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Temporomandibular Joint Dysfunction and Disorders in the Development of the Mandible in Patients with Juvenile Idiopathic Arthritis – Preliminary Study

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
D – writing the article; E – critical revision of the article; F – final approval of article; G – other

Abstract

Background. The temporomandibular joint (TMJ) may be affected unilaterally or bilaterally in the course of juvenile idiopathic arthritis (JIA). Permanent complications involve joint damage or stiffness and disorders in the development of the mandible, such as micrognathia, posterior rotation of the mandible, crowding and protrusion of the front teeth and malocclusion.

Objectives. The aim of the study was the clinical and radiological assessment of TMJ dysfunctions and disorders in the development of the mandible in patients suffering from JIA, depending on the duration of the disease.

Material and Methods. The research involved 46 patients with JIA, recognized according to the criteria established by the International League of Associations for Rheumatology (ILAR). Among the patients, 20 suffered from polyarticular JIA and 26 from pauciarticular JIA. The clinical assessment included determination of the facial profile according to Ricketts, intraoral assessment according to Angle's classification and canine class. There were 15 patients (9 with polyarticular and 6 with pauciarticular JIA) qualified for radiological examination. The location of the mandible was determined with the use of a lateral cephalometric image on the basis of a compilation of various analyses.

Results. Out of the 46 patients, 15 individuals (32.6%) displayed clinical features of TMJ dysfunction (pain, clicking, crepitus). Of these, 6 patients (40.0%) reported disorders in mandibular development typical of JIA in the form of retrognathia and posterior rotation of the mandible. According to the grading system developed by Rohlin and Petersson, articular surface damage was considerably higher in the patients with a longer history of the disease ($p < 0.01$) and positively correlated with the deficit in mandible growth ($r = 0.66$, $p < 0.008$).

Conclusions. Early detection of temporomandibular joint damage, even in the case of juvenile idiopathic arthritis with a low level of inflammation, may prevent permanent and significant facial deformities when combined with orthodontic treatment of disorders in the development of the mandible (*Adv Clin Exp Med* 2014, 23, 5, 797–804).

Key words: juvenile idiopathic arthritis, temporomandibular joint, malocclusion.

Juvenile idiopathic arthritis (JIA) is a systemic connective tissue disease characterized by a self-sustaining immunological inflammatory process which can affect 1 or multiple joints, including temporomandibular joints (TMJ) [1]. Synovial inflammation and hyperplasia form the pathologic basis of inflammatory changes in the TMJ and in peripheral joints. Pannus, which is formed

as a result of chronic inflammation, expands towards the surface of the articular cartilage, leading to its damage or sometimes complete stiffness of the joint. Condylar erosions, flattening or defects, which are formed simultaneously, may be detected radiographically [2]. Chronic inflammation causes permanent complications including micrognathia, posterior rotation of the mandible, anterior open

bite malocclusion or crowding and protrusion of the lower incisors [3–5].

The clinical symptoms of damage to the TMJ in the course of JIA may appear both during exacerbations and remissions of the disease. They usually involve transient pain in the TMJ and acoustic symptoms such as clicking and crepitus during the movement of the joint, which are often unconnected with the rheumatoid process [6, 7].

A dangerous clinical consequence of inflammation of the TMJ is restriction in opening the mouth, leading to difficulty in eating solid food [6, 8].

The aim of this study was the clinical and radiological assessment of TMJ dysfunctions and disorders in the development of the mandible, such as retrognathia and posterior rotation, in patients suffering from juvenile idiopathic arthritis, depending on the clinical form of the disease and its duration.

Material and Methods

The study involved 46 patients (27 girls and 19 boys), aged 6–19, whose JIA had been diagnosed according to the ILAR classification criteria of 2001; 20 of them suffered from polyarticular JIA and 26 from pauciarticular JIA. The patients were under medical supervision at the Pediatric Clinic of the Warsaw Institute of Rheumatology.

