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Ankylosis of the Temporomandibular Joint and Its Surgical Reconstruction – Case Reports. Modern Methods of Treatment

Ankyloza stawu skroniowo-żuchwowego i jego chirurgiczna odbudowa – opis przypadków. Współczesne możliwości leczenia

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A – research concept and design; B – collection and/or assembly of data; C – data analysis and interpretation;
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Abstract

Ankylosis of the temporomandibular joint (TMJ) is a very rare disease that causes dysfunction of the joint. This disorder may develop in the perinatal period or during infancy as the result of improper treatment of an infection of the inner ear which may lead to edema of the infected part of the facial skeleton. This, in turn, may cause hearing disorders and problems with swallowing and breathing. The infection of the inner ear may be viral or bacterial but the injury is usually an iatrogenic effect caused by forceps delivery. The disease may be a result of developmental disorders. Very often the disease is diagnosed too late in spite of many symptoms.

In our study, reconstruction was performed in two children aged 16 and 17. They each suffered from ankylosis of the TMJ. The former case involved surgical reconstruction of the TMJ with a prosthesis and the latter was reconstructed with a distractor. In the former case, an artificial prosthesis was used and in the latter case the TMJ was reconstructed by means of vertical distraction of the left ramus of the mandible. Patients before and after the surgical treatment were treated with braces. In this work, the authors show two successful multistage holistic treatments of ankylosis of the TMJ which prove that even though the treatment is multistage and long-term, surgical intervention may, to a great extent, help and accelerate the healing process (**Dent. Med. Probl.** 2014, 51, 4, 519–530).

Key words: temporomandibular joint disorder therapy, joint prosthesis, ankylosis, distraction.

Streszczenie

Ankyloza stawu skroniowo-żuchwowego jest rzadką chorobą powodującą zaburzenia czynności stawu. Wada ta może powstawać w okresie okołoporodowym lub w wieku niemowlęcym na skutek niewłaściwie leczonego zapalenia ucha wewnętrznego, a w rezultacie może prowadzić do obrzęków chorej części twarzoczaszki. Jej powikłaniami mogą być: uszkodzenie narządu słuchu oraz problemy z przelknięciem i oddychaniem. Prawdopodobieństwo wystąpienia zapalenia ucha wewnętrznego jest skutkiem infekcji bakteryjnej bądź wirusowej. Uraz natomiast ma najczęściej pochodzenie jatrogenne. Wynika on z niewłaściwie przeprowadzonego porodu kleszczowego. Do tego zaburzenia mogą doprowadzić wady rozwojowe płodu. Bardzo często jest późno diagnozowane mimo licznych objawów.

Wykonano rekonstrukcje u 2 dzieci w wieku 16 i 17 lat, u których nastąpiła ankyloza i niedorozwój stawu skroniowo-żuchwowego. Następnie chirurgicznie odbudowano staw skroniowo-żuchwowy za pomocą protezy. W pierwszym przypadku była to proteza sztuczna stawu prawego, w drugim natomiast wykonano rekonstrukcję za pomocą pionowej dystrykcji lewej gałęzi żuchwy. Pacjenci przed zabiegiem i po nim byli leczeni aparatami ortodontycznymi

stałymi. Opisane przypadki pokazują, że jest możliwe skuteczne leczenie zaburzeń czynności s.s.ż. w skrajnie trudnych przypadkach, jakimi są zeszywnienia. Mimo że leczenie jest wieloetapowe i trwa wiele lat, interwencja chirurgiczna w znacznym stopniu ułatwia proces leczenia pacjenta (**Dent. Med. Probl.** 2014, 51, 4, 519–530).

Słowa kluczowe: proteza stawu skroniowo-żuchwowego, ankyloza, dystrakcja.

Ankylosis of the temporomandibular joint is a rare disease of the human skeleton, most frequently caused by anomalies during the prenatal period. It can arrest the growth of the jaw, distort the dental arch – a bird-like profile, and may lead to lockjaw, inappropriate articulation or trouble breathing [1–4]. Joint hypoplasia or hyperplasia are its common signs. Ankylosis is accompanied by such disorders as Franceschetti-Klein, Hallermann-Streiff and Pierre Robin syndromes [1]. Ankylosis may be divided into unilateral or bilateral types. The disorder may be limited to the upper or the lower portion of the joint, or it may take over the whole joint. Histologically, the adhesion is divided into fibrous or fibrous and bone [3, 4]. The bone adhesion of the temporomandibular joint is further divided into: zygopophysis bone adhesion, adhesion of both apophyses and adhesion of the jawbone (division according to Dorski). The disorder may be also divided depending on the place of appearance, for example intra-articular (true ankylosis) or extra-articular (false ankylosis) [3, 4]. It may also be secondary to Still's disease – youthful rheumatoid joint inflammation. The disease, however, covers many joints and leads to fibrosis, distortions, bone adhesion and micrognathia. In the case of ankylosing spondylitis, frequent changes in the jaw structure occur, including joint space narrowing as well as lockjaw [3]. This makes it impossible to keep proper oral hygiene and perform dental exams [3]. Functional disorders should also be considered, because, if permanent, they may lead to degenerative joint changes [1]. They are most often caused by the ageing process. Osteoporosis and deterioration of the support zones in the stomatognathic system are among such functional disorders. This causes degeneration of the fibrous cartilage and articular disk and its perforation in extreme cases [1].

