

A rare case of unilateral fibrous dysplasia of the condyle of the mandible. Diagnosis and therapy using an axiographic technique and digital X-ray – a case report

Rzadki przypadek jednostronnej włóknistej dysplazji kłykcia żuchwy. Diagnoza i terapia z wykorzystaniem techniki aksjograficznej i cyfrowej techniki RTG – opis przypadku

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SUMMARY

Introduction: The aim of the paper is to present the diagnostic procedure and treatment of a female patient with dysfunction of the right temporomandibular joint, caused by fibrous dysplasia of the head of the right mandibular condyle.

Material and methods: In the diagnostic process of bony structures digital radiography (Digora) was used. To examine the mobility of the mandible axiographic measurements were performed using the Cadiax (Gamma Diagnostic) device.

Conclusions: These methods allowed the confirmation of the diagnosis and the determination of a treatment plan which included the use of a therapeutic appliance. The performed procedures had a positive therapeutic effect.

Key words: fibrous dysplasia, therapy, diagnosis, axiographic visualization, digital X-ray.

STRESZCZENIE

Wstęp: Celem pracy było przedstawienie diagnostyki i terapii pacjentki z dysfunkcją stawu skroniowo-żuchwowego, spowodowane zmianami o charakterze dysplazji włóknistej głowy wyrostka kłykcia żuchwy po stronie prawej.

Materiał i metody: W procesie diagnostycznym struktur kostnych wykorzystano technikę radiografii cyfrowej – system Digora, natomiast ruchomość żuchwy oceniano za pomocą

techniki aksjograficznej. Pomiary przeprowadzono za pomocą urządzenia Cadiax (Diagnostic) Gamma.

Wnioski: Zastosowane metody diagnostyczne mogą potwierdzić diagnozę i ustalić plan leczenia. Przeprowadzone procedury przyniosły pozytywny efekt terapeutyczny.

Słowa kluczowe: dysplazja entodermalna, terapia, diagnostyka, wizualizacja aksjograficzna, cyfrowa radiologia.

INTRODUCTION

Fibrous dysplasia is a rare chronic osseous disease. Its origin is unknown but it is believed to be a developmental, inherent condition [1, 2, 3, 4]. It is usually asymptomatic and concerns long bones, ribs, facial bones and the mandible, where healthy bone is replaced by hyperplastic osseous and fibrous tissues [5, 6, 7]. It usually begins in early childhood and adolescence, and lasts the whole life [3, 8]. Patients usually start to express their complaints and are subject to diagnostic procedure and therapy only in disrupted growth, dysplasia of the affected structures, pathological fractures, and during compression of vascular vessels and nerves by dysplastic pathological fibrous structures [9, 10]. Some patients have an elevated level of acid phosphatase in the blood serum [11, 12, 13, 14].

The aim of this paper was to present the therapeutic procedure applied in the treatment of a patient at the Department of Dental Prosthetics of the Pomeranian Medical University. The patient complained of pain in the area of the right temporomandibular joint.

CASE REPORT

A patient with the initials K.K., clinical card no. 5/K/2008, was referred to the Department of Dental Prosthetics of the Pomeranian Medical University in 2008, in order to diagnose and treat dysfunction of the temporomandibular joint. On the day of admission the patient complained of swelling of the soft tissues on the right side of her face, and acute pain in the



FIGURE 1. A) The patient on the day of admission for treatment; B) The patient at the age of 18; C) The patient at the age of 25

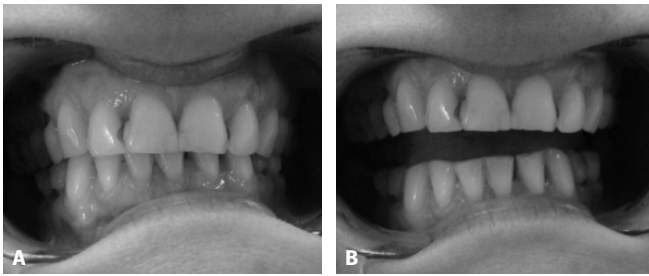


FIGURE 2. A, B) Intraoral photograph before treatment

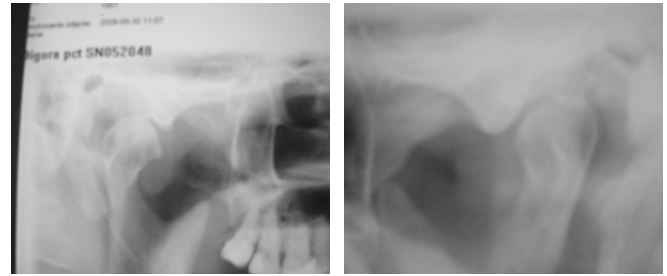


FIGURE 3. Orthopantomogram of the mandible before treatment, changes in the right head of condyle