The following aspects were evaluated in the examined patients:

- the existence of pain and acoustic symptoms in the area of TMJ during the medical inquiry and clinical examination (during palpation and during maximal mouth opening);

- occlusion according to Angle's class and canine class, determining the relations between the first molars and canines of the jaw and mandible:

- Angle's Class I: the first anterior molar cusp of the superior first molar is located in the anterior molar intercusp sulcus of the inferior first molar;

- Angle's Class II: the first anterior molar cusp of superior first molar is located in the space between the second premolar tooth and the first molar of the mandible;

- Angle's Class III: the first anterior molar cusp of the superior first molar is located in the posterior molar intercusp sulcus of the inferior first molar or in the space between the first and second molars of the mandible;

- Canine Class I: the cusp of the superior canine impinges on the distal plane of the inferior canine;

- Canine Class II: the cusp of the superior canine impinges on the proximal plane of the inferior canine tooth;

- Canine Class III: the cusp of the superior canine tooth impinges behind the inferior canine tooth;

- facial profile according to Ricketts analysis:

- straight = normal profile: the lower lip is located 2–3 mm behind the so-called “esthetic line” *E* from the tip of the nose to the most forward projecting point on the anterior surface of the chin (the pogonion); the lower lip is located farther behind line *E*;

- Convex profile: the lower lip is located over 1 mm in front of line *E*;

- Concave profile: the lower lip is located over 5 mm behind line *E*;

- maximal mouth opening: measurement of the vertical aperture between the incisal edge of the maxillary central incisor and the labio-incisal edge of the opposing mandibular incisor with maximum mandibular abduction.

No previous cases of malocclusion in the family eliminated the possibility of a genetic background of the malocclusion.

From among the examined patients, 15 were qualified for radiological examination and statistical analysis (8 with polyarticular JIA and 7 with pauciarticular JIA).

The location of the mandible was determined with the use of a lateral cephalometric image on the basis of a compilation of various analyses (Fig. 1). All lateral cephalometric images were taken in maximal intercuspitation using an OP 100 orthopantomograph with Kodak T-Mat E film.

The articular surface of the TMJ was described according to the grading system developed by Rohlin and Petersson [9]:

Grade 0 – normal conditions: marked border of the cortical bone of articular surface;

Grade 1 – slight abnormality: single, mild osteophytes, sclerosis and flattening of the articular surface;

Grades 2 and 3 – early and moderate destructive abnormality, such as erosion and cysts;

Grade 4 – severe destructive abnormality: extensive erosions of the lateral and medial parts of the condyle and the temporal joint;

Grade 5 – mutilating abnormality: temporomandibular joint ankylosis.

The assessment of the TMJ surface and periapical changes in the teeth was possible on the basis of pantomographic images taken with an Orthoceph OC 100 using Kodak T-Mat G/RA film.

Ethical Issues

The study protocol was approved by the Bioethics Committee in Białystok, Poland.

