Temporomandibular joint involvement in a cohort of patients with Juvenile Idiopathic Arthritis and evaluation of the effect induced by functional orthodontic appliance: clinical and radiographic investigation

**Abstract**

**Aim** The aim of the study was to assess possible correlations between the clinical parameters of temporomandibular joint (TMJ) arthritis and pathologic MRI findings of the TMJ in patients affected by juvenile idiopathic arthritis (JIA), and the effect of a functional orthodontic therapy on the evolution of TMJ disorders.

**Material and methods** A prospective clinical and nuclear magnetic resonance (NMR) investigation was conducted on a sample of 53 patients (41 female, 12 male) with JIA, treated for 24 months with an Andresen appliance. The involvement of TMJ was defined by clinical and radiological signs. NMR assessments were performed in closed and maximum opening mouth position before (T0) and at the end of functional orthodontic therapy (T1).

**Results** Fifteen patients showed physical and radiologic TMJ abnormalities. Changes were not uniformly distributed among the different JIA subtypes. Patients with polyarticular JIA (≥5 peripheral joints affected) showed more destructive bony changes. No correlation existed between clinical symptoms and NMR alterations.

**Conclusion** The prevalence of TMJ involvement in patients suffering of JIA, and the improvement of TMJ and muscular pain associated with the use of functional appliance found in the present study, suggest an alert for TMJ dysfunction in patients with JIA and demonstrate the utility of functional orthodontic therapy in preventing the morbidity associated with TMJ arthritis in JIA.

**Keywords** Juvenile Idiopathic Arthritis; Nuclear magnetic resonance; Temporomandibular Joint Arthritis.

**Introduction**

Juvenile Idiopathic Arthritis (JIA), previously termed Juvenile Chronic Arthritis (JCA) in Europe and Juvenile Rheumatoid Arthritis (JRA) in North America, is one of the most common chronic diseases in childhood, with an estimated annual incidence of 10 per 100,000 children worldwide, and a prevalence of 65 per 100,000 [Minden, 2007; Baum, 1977]. The term JIA indicates a chronic arthritis with an onset before the age of 16, persisting for at least 6 or more weeks, without a known cause [Stoll et al., 2008]. JIA encompasses a spectrum of diseases that widely varies for onset characteristics, clinical course, associated features and ultimate outcomes. JIA includes seven different subtypes: systemic JIA (sJIA), oligoarticular JIA, rheumatoid factor negative polyarticular JIA, rheumatoid factor positive polyarticular JIA, enthesitis related arthritis (ERA), psoriatic arthritis, and undifferentiated JIA. All subtypes of JIA are characterised by persisting joint swelling caused by accumulation of synovial fluid and thickening of the synovial lining. There is evidence to support the involvement of different components of the immune system in the etiopathogenesis of JIA [Lee et al., 2012]. The synovial tissue contains various inflammatory cells including neutrophils, plasma cells, dendritic cells, and a high number of activated T-cells. Thus there is compelling evidence that activated T-lymphocytes play a role in the pathogenesis of JIA [Bendersky et al., 2012]. Involvement of the other component of the immune system is also evident in JIA. For instance a significant proportion of children with JIA have antinuclear antibodies, and some children have the rheumatoid factor, suggesting a role of the humoral component of the immune system [Yilmaz et al., 2001]. The Temporomandibular joint (TMJ) can be affected by JIA with a prevalence that varies between 38% and 72%, depending on the JIA subtype [Pedersen et al., 2001], the diagnostic method used and the population studied [Bayar N et al., 2002; Twilt et
al., 2004]. The worst outcomes are reported in patients with systemic or polyarticular arthritis; the pathological changes include hypertrophic inflammatory synovitis characterised by cellular infiltration, with proliferation and congestion of blood vessels. In chronic disease, pannus infiltration gradually extends over the surface of the articular cartilage, leading to significant limitations in mandibular growth and development [Bakke et al., 2001]. Tendons and muscles become edematous and infiltrated with cells and any effect on the chondrogenic zone of the TMJ may lead to delayed growth. Joint damage and the changes in muscle function cause developmental changes in the mandibular morphology and an unstable occlusion. This results in a decreased total mandibular function causing further joint damage, and a vicious circle is established [Pirttiniemi et al., 2009]. Maximal molar bite forces and endurance times are significantly reduced in children with JIA [Farronato et al., 2011]; the effects on facial growth are well described and may be predominantly due to condilar abnormalities with characteristic features of micrognathia (30% of cases), Angle Class II malocclusion (69%), anterior open bite, lower incisor crowding and incisal protrusion. The characteristic “bird face deformity” is often used to describe the facial morphology associated with JIA, however this feature is not pathognomonic of the disease [Portelli et al., 2009]. Clinical signs associated with TMJ inflammation include pain with jaw excursion, asymmetric jaw opening, crepitation and absence of the jaw translation movement. TMJ arthritis can be asymptomatic and not associated with any detectable swelling at the examination, but facial growth disturbance may still be present without TMJ symptoms.