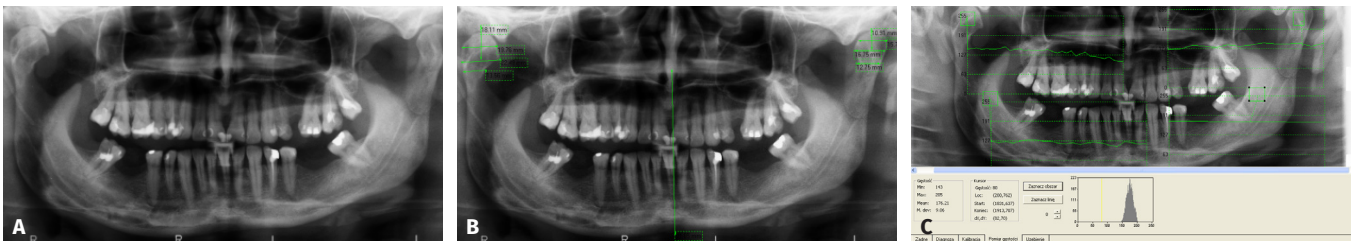


FIGURE 4. A) Orthopantomogram of the mandible on the day of application; B) Orthopantomogram of the mandible with comparative measurements of condyle widths; C) Orthopantomogram of the mandible using the Digora system, with the assessment of the density of selected points, quantified by the measurements of grey level in selected points

area of the right mandibular joint. She had suffered from this pain for many years, albeit with varying intensity. It had been stronger in her childhood and subsided in her adolescence. It recurred a few years before she registered at the Department.

Three months earlier the pain had become so strong that the patient contacted her local primary care physician, who observed asymmetry in facial features and impaired mobility of the mandible. He referred the patient to the Department of Dental Prosthetics at the PMU for further examination (Fig. 1a). Based on the survey and photographs, it was ascertained that the patient had already had asymmetry of the mandible in her childhood (Fig. 1b, 1c). Additionally, the patient reported neurotic states and teeth clenching, associated with personal difficulties, i.e. raising her disabled son for more than ten years. Her emotional style was assessed to be 'task-oriented'. A clinical examination revealed limited mandibular mobility.

During mouth opening, a steady right side deviation was observed. The tension of the masseter muscles was greater on the right side. There was also some asymmetry in facial features, in the area of the ramus and body of the mandible. An intraoral examination showed a distinct difference in

opening (Fig. 2a, 2b). Orthopantomograms of the mandible showed changes in the length of the right ramus compared to the ramus of the left mandible. The right ramus was longer than the left ramus. The head of the right condyle was markedly larger and had very distinct edges (Fig. 3, 4a, 4b).

The orthopantomogram of the mandible (Digora) was used to compare the density of the heads of the right and left condyles and the body of the mandible (Fig. 4c). Significant differences in the grey level between the heads were found; similar values were found in the area between the ramus and body of the mandible on both sides. The median was 176.2, minimum 143, maximum 205. Comparisons were made in the respective sites: right head 94, left head 146, right body of the mandible 196, left body of the mandible 197. Saturation of the heads differed significantly, at $p < 0.001$.

Due to the increased size of the head of the right mandibular condyle, the differences in bone densities, pain and limited mandibular mobility, the patient was sent for observation at the Clinic of the Maxillofacial Surgery of the Pomeranian Medical University. Based on the clinical and laboratory examinations, and computer tomography, dysplasia fibrosa ossium caput

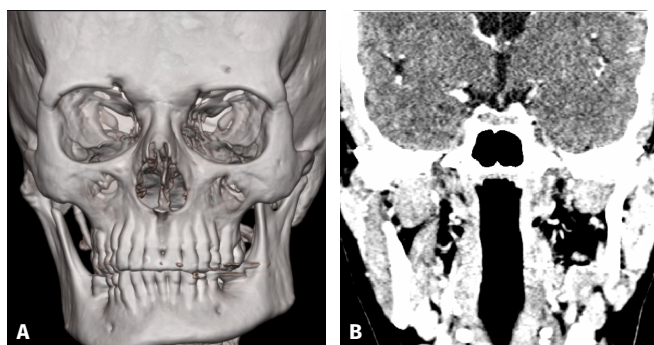


FIGURE 5. A) CT radiographs with a 3D presentation. Changes in the length of the ramus and in the shape of the head of the right mandibular condyle; B) CT radiographs of the mandible – visible changes in the right condyle