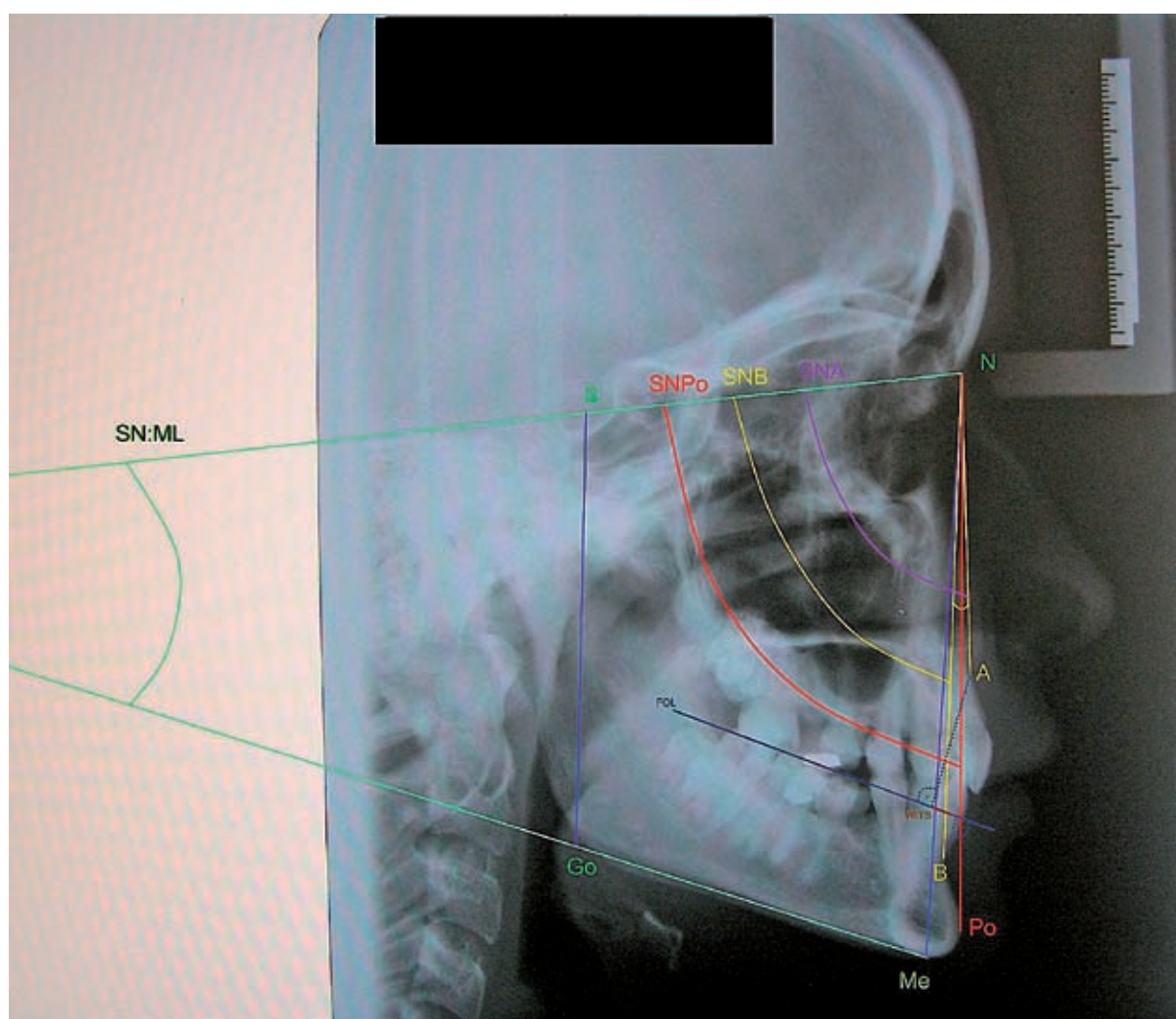


Fig. 1. A compilation of various analyses of a lateral cephalogram of one of the examined patients (S – the sella point; N – the nasion point; A – the subspinale; B – the supramentale; Po – the pogonion point; Me – the menton point; Go – the gonion point; SN – the line of the anterior cranial fossa; ML – the contiguous line to the lower edge of corpus of the mandible; SNA – the angle between the lines joining points S to N and N to A; SNB – the angle between the lines joining points S to N and N to B; SNPo – the angle between the lines joining points S to N and N to Po; ANB – the angle between the lines joining points A to N and N to B; WITS – the measurement defined by the projection of points A and B onto the occlusal plane FOL; ML:SN – the angle between planes ML and SN; SGo:NMe – the proportional relation of the back facial height to the front facial height)

Statistical Analysis

The results obtained were subjected to statistical analysis in which the arithmetic mean and standard deviation were calculated for the measurable features, and the distribution in quantity-percentage terms was calculated for the qualitative features. The Shapiro-Wilk test was used to check for normal distribution of the data. The parametric Student *t*-test or Mann-Whitney *U*-tests were applied as appropriate.

The relationships between selected parameters were determined using the Spearman rank correlation coefficient. $P < 0.05$ was assumed as statistically significant. The statistical power of the tests was also calculated. The calculations were carried out using the Statistica 10 software package (StatSoft).

Results

The age range in the group of patients involved in the study was 6–19 years (mean age 13.8 ± 4.52 years), and the range of duration of the disease was 2–13 years (mean 6.7 ± 4.25 years). The disease activity was determined using the Juvenile Arthritis Disease Activity Score (JADAS) [10]. The JADAS score consists of a global disease activity assessment by a doctor, a global assessment of the child's well-being by the patient/parent, the number of joints involved, and the erythrocyte sedimentation rate (ESR) value. Three grades of disease activity are distinguished: low, medium and high. According to this scale the study population was divided into 2 groups: 35 patients (76%) were classified as having low disease activity, while the

