

## Article

# Electromyographic Activity of Masticatory Muscles in Subjects with Juvenile Idiopathic Arthritis: A Case—Control Study

Francesco Caroccia <sup>1,2</sup> , Ludovica Passanello <sup>1</sup>, Rossana Pipitone <sup>1</sup>, Francesco Moscagiuri <sup>1</sup>, Paolo Asperio <sup>3</sup>,  
Alessandra Lucchese <sup>4</sup> , Luciana Breda <sup>5</sup> and Michele D'Attilio <sup>1,\*</sup> 

<sup>1</sup> Department of Innovative Technologies in Medicine & Dentistry, University "G. d'Annunzio" Chieti-Pescara, 66100 Chieti, Italy; francesco.caroccia@unifi.it (F.C.); ludovica.passanello@studenti.unich.it (L.P.); rossana.pipitone@studenti.unich.it (R.P.); francesco.moscagiuri@unich.it (F.M.)

<sup>2</sup> Unit of Orthodontics, Department of Experimental and Clinical Medicine, The University of Florence, 50127 Firenze, Italy

<sup>3</sup> Division of Maxillo Facial Surgery, Cardinal Massaia Hospital, 14100 Asti, Italy; asperio@asl.at.it

<sup>4</sup> Unit of Orthodontics, School of Dentistry, Vita-Salute San Raffaele University, 20132 Milan, Italy; lucchese.alessandra@univr.it

<sup>5</sup> Department of Paediatrics, University of Chieti-Pescara, 66100 Chieti, Italy; luciana.bredach@gmail.com

\* Correspondence: michele.dattilio@unich.it; Tel.: +39-335-7489425

**Abstract:** The aim of this study was to evaluate the effect of juvenile idiopathic arthritis (JIA) on the activity of masticatory muscles using surface electromyography (sEMG). Forty-one JIA subjects (ten males, thirty-one females; average age 13 years  $\pm$  3) and thirty-two healthy control subjects (twenty-seven females, five males; average age 14 years  $\pm$  2) were recruited. sEMG of anterior temporalis (TA), masseter (MM), and sternocleidomastoid (SCM) muscles was performed by using the occlusal contact analyzer software called Teethan (BTS S.p.A., Garbagnate Milanese, Milan, Italy). Comparisons between groups were assessed with unpaired *t*-tests for non-normally distributed data and with Mann–Whitney U tests for normally distributed parameters. The JIA group showed a significant increased percentage overlapping coefficient of TA (POC TA) ( $p = 0.01$ ) and impact index (IMP) ( $p = 0.003$ ). No significant differences were observed for the POC MM, POC SCM, percentage overlapping coefficient between posterior and anterior teeth contact (BAR), the torsion index (TORS), and the asymmetry index (ASIM). Masticatory muscles seemed to be slightly affected by JIA. sEMG could be an effective aid in the early clinical detection of TMJ involvement in JIA. Further research is needed to confirm its validity.

**Keywords:** juvenile idiopathic arthritis; temporomandibular joint; temporomandibular disorders; electromyography; masticatory muscles



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## 1. Introduction

Juvenile idiopathic arthritis (JIA) is the most common chronic inflammatory rheumatic disease that affects children [1]. It refers to a group of heterogeneous disorders sharing the common feature of chronic inflammatory arthritis of unknown cause. This inflammatory status lasts longer than 6 weeks and has an early onset before the age of 16 [2]. An incidence and prevalence of 1–22 in 100,000 and 7–150 in 100,000, respectively, was estimated [3]. JIA is defined as a multifactorial disorder, and its etiology still remains unclear. The most reliable theory is the one supporting the influence of immunogenic mechanisms underlying several genetic and environmental factors (e.g., infections, antibiotic exposure, stress, and trauma) [4–6], even if a minority of the population seems to show familial factors [7]. Human leukocyte antigen (HLA) B-27 and other HLA tissue types are the most commonly mentioned genetic factors [8], but many uncertainties about the role of these factors—as well as many others—remain in JIA pathogenesis.

