected by the use of submental-vertex radiograph).



FIGURE 7: The anterior and lateral view of the patient's occlusion after the removal of fixed orthodontic appliances.



FIGURE 8: The anterior facial view of the patient at rest and smiling after the completion of her orthodontic treatment.

The severe dental crowding, in conjunction with the patient's unfavourable facial growth pattern and the incompetent lips, determined the orthodontic treatment plan, comprising the extraction of all four first premolars. The case was assessed as one requiring maximum anchorage, which initially involved a palatal bar connecting the banded upper first permanent molars, followed by a high-pull headgear. Full-mouth fixed orthodontic appliances were sequentially applied for a total period of two years and six months. On successive appointments, specific emphasis was placed on the importance of the patient maintaining optimal dental hygiene. After the completion of treatment the appliances were removed and the retention protocol followed. A Hawley-type retainer was used for the upper arch and a fixed lingual retainer on the lower anterior teeth.

Overall, the post orthodontic extra-oral and intra-oral clinical examination of the patient was quite satisfactory with regard to the occlusal and aesthetic dental and facial characteristics. No dental pathology was observed regarding the development of either carious (or even white spot) lesions or periodontal complications. The gingival contour of all teeth was firm and lacking severe inflammatory signs (no bleeding on probing, periodontal sulcus depth less than 3mm) although some localised plaque accumulation was evident. The occlusal view of both dental arches was normal and co-ordinated with each other, and their midlines coincided (Figure 7). Overall, an aesthetically pleasing smile was accomplished by the proper alignment of the upper anterior teeth (Figure 8). The post-treatment profile line of the patient had become almost orthognathic, although reminiscent of its former concave form (Figure 9). Even more important was the fact that the vertical dimension of the craniofacial growth was controlled. The patient's lips were in contact at rest position, although the upper lip was flatter and the nose a bit more prominent. The post-treatment panoramic radiograph did not reveal any clinically significant apical root resorption, interproximal dental surfaces caries, or loss of periodontal support. The mesiodistal axial inclination of maxillary left lateral incisor and canine did not appear ideal as viewed in the panoramic radiograph; however, it was





FIGURE 9: The profile view of the patient and the lateral cephalometric radiography after the completion of her orthodontic treatment.



FIGURE 10: The final panoramic radiograph of the patient.



FIGURE 11: The juxtaposition of the tracings of the lateral cephalometric radiographs taken before (white line) and after (navy blue line) the completion of the orthodontic treatment.

satisfactory on an intra-oral view. A further concern was the direction of eruption of the lower third molars, which warranted some attention in future recall visits (Figure 10).

The post-treatment lateral cephalometric study, although still determining a Class II skeletal relationship, revealed a reduction of initial values of respective variables. In addition, improved ratios of lower anterior to total anterior facial height, and posterior to anterior facial height, were noted. The interincisal angle was almost normalised owing to the favourable uprighting of upper incisors and some labial inclination of the lower incisors (**Figure 11**).

Finally, but most importantly, no signs or symptoms of TMJ dysfunction were recorded throughout orthodontic treatment.

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