The gold standard for temporomandibular disorders diagnosis is represented by NRM; some authors suggest NRM of the TMJ in patients who less than 4 years of age at the onset of JIA, and in those with systemic JIA, regardless the age of onset [Argyropoulou, 2009].

Treatment of TMJ arthritis in JIA included systemic drugs, such as methotrexate (MTX), as well as local treatments with arthrocentesis, intra-articular injections of steroids or sodium hyaluronate [Moystad et al., 2008]. In JIA children the use of Class II functional appliances is a common clinical practice; these devices are used to modify the unfavourable growth pattern, for advancement and anterotation of the mandible in order to improve mandibular retrognathia. In the arthritic child, however, some authors advise not to strain the TMJ with mandibular advancement procedures, because they fear accelerated skeletal destruction of the TMJ as a result of the increased bone turnover rate [Van Venrooy, 1985]. A recent systematic review of the literature concludes that the evidence on orthodontic treatment principles for JIA children with temporomandibular joint involvement is very low because of the heterogeneity of the studies actually present in the literature [Von Bremen and Ruf, 2011].

The aim of this study was to assess possible correlations between the clinical parameters of temporomandibular joint arthritis and pathologic MRI findings of the TMJ in patients affected by JIA, and the effect of a functional orthodontic therapy with a Class II activator.

**Material and methods**

Fifty-three patients (41 girls and 12 boys) with age ranging from 5 to 17 years were selected from January 2009 to February 2011 at the Department of Paediatric Rheumatology of the University of Messina, “G. Martino” Hospital. Each patient had JIA based on the ILAR criteria with a median age of 5.5 years at disease onset and a median disease duration of 2 years. For the study, the following data were collected.

- Age of onset of JIA.
- Disease subtype.
- Sex and family history.
- Number of affected joints.
- Age at onset of TMJ involvement (first complaints reported and/or radiographic alterations).
- Unilateral or bilateral temporomandibular involvement.
- Symptoms linked to TMJ involvement (headache, grinding, crepitations, clicking, reduced mouth opening, chewing pain).

The following laboratory data were acquired.

- Antinuclear anti-body (ANA).
- Rheumatoid factor (RF).
- Erythrocyte sedimentation rate (ESR).
- C-Reactive protein (CRP).

Clinical examination included facial evaluation, intraoral examination and functional examination of the TMJ. To assess TMJ function the following were assessed: condylar excursion during mouth opening and closing; presence of mandibular deviation during mouth opening, or at maximum vertical opening; presence of clicking and/or pain. Manual palpation of the condyle was useful to detect abnormal condylar movements; auscultation of the TMJ using a stethoscope and palpation of the TMJ and masticatory muscles were also performed. A specific questionnaire was devised to evaluate the orofacial pain considering pain characteristics, location, intensity, duration, individual perception, triggering factors and aggravating movements. Each patient with TMJ involvement was treated for 24 months with an Andersen activator, with a central screw and a vestibular arch, fabricated through a wax bite registration: with this procedure a piece of softened wax is placed on the upper dental arch and the patient is guided to close the mouth in mandibular protrusion with coincidence of the upper and lower midlines. The registration bite was very thin in order to obtain a minimum vertical dimension. The activator was modified every three months increasing its posterior vertical dimension in order to stimulate the mandibular ramus growth thanks to the dislocation of
the mandibular condyle. Magnetic resonance imaging included T1 (pre-treatment) and T2 (post-treatment) sequences to assess condylar configuration, flattening of condyles, glenoid fossa, presence of bony erosions, disk abnormality, range of motion, presence of joint effusions or pannus. The severity of TMJ inflammation seen on MRI was assessed as follow, based on the grading system developed by Cahill.

- Age Grade 1: Normal Joint.
- Age Grade 2 (Acute): joint effusion, synovial thickening or marrow oedema.
- Age Grade 3 (Chronic): juxta-articular erosions.
- Age Grade 4 (Chronic): Condylar sclerosis or loss of articular cartilage.
- Age Grade 5 (Chronic): TMJ ankylosis.

The protocol was approved by the Scientific and Ethic Committee of the A.O.U. “G. Martino”.