Table 1. Clinical assessment of temporomandibular joints and occlusion in patients with JIA with TMJ dysfunction

| Parameters | | JIA (n = 15) MIZS (n = 15) | (%) |
|------------------------------------|----------|-------------------------------|------|
| Maximum mouth opening (mm) | 15–34 | 4 | 26.7 |
| | 35–38 | 6 | 40.0 |
| | 42–44 | 5 | 33.3 |
| Face profile according to ricketts | straight | 9 | 60.0 |
| | concave | 6 | 40.0 |
| Occlusion – angle's classification | I | 5 | 33.3 |
| | II | 10 | 66.7 |
| Occlusion – canine class | I | 5 | 33.3 |
| | II | 9 | 60.0 |
| | III | 1 | 6.7 |

disease activity in 11 patients (24%) was moderate. Clinical symptoms in the temporomandibular joints mainly included pain, crepitus and restricted mouth opening. The highest value of maximal mouth opening in patients with these symptoms ranged from 44 mm to 35 mm.

Symptoms of joint dysfunction or changes within the facial skeleton were observed in 15 out of the 46 patients, which constituted 32.6% of all the participants of the study; 7 of those 15 (46.7%) had pauciarticular JIA and 8 (53.3%) had polyarticular JIA. Inflammatory disease activity in these 15 patients during the examination was low, according to JADAS. Table 1 shows the frequency of dysfunctions within the facial skeleton observed during clinical examinations. It should be emphasized that only 2 patients with TMJ dysfunction had no changes in the mandibular condyle and articular

Table 2a. Pantomographic analysis in patients with juvenile idiopathic arthritis with TMJ dysfunction

| Parameters | | JIA (n = 15) MIZS (n = 15) | (%) |
|--|-----------------|-------------------------------|------|
| Rohlin and Petersson grading system* | 0° | 2 | 13.3 |
| | I° | 7 | 46.7 |
| | II° and III° | 4 | 26.7 |
| | IV° | 2 | 13.3 |
| Periapical changes in the teeth (pantomographic imaging) | no changes | 3 | 20 |
| | changes present | 12 | 80 |

* degree of articular surface damage to the temporal bone and mandibular condyloid process.

Table 2b. Cephalometric analysis (measurements of cranium and mandible) in patients with JIA with TMJ dysfunction

| Cephalometric measurements: | Mean | SD ± |
|--|-------|------|
| SGo:NMe (norm $58^\circ \pm 3.6^\circ$) | 64.14 | 6.36 |
| ANB (norm $2^\circ \pm 3^\circ$) | 3.0 | 5.42 |
| SN:ML (norm $33^\circ \pm 6^\circ$) | 35.47 | 9.10 |
| SNA (norm $82^\circ \pm 3.5^\circ$) | 80.13 | 4.96 |
| SNB (norm $80^\circ \pm 3.5^\circ$) | 76.30 | 5.70 |
| SNPo (norm $81^\circ \pm 3.5^\circ$) | 76.80 | 6.50 |

surface, and 6 demonstrated significant damage, including 2 patients with very serious changes. In addition, periapical tissues of teeth were evaluated in pantomographic images, and changes were noted in 12 patients (Table 2a). The results presented in Table 2b show a higher tendency toward retrognathia and posterior rotation of the mandible in patients with JIA and TMJ dysfunction in comparison to healthy children. No significant differences were identified between patients with pauciarticular JIA and patients with polyarticular JIA in terms of temporomandibular joint destruction (85.7% to 75%). Dysfunction of these joints, indicated in maximal mouth opening restriction, increases with the duration of the disease (Fig. 2; statistical power = 1.37), as does damage to the temporomandibular bones observed in radiographic imaging (Fig. 3; statistical power = 0.54). A significant positive correlation between the degree of mandibular condylar damage and the suppression of mandible development was observed (Fig. 4; statistical power = 0.81). A significant negative correlation between the ANB angle (retrognathia) and maximal mouth opening was also ascertained (Fig. 5; statistical power = 0.69).