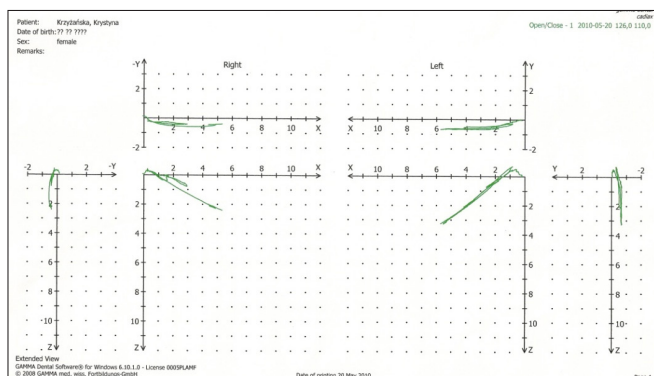


FIGURE 6. Axiography of mandibular movements before therapeutic exercises and the use of a relaxation splint



FIGURE 7. Photograph of the applied relaxation splint in the oral cavity

mandibulae dexter was diagnosed (Fig. 5a, 5b). Surgery was not recommended because pain and swelling had subsided. Instead, further observation and conservative treatment were recommended.

At the Department of Dental Prosthetics of the Pomeranian Medical University, the patient was informed about the nature of the disease and associated risks. Movements of the mandible were recorded with the Cadiax system (Gamma) and presented in a 3D system (Fig. 6). Relaxation exercises for the masseter muscles were recommended. After two weeks, a total occlusal splint was prepared in order to maintain the relaxation of muscles and enhance the mobility of the temporomandibular joint (Fig. 7).

Self-observation and self-checking were recommended. The patient was also asked to pay regular check-up visits. After

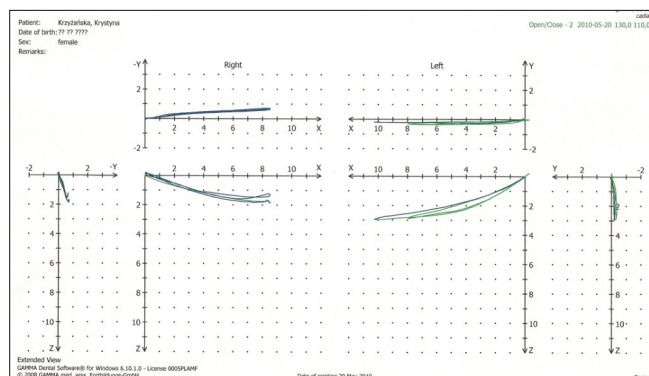


FIGURE 8. Axiography of mandibular movements after 6 months of muscle exercises.

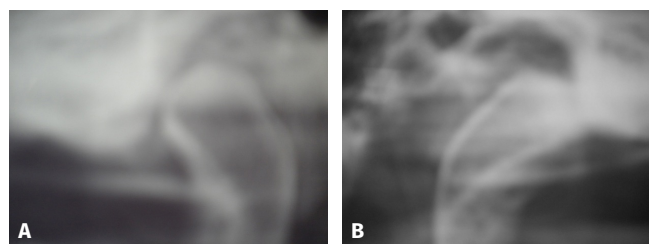


FIGURE 9. A, B) Radiographs of mandibular condyles, 2 years after starting the treatment – mouth open

six months of using the occlusal splint and the recommended exercises for the enhancement of mandibular mobility, another axiographic examination was made (Fig. 8). The pain had subsided and mobility improved. It was recommended for the patient to use the splint in stressful situations (apart from self-checking). After two years, during a check of the morphological state of the mandible, a radiograph was taken (Digora), both with opened and closed mouth. The patient is under continuous radiological assessment.

The mandibular mobility was correct. Mineralization in the area of the right cavity and head of the condyle was found (Figs. 9a, 9b, 10). The patient reported that the pain in the area of the temporomandibular joint had subsided and she could notice an improvement in mandibular mobility (Figs. 11a, 11b and 11c). Correct mobility was confirmed by the axiographic records using the Cadiax system (Fig. 12). Based on conversations with the patient, clinical examination, and additional X-ray photographs, it was observed that the dysfunction of the temporomandibular joint may have been caused by the monofocal form of fibrous dysplasia in the head of the right condyle. The patient is still under the supervision of the Department of Dental Prosthetics of the Pomeranian Medical University.