**Results**

In the sample of patients we observed 23 cases of oligoarticular JIA, 21 of polyarticular JIA, 7 of systemic JIA, 1 psoriatic arthritis and 1 of enthesitis related arthritis (Table 1). In the sample selected for the study, only 15 patients (10 girls, 2 boys) showed TMJ involvement; 7 patients were affected by oligoarticular JIA, 8 patients by a polyarthritis JIA. Duration of disease was similar between the two groups, with a median age of 9.5 years; 80% of these patients were positive for rheumatoid factor, while none was positive for ANA or HLA-B27. ESR was increased in 73.3% of patients. Six patients were affected by uveiti; 46.6% of patients were treated with nonsteroidal antiinflammatory drugs (NSAIDs) and MTX, 20% were taking NSAIDs, MTX and Etanercept and the remaining 33.4% were treated with NSAIDs or MTX as monotherapy. Among the 15 patients with TMJ involvement the pathologic findings observed were (Table 2): bilateral TMJ click (25); bilateral functional limitation (12); monolateral TMJ click (7); monolateral chewing pain (7); monolateral chewing pain + monolateral TMJ click (7); bilateral chewing pain + monolateral functional limitation (7); bilateral functional limitation + monolateral TMJ click (7); monolateral swelling + bilateral click + deviation (6); bilateral functional limitation + deviation (7); bilateral chewing pain + functional limitation + deviation (7); deviation (7).

All patients with TMJ involvement were treated with Andresen activator for 24 months. Comparing pre-treatment and post-treatment clinical signs, a significant improvement of TMJ symptoms was observed; TMJ pain, jaw deviation and mouth opening limitation were considerably improved in almost all patients. A better facial profile with a reduced skeletal discrepancy and an increased function was observed. All patients reported a significant regression of pain, morning rigidity and chewing problems; subsided TMJ inflammation and a significant reduction of inflammation exacerbation frequency in the sample studied has been observed. In the unilateral TMJ arthritis it was observed a more balanced development of the mandibular ramus, with a significant reduction of mandibular deviation in the direction of the affected side.

**Discussion**

TMJ arthritis is a common finding in JIA, and affects all disease subtypes. The majority of patients selected for this study were female subjects positive for rheumatoid
factor with polyarticular JIA. None of them was ANA and HLA-B27 positive and the ESR was increased. This pattern is consistent with a previous description of risk factors in patients with TMJ inflammation [Kuseler et al., 2005]. Bilateral TMJ involvement was more common than unilateral. Although our patients reported jaw pain, the presence of pain did not significantly correlate with the presence of TMJ effusions of erosions on NMR. Condilar NMR alterations were observed more frequently in patients who did not have pain compared to symptomatic ones. However NMR findings demonstrate that jaw deviation or limited maximal mouth opening, although without pain, often reflect an advanced TMJ arthritis in most of the examined patients. TMJ clinical evaluation was quite difficult, especially in young children, because symptoms such as pain can often be evaluated only indirectly, by asking to the parents. Furthermore, some clinical symptoms could be masked by antirheumatic therapy.

According to the results of the present study is possible to state that an early treatment of patients with TMJ involvement, using a Class II activator, could prevent severe problems of the TMJ caused by hypoplastic condyles and growth alterations of the mandible [Marsico et al., 2011]. The increase of posterior vertical facial height and the consequent mandibular counterclockwise rotation produce an improvement of the occlusion [Portelli et al., 2012], masticatory function, profile [Bellintani et al., 2005], and cervical spine posture [Tecco et al., 2005]. Imaging of the TMJ is important for diagnosis and treatment planning; NMR is accepted in the literature as the gold standard in the early detection for TMJ involvement in JIA [Kuseler et al., 1998]. Conventional Rx such orthopantomography, show destructive changes in the TMJ only in a late stage, and CT scans do not show soft tissue structures like the articular disk [Matarrese et al., 2006; Portelli et al., 2013] On the basis of the results obtained, an early screening program to detect TMJ arthritis in children with JIA would allow an earlier therapeutic intervention that could prevent subsequent deformities and preserve the normal jaw structure and function. TMJ involvement in children with JIA can lead to further suffering and for this reason any type of intervention that improve TMJ disorders is well accepted both by patients and their families.

Conclusion

- Experienced paediatric dentists in cooperation with paediatric rheumatologists should be trained to recognise TMJ disorders in patients suffering of JIA. Regular clinical examination and early treatment, together with progressive monitoring of the craniofacial development is advised.
- Early treatment of patients with TMJ involvement with a Class II activator could prevent severe TMJ disorders. Increasing of posterior vertical facial height and the consequent mandibular counterclockwise rotation can improve occlusion, masticatory function and facial profile.
- Not all children with JIA have TMJ disorders and further studies are needed to identify risk factors for TMJ involvement, in order to propose interceptive therapies that may prevent more severe damages.

References

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