Discussion

The chronic inflammatory process in the course of JIA can affect the temporomandibular joints as well as the surfaces of the bones which form them, including the temporal bones and mandibular condyloid processes. Symptoms of TMJ inflammation in JIA may occur at the beginning of the disease, during its course, or may even precede the disease itself [11, 12]. In the study group, clinical symptoms of TMJ inflammation such as transient pain, clicking and difficulty in opening the mouth were observed in 15 out of 46 patients (over 1/3 of the patients). Among those 15, the maximal mouth opening during the examination was 38 mm, and

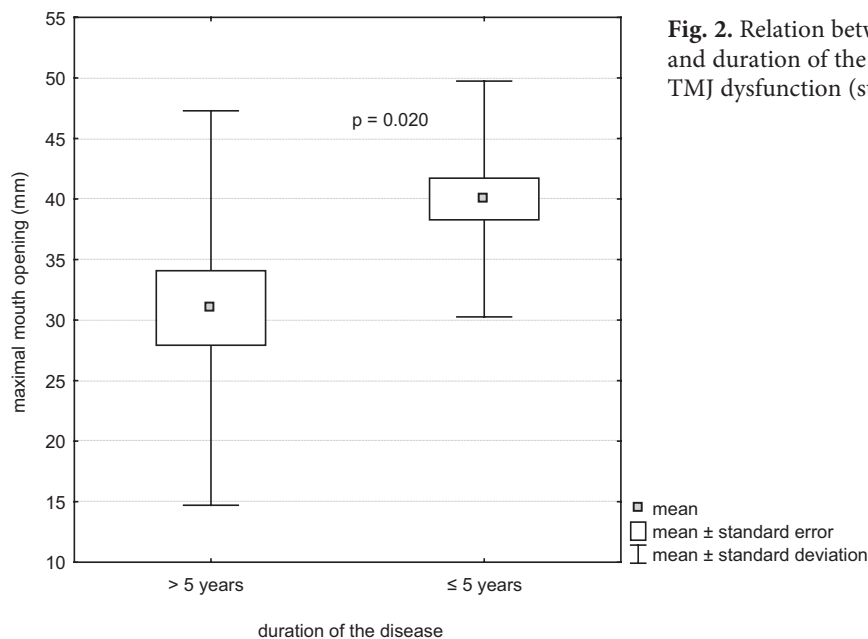


Fig. 2. Relation between maximal mouth opening and duration of the disease in patients with JIA with TMJ dysfunction (statistical power = 1.37)

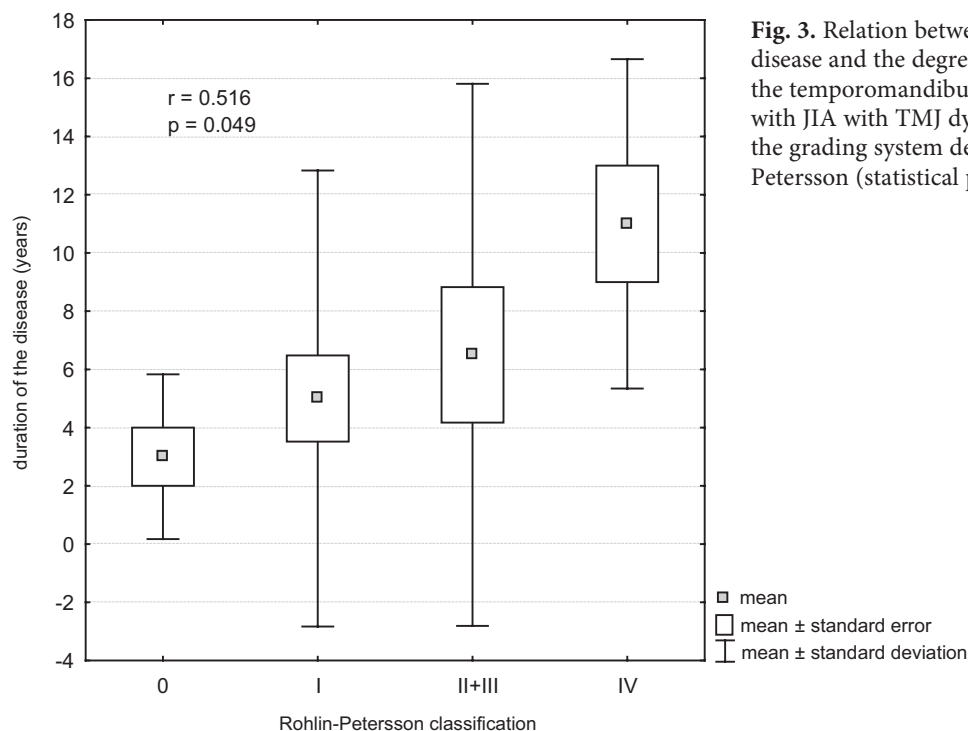


Fig. 3. Relation between the duration of the disease and the degree of surface damage in the temporomandibular joints in patients with JIA with TMJ dysfunction, according to the grading system developed by Rohlin and Petersson (statistical power = 0.54)