JIA was classified by the International League of Association for Rheumatology (ILAR) in 1995, then revised in 2001 and subdivided in seven categories according to clinical

features, treatment modalities, and disease prognosis. The identified categories were: systemic arthritis, oligoarthritis (persistent or extended), polyarthritis rheumatoid factor (RF)-positive, polyarthritis RF-negative, enthesitis-related arthritis (ERA), psoriatic arthritis (PsA), and undifferentiated arthritis. The latter category involves patients not fitting any of the above-mentioned criteria or those fitting more than one [9]. A process has been initiated by the Pediatric Rheumatology International Trials Organization (PRINTO) to provide evidence-based, validated new criteria in order to identify the various types of JIA and distinguish the disorders observed in both adults and children from those observed only in children [10]. Like other forms of arthritis, JIA is characterized by inflammation of the synovium of one or more joints, causing persistent swelling, reduced mobility, and pain when moving [9]. The temporomandibular joint (TMJ) can also be involved in all JIA subtypes, with a reported prevalence between 17% and 87%, depending on the diagnostic criteria and methodology [11,12]. TMJ involvement may occur at the onset or during the course of JIA [13,14]. TMJ involvement develops during the active phase of JIA, when inflammation generates chondral and subchondral bone lesions, eventually leading to condylar resorption [15]. Orofacial consequences range from clinically asymptomatic conditions to dysfunctions and/or morphological deformities [16–18], with uni- or bilateral impairment. Untreated TMJ involvement in children with JIA can lead to myalgia of head and neck muscles [19] and the disturbance of mandibular growth (mandibular micrognathia and retrognathia) [20,21]. It has been reported in the literature that altered growth may also cause malocclusion and jaw functional deficits such as jaw asymmetry, limited maximal incisal opening, increased profile convexity, and significant steepness of the mandibular plane angle [20,21]. Pain mostly occurs during mouth opening and chewing, especially in the TMJ area as well as the masseter muscle region [7]. The importance of early orthognathodontic intervention as well as the role of other clinicians in the framework of a broader and more specific multidisciplinary approach has thus become evident in reducing joint damage sequelae [22]. According to several studies, signs in radiographic images as well as magnetic resonance imaging (MRI) may be very common, despite the absence of clinical symptoms. [15,23,24]. MRI is currently considered the gold standard in the diagnosis of TMJ damage and TMJ arthritis in JIA patients [25]. Despite the low sensitivity and specificity of clinical orofacial examination for the diagnosis of TMJ arthritis in JIA patients, this constitutes an important part of the general clinical assessment, also to evaluate treatment options, assess response to therapy, and provide ongoing monitoring of a patient with existing TMJ arthritis [26]. According to D'Attilio et al. [19], instrumental examination may take place in the evaluation of masticatory muscles' activity and TMJ damage. Surface electromyography (sEMG) is an important tool for both the analysis of muscular performance and physiopathological changes affecting muscles, joints, and related structures [27,28]. The aim of the present study was to analyze the effect of JIA on the activity of masticatory muscles using sEMG and then to compare it with a group of healthy subjects. The null hypothesis is that there is no difference in the activity of masticatory muscles between the two groups.

## 2. Materials and Methods

This study was conducted according to STROBE guidelines for observational studies [29]. The JIA group was composed of 41 subjects (10 males, 31 females; average age 13 years  $\pm$  3) consecutively recruited from September 2020 until June 2021 in the Paediatric Department of the SS. Annunziata Hospital in Chieti, Italy. All subjects fulfilled the following inclusion criteria: (i) subjects with JIA diagnosed according to the ILAR criteria [10] by the Paediatric Department of the SS. Annunziata Hospital in Chieti, Italy; (ii) aged between 5 and 18 years old; (iii) no ongoing or previous orthodontic treatment; (iv) compliance and ability to reliably undergo sEMG. Subjects with the following exclusion criteria were, thus, excluded: (i) incomplete medical history; (ii) congenital craniofacial syndrome diagnosis (e.g., hemifacial microsomia, cleft lip/palate, Treacher Collins syndrome, or TMJ ankylosis); (iii) the presence of craniofacial bone fractures in medical history; (iv) previous