MATERIAL AND METHODS

X-ray pictures analysis

The presentation of images generated by an X-ray detector device on a monitor screen is based on a matrix of pixels. An image is presented in 8 bit form (although it is created as a 10 bit one), which makes 2^8 , 256 grey levels. Every pixel of an

DISCUSSION

Dysfunction of the temporomandibular joint is one of the most common causes of pain in this area. Pain and changes in facial features, especially asymmetry, should warn physicians and dentists of possible diseases, including fibrous dysplasia [6, 7, 12]. It especially concerns juvenile patients, as suggested by the works of Cohen and *Syryńska et al.* [17, 18, 19]. In adults, facial asymmetry is often ignored by patients and their physician until the onset of pain in the facial part of the skull, accompanied by significant asymmetry of facial features.

In this study, the patient complained of swelling and pain in the area of the temporomandibular joint. On interview, she did not report any traumas in childhood or any later period. Examinations showed asymmetry in the length of the rami of the mandible, and limited mobility in the right joint. Orthopantomograms and Schüller radiographs showed changes in the structure of the head of the right mandibular condyle, and in the length of the right ramus of the mandible.

The most frequent cause of ankylosis is a trauma experienced in the early childhood, inside or outside the joint. The trauma may concern articular surfaces or, additionally, the surrounding tissues [7, 20, 21], resulting in ankylosis, initially fibrous and then osseous, of the temporomandibular joint. If the trauma occurs in the developmental stage, it may be the cause of serious morphological and aesthetic dysfunction, which, if untreated, may lead to acute disability [21, 22]. In adults, ankylosis may occur as a complication of a trauma or arthritis [14].

Fibrous dysplasia of bones is a disease of unknown aetiology, usually inherent and asymptomatic. It is a fibrocystic degeneration of bones, usually occurring in childhood or in menopause, and is more frequent in women. It is inhibited during adolescence, after reaching sexual maturity. In the disease, correct bone is replaced by fibrous connective tissue and decalcified bone tissue³. Fibrous dysplasia presents about 5% of benign tumours of the bone. In 70% of cases it occurs in a form restricted to one bone. It is believed that it has a genetic background [23].

The degeneration may concern one or many bones of the skull, mandible, long bones or ribs. Changes may be located only on one side of the body. The unknown aetiology of the fibrous dysplasia makes it impossible to implement casual treatment [4, 12]. The therapeutic procedure may involve surgical removal of the incorrect tissues, especially in the area of blood vessels or nerves, or to prevent possible fractures [17, 19]. Data on the acuteness of asymmetry can be provided by radiological techniques and computer tomography [12, 24, 25, 26, 27, 28, 29, 30].

Laboratory examinations sometimes show an elevated level of acid phosphatase in patients with fibrous dysplasia. However, it not accepted as a symptom that could confirm the occurrence of the disease [3, 7, 11, 12, 13, 14]. It is therefore recommended to perform careful observation and monitoring of patients, especially children and adolescents [18]. In adults with a significant asymmetry caused by fibrous dysplasia in the facial part of the skull, removal of the affected tissues is usually performed for aesthetic reasons. When surgery is not recommended,

the patient should be monitored as the rapid increase in dysplasia may suggest cancerous metaplasia [11, 24, 26, 31].

In the discussed case, the mild and long-term nature of the disease, and the present clinical state of the patient, indicated a monofocal and mild form of fibrous dysplasia, not intra-articular ankylosis. This was confirmed by a survey, laboratory and clinical studies, additional radiological examinations, and consultation at the Department of Maxillofacial Surgery of the Pomeranian Medical University.

In this case the performed X-ray and axiographic examination were sufficient for diagnosis. Biopsy was therefore unnecessary [32].

CONCLUSIONS

The use of the Digora digital radiography system and analysis of axiographic data collected with the Cadiax device allowed a precise diagnostic and treatment to be performed, along with the following observation. Asymmetric mild fibrous dysplasia does not always require surgical treatment, but always requires continuous monitoring of a patient. The applied therapeutic treatment eliminated pain and restored correct mandibular mobility. Because the patient's bone growth has been completed, an invasive investigation procedure, such as a biopsy, was abandoned. Diagnosis based on radiographic imaging and characteristic clinical symptoms, was confirmed by the good results of conservative treatment.

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