the minimal value was 15 mm. The differences depended on the severity of the symptoms and the degree of articular surface damage. Due to the considerable discrepancies in the ages of the examined patients, these values should be compared cautiously with the results of other studies, which indicate a maximal mouth opening of 35 mm [6], 29.2 mm [13] and 49.5 mm [14]. It should be emphasized that persistent pain in TMJ may lead to a weakening of the muscles or even to masseter muscle atrophy, which affects the functioning and development of the mandible [15]. The current study confirms this, because an increased ANB

angle reduced maximal mouth opening (negative correlation – Fig. 5).

Furthermore, in the developmental years the mandible is especially prone to inflammatory factors, partly because the mandibular growth center is located just below the articular surface of the mandibular condyloid process. Therefore, it can be easily damaged, which can lead to mandibular underdevelopment, abnormality in facial skeleton morphology or (ultimately) to “bird face” features [7, 8, 16, 17].

According to the available literature, the frequency of inflammatory changes in the TMJ ranges

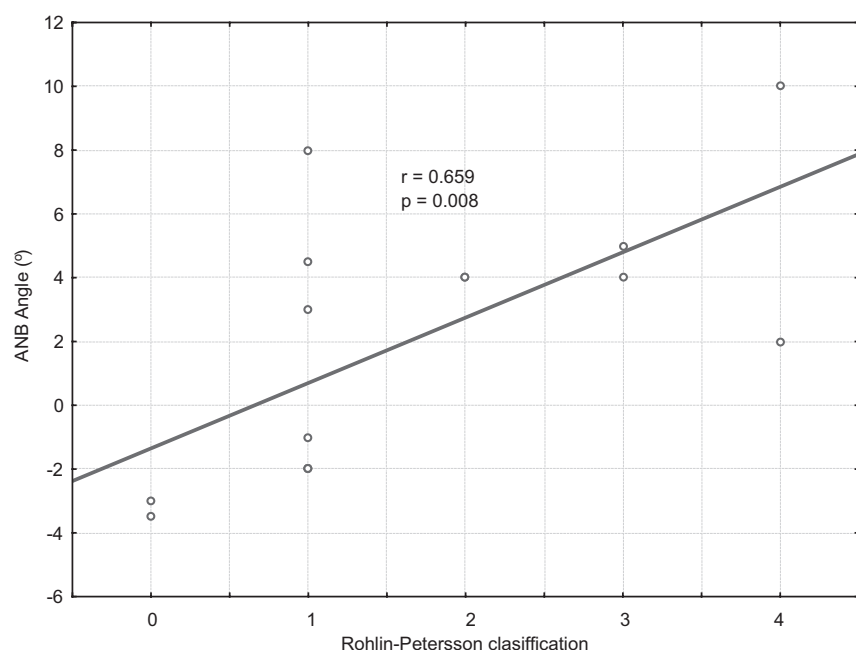


Fig. 4. Relation between the ANB angle and the degree of surface damage to the temporomandibular joints in patients with JIA with TMJ dysfunction, according to grading system developed by Rohlin and Petersson (statistical power = 0.81)