TMJ treatment or craniofacial surgery in medical history; (v) the presence of any other systemic illness. The control group was composed of 32 healthy age- and gender-matched subjects (27 females, 5 males; average age 14 years  $\pm$  2) with no TMD signs or symptoms nor any ongoing or previous orthodontic treatment. Control group data were extracted from the electromyographic database of the Department of Orthodontics of the University of Chieti, Italy. The subjects and their parents were informed about the nature of the investigation, and an informed consent form was signed prior to participation. Subjects could withdraw from this study for any reason and without prejudice at any time. The protocol was reviewed by the Local Ethical Committee of the University of Chieti (protocol no: MGB\_AIG). Moreover, the study was conducted according to the criteria of the Helsinki Declaration and the ICH Guideline for Good Clinical Practice.

### 2.1. Procedure

All subjects underwent an orthodontic and gnathological examination to assess medical and dental history. Gnathological examinations were carried out according to the DC/TMD protocol [30]. Subsequently, sEMG of anterior temporalis (AT), masseter (MM), and sternocleidomastoid (SCM) muscles was performed with the occlusal contact analyzer software called Teethan (BTS S.p.A., Garbagnate Milanese, Milan, Italy), consisting of 6 wireless probes with an acquisition frequency of 1 kHz [31]. The instrument characteristics and probes' positioning were described in a previous investigation by D'Attilio et al. [19]. sEMG acquisition was performed by two blinded expert operators in a quiet room with the subject seated on a comfortable chair without a headrest, with their feet resting on the floor and their arms resting on their lap. The subject was asked to look straight ahead towards a mirror adjusted at eye level to maintain a "natural head position" [32]. As described by D'Attilio et al. [19] and Michelotti et al. [33], three consecutive tests were performed for each subject: the Cotton test (COT6), Clenching test (CLE6), and Rotation test (ROT6). The first two tests allowed measurements of muscle activity in maximum intercuspation: COT6 consisted of biting two cotton rolls positioned on the occlusal surface of the posterior teeth behind the first premolar in order to calibrate maximum muscle activity in the absence of dental contact; CLE6, performed in natural intercuspation, allowed the operator to detect the occlusal parameters. The third test, ROT6, consisted of asking the subjects to rotate their head—first on one side, then on the other—while the operator ensured they did not tilt their head or move their shoulders away from the back of the chair, allowing the SCM measurement. The Teethan software (BTS S.p.A., Garbagnate Milanese, Milan, Italy) then combined all the tests, hence leading to the extraction of the following indices [34]:

- POC: percentage overlapping coefficient (norm values  $83\% \leq X \leq 100\%$ ) [35–37]. The POC indicates the predominance of one of the two sides of each pair of muscles. A POC value of 100% indicates perfect symmetry between the muscles of the two sides; conversely, a value tending to 0% means no symmetry. Three indices were computed for each subject (POC TA for the anterior temporalis muscles; POC MM for the masseter muscles; POC SCM for sternocleidomastoid muscles). POC TA correlates to the anterior teeth contacts, while POC MM to the posterior teeth contacts.
- BAR: percentage overlapping coefficient between posterior and anterior teeth contact (norm values  $90\% \leq X \leq 100\%$ ).
- TORS: torsion index (norm values  $90\% \leq X \leq 100\%$ ) [36–38]. The TORS measures the percentage of torsion of the mandible resulting from the analysis of the torque of the crossed muscle pairs: left TA with right MM and right TA with left MM. The prevalence of one pair of crossed muscles over the other indicates a torsion of the lower jaw. The greater the torquing effect, the more the TORS approaches zero, as no symmetric activation of the couple of muscles exists.
- IMP: impact index (norm values  $85\% \leq X \leq 115\%$ ) [36]. The IMP indicates the muscle workload during clenching. Lower values indicate reduced muscular strength during clenching.