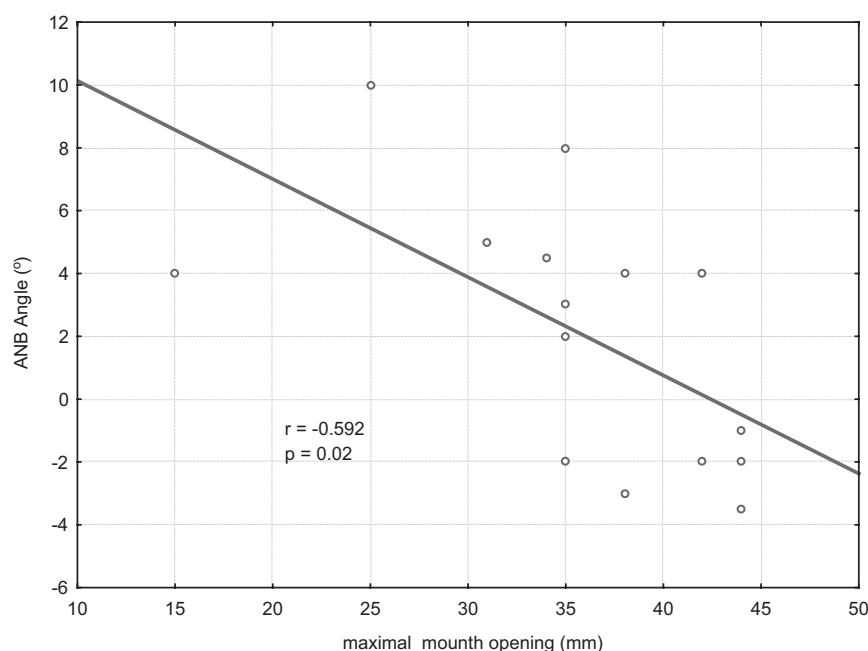


Fig. 5. Relation between the ANB angle and maximal mouth opening in patients with JIA with TMJ dysfunction (statistical power = 0.69)

from 17% to 87%. These differences may result either from the degree of severity of the symptoms and articular surface damage, or from the different populations examined the use of different methods of assessing the affected area of the joint [2, 7, 18–23].

The present study was conducted with the use of equipment allowing for cephalometric and pantomographic imaging. The accuracy of assessment of bone changes with the use of these methods has been evaluated as being from 71% to 84% [24, 25]. Arguably, MRI or CT examination are far more accurate methods [26]. However, they are much more expensive, and in the case of CT, the patient is exposed to a considerably higher dose of

radiation than in the case of cephalometric or pantomographic imaging [27]. Furthermore, the results of a preliminary study by Abramowicz et al. indicate that in children with JIA, the combination of abnormal condyle morphology and accentuated antegonial notching on a panoramic radiograph correlates with TMJ synovitis on an MRI [28].

The findings of this study may be compared to a study by Twilt et al., which also made use of the Rohlin and Petersson grading system [12]. Twilt et al. found the occurrence of changes in the TMJ to be from 37% to 46%. In the present study, clear abnormalities in the cephalometric examination (above Grade 2 according to the Rohlin and Petersson grading system) were observed in 39.9% of the patients.

The findings of the present study confirm the findings of other authors [2, 29] that relate the inflammatory process in TMJ to bigger changes, which may include a retrognathic mandible and impaired maximal mouth opening; the range of maximal mouth opening was significantly lower in patients who had suffered from JIA for over 5 years ($p < 0.02$, Fig. 2). Similarly, damage to the TMJ according to the grading system developed by Rohlin and Petersson was significantly higher in patients with a longer duration of the disease ($p < 0.049$) and positively correlated with an increased ANB angle expressing a retrognathic position of the mandible ($r = 0.66$, $p < 0.008$, Fig. 4).

It is a common conviction that polyarticular JIA may have a greater influence (3 : 1) on damage to the articular surface than pauciarticular JIA [2]; however, according to the current study, the proportions are almost equal (7 : 6).

According to the available literature, bone erosions in JIA occur later than in rheumatoid arthritis, which is a result of the wider articular cartilage, especially in the case of younger children. However, the resulting impairment of the mandible function in the developmental years can be considerable, to such extent that juvenile idiopathic arthritis which affects the TMJ is considered a malicious localization of JIA [30].

To sum up, the use of the presented methods for the early detection of TMJ and mandibular damage may prevent permanent and significant facial deformities that can have a serious impact on the quality of life of young patients, regardless of the disease activity or remission. A lack of standards for the diagnosis and treatment of patients with TMJ arthritis is undoubtedly still a problem [31], but the authors believe that this study will contribute to the development and elaboration of such standards.

The authors concluded that clinical and radiological assessment of the mandible and temporomandibular joints indicated posterior rotation and Class II malocclusion in over 1/3 of the study population of children and teenagers suffering from juvenile idiopathic arthritis. This suggests that it is important to include the clinical and radiological assessment of these joints during each rheumatological control visit. Cephalometric analysis confirms the degree of articular surface damage of the temporomandibular joint in approximately 40% of patients with clinical signs of dysfunction, which indicates the need for early diagnostic imaging. The treatment of patients suffering from juvenile idiopathic arthritis with the symptoms of temporomandibular joint dysfunction should be multidisciplinary, and an orthodontist should be included in the treatment team.

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