- ASIM: asymmetry index (norm values  $-10\% \leq X \leq +10\%$ ) [39]. The ASIM measures the distribution of the occlusal contacts on the right and left side, comparing the activity of TA and MM of the left side with TA and MM of the right side. A value of zero indicates perfect symmetry of the two couples of muscles; negative values conventionally indicate a predominance of the left couple, while positive values indicate a predominance of the right couple.

## 2.2. Sample Size Calculation and Statistical Analysis

A sample size calculation was performed before recruitment. The primary outcome measure of this study was the POC MM index. Based on previous investigations [34], a sample size of 10 subjects was calculated, in order to obtain a possible statistically significant difference between the observations at the end of the study. The value of  $\alpha$  was determined at 0.05, while the power of the test was determined at 0.90. The website <https://clincalc.com/stats/samplesize.aspx> (accessed on 20 July 2020) was used for the calculation [40].

Statistical analysis was performed using Prism-GraphPad software (Graphpad software, LLC, San Diego, CA, USA). The Kolmogorov–Smirnov normality test was applied for each of the variables to check whether data were normally distributed. For each index, the mean and standard deviation were calculated. Data were only normally distributed for the POC SCM, so a parametric test named the *t* test was applied for this variable. For the other indices with no normal distribution, a non-parametric test—the Mann–Whitney U test—was used to compare the difference between groups. The level of statistical significance was 0.05.

## 3. Results

A total of 41 subjects with JIA and 32 healthy control subjects completed the study.

The mean and standard deviation values of each of the seven variables considered in the study for the JIA and the control group are shown in Table 1.

**Table 1.** sEMG parameters between JIA and control groups and with normal range.

Index	Normal Range	JIA Group (Mean $\pm$ SD)	Control Group (Mean $\pm$ SD)	<i>p</i> -Value
POC TA	$83\% \leq X \leq 100\%$	$80.7 \pm 9.9$	$83.6 \pm 10.2$	0.011 <sup>a,*</sup>
POC MM	$83\% \leq X \leq 100\%$	$81.6 \pm 9.3$	$83.3 \pm 9.27$	0.369 <sup>a</sup>
POC SCM	$83\% \leq X \leq 100\%$	$83.3 \pm 5.5$	$80.4 \pm 7.2$	0.066 <sup>b</sup>
BAR	$90\% \leq X \leq 100\%$	$79.5 \pm 13.8$	$83.2 \pm 9.34$	0.207 <sup>a</sup>
TORS	$90\% \leq X \leq 100\%$	$87.8 \pm 4.8$	$89.4 \pm 3.22$	0.192 <sup>a</sup>
IMP	$85\% \leq X \leq 115\%$	$173.1 \pm 82.9$	$114.4 \pm 75.5$	0.003 <sup>a,*</sup>
ASIM	$-10\% \leq X \leq +10\%$	$2.75 \pm 14.6$	$-2.83 \pm 14.2$	0.225 <sup>a</sup>

Legend: POC TA: percentage overlapping coefficient, anterior temporalis muscles; POC MM: percentage overlapping coefficient, masseter muscles; POC SCM: percentage overlapping coefficient, sternocleidomastoid muscles; BAR: percentage overlapping coefficient between posterior and anterior teeth contact; TORS: torsion index; IMP: impact index; ASIM: asymmetry index; SD: standard deviation; a: Mann–Whitney U test; b: *t*-test; \*: level of significance was set at  $p < 0.05$ .

Four out of seven variables (POC TA, POC MM, IMP, and ASIM) were within the normal range in the control group, while the other three variables (POC SCM, BAR, and TORS) were slightly distant from the normal range.

Five out of seven variables (POC TA, POC MM, BAR, TORS, and IMP) did not fall within the normal range, and only two variables (POC SCM and ASIM) were normal according to the literature in the JIA group.

Comparing the JIA and the control group, the Mann–Whitney U test resulted in statistical significance for only two out of six variables with no normal distribution: POC TA ( $p = 0.011$ ) and IMP ( $p = 0.003$ ). The *t*-test showed no statistically significant difference for the POC SCM between the JIA and control groups ( $p = 0.066$ ).

#### 4. Discussion

The present observational study evaluated the masticatory muscles (MM and TA) and SCM activity in subjects affected by JIA. Data extracted from sEMG in a sample of 41 JIA subjects were compared with those of 32 healthy subjects [41]. Statistically significant differences were obtained for the variables POC TA and IMP ( $p = 0.011$  and  $p = 0.003$ , respectively) between the two groups; thus, the initial null hypothesis could be rejected.

Overall, the control group was shown to be a fairly realistic sample of the normal general population, showing all the variables within the normal range or slightly distant from the normal range [35–39]. Otherwise, the means of the variables extracted from the JIA group was shown to be far or slightly inside the normal range [35–39].

Comparing the two groups, two variables showed statistically significant differences, suggesting an involvement of the mandibular condyle. Regarding the percentage overlapping coefficient (POC) of each muscle pair analyzed in this study, only the POC TA was shown to be statistically significant ( $p = 0.011$ ). Otherwise, the POC values of the MM and SCM muscles were not significant and at the same time within or closer to the normal range. The POC TA suggests a workload imbalance between the anterior temporalis muscles of the two sides [35–37]. Given that only the POC TA showed a significant difference, it could be supposed that the proximity of the muscle insertion with respect to the joint structures could determine the extent of the disease. At the same time, no predominance between the two sides for the masticatory muscle activity was suggested by the lack of significance for the parameters POC MM, POC SCM, and ASIM [35–39]. Therefore, the bilateral involvement of the two articular structures could be supposed [14].

The IMP value evaluates the intensity of the muscular work of the analyzed muscles, proving to be strictly related to the bite strength [36]. In our study, the IMP of the JIA group was shown to be significantly increased compared to the normal control group ( $p = 0.003$ ), thus suggesting a greater clenching tendency. This could be explained by the necessity of the JIA group to bring the affected joint heads closer during the closing movements of the mouth. Not surprisingly, the chronic inflammation of the articular structures leads to progressive condylar erosion [42], increasing the distance between the two joint heads, thus requiring increased masticatory muscle recruitment to keep them closer during mouth movements.

The use of electromyography to investigate somatic muscle activity (especially those of the limbs) in subjects with JIA was largely investigated [43,44], and an Electromyography-Scoring Protocol for the neurophysiologic evaluation and grading of muscle involvement was also proposed [45]. However, few investigations focused their attention on the masticatory muscles. Kreiborg et al. [46] reported a case report of a girl affected by JIA and followed annually from 9 to 17 years of age. The electromyographic observation revealed that relative contraction strength was significantly enhanced, especially in the temporal muscles, confirming our results in POC TA values. Nevertheless, Kreiborg's study [46] was a case report, and different electromyographic equipment was used.

Our indices cannot be compared with other studies, since they had never been used before in subjects affected by JIA. To our knowledge, another observational study [19] performed sEMG of masticatory muscles in children with and without JIA with the same equipment applied in this study, but different indices were extracted. D'Attilio et al. [19] observed an incorrect function of the masticatory muscle system expressed in terms of pathological ratio ( $MM/TA < 1$ ) according to Jankelson [47]. Jankelson's pathological ratio investigated by D'Attilio et al. [19] and the POC TA investigated in the present study equally represent an elaboration of sEMG data recorded by the sensor positioned on the anterior temporalis muscle, indicating the involvement of this muscle in JIA.

One limitation of this study is the lack of differentiation in the JIA group according to the seven categories assessed by the ILAR [9]; future research is needed to evaluate any correlation between the JIA subtypes and the masticatory muscle involvement. Furthermore, it will be of great interest for the future to longitudinally observe the effects of JIA therapies by means of electromyography.

## 5. Conclusions

sEMG highlights changes in muscle activity, and masticatory muscles seem to be slightly affected by JIA. Despite the difficulty in clinically assessing the signs and symptoms of TMJ involvement in JIA, instrumental appliances such as sEMG could represent a non-invasive and more reliable diagnostic method for early detection. In addition, good reproducibility due to a standardized protocol could also be obtained. Further research is thus needed to confirm its validity.

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**Data Availability Statement:** The data presented in this study are available upon request from the corresponding author.

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