Cases reports

Case 1

A female 16-year-old came to the Children's Center for Craniofacial Anomalies and Oral and Maxillofacial Surgery in Olsztyn for a surgical consult due to the increasing lockjaw and facial disproportions on the right side of the jaw (Fig. 1). The documentation supplied by the parents con-



Fig. 1. Patient pretreatment face photo in the hospital in Olsztyn. Photo: K. Dowgierd

Ryc. 1. Fotografia twarzy pacjentki przed leczeniem w szpitalu w Olsztynie. Fot.: K. Dowgierd

firmed that the patient was treated in the Independent Public Clinical Hospital No. 1 of the Pomeranian Medical University in Szczecin and at the Children's Memorial Health Institute in Warsaw.

In 2000, in Szczecin, the girl was diagnosed with right ankylosis of the temporomandibular joint and right hypoplasia of the ramus and body of the mandible. Additional examination (craniofacial computer tomography) showed a distortion of the coronoid and condylar process of the right ramus and the squama of the temporal bone and the greater wing of the sphenoid bone on the right side. There was residual right temporomandibular joint space, partly preserved in the front. The diameter of the right condylar process was 22 mm. There was also an obstruction of the right maxillary sinus and a complete obstruction of the left nasal duct with deviation of the nasal septum to the right. This would explain the patient's asymmetrical jaw position and malocclusion. The panoramic image from 2000 does not show any bone block between the condylus mandible and the socket (Fig. 2). Distortion of the right socket of the condylar process was also observed. Preventive and orthodontic treatment as well as mechanotherapy and the use of the "TheraBite System[®]" was recommended. The patient was qualified and referred



Fig. 2. Pretreatment panoramic X-ray

Ryc. 2. Zdjęcie pantomograficzne pacjentki przed zabiegiem

to the Children's Surgery Clinic at the Mother and Child Institute in Warsaw to undergo surgery.

In 2008, a surgery to create an artificial joint and mandibular osteotomy with bone grafting from the rib was performed. During this procedure, bone adhesions were surgically removed together with a part of the condylar process at the right side, which impeded joint mobility. Part of the rib was prepared and transplanted in place of a missing condylar process on the right side. The bones were joined by micro platelet anastomosis.

In 2012, the patient was referred to the Craniofacial, Reconstructive and Aesthetic Surgery Department in Olsztyn. During a laryngological consult, limited temporomandibular joint mobility was diagnosed, together with a 1-cm opening of the mouth and facial asymmetry. The nasal ducts were blocked, with planar adhesion in the right duct and a deviation of the nasal septum to the left was found. Due to the limited jaw mobility and the obstruction of the nasal ducts, surgery to treat the ankylosis was postponed until the nasal ducts were unblocked and the adhesion released. This would allow for nasal intubation. The patient consulted several times with a psychiatrist and a psychologist due to her depression, weepiness and emotional imbalance. This was connected with her appearance (the patient could not accept her appearance) and her fear of the upcoming surgery. The patient's mother also needed a psychological consult. A specialist recommended psychological support during and after the surgery.

Due to some difficulties in intubating (1-cm opening of the mouth) and partial nasal duct obstruction, the surgery was postponed twice. Finally, the surgery took place in the middle of 2012.

The patient was intubated through the nose and resection of an ankylotic tumor from the base of the skull was performed under general anesthesia together with a resection of the ramus of the mandible on the right and a removal of a cyst from tooth no. 47 and tooth no. 48 with bone reconstruction. The surgery was performed from an extra-oral approach around the right cheek (preauricular approach) (Figs. 3–5). *Temporal tracheotomy* was also performed.

The patient was told to come to the Craniofacial Clinic in 2 months for a follow-up. Intensive mechanotherapy was recommended through a wide opening of the mouth every day, every hour.

After 2 months, a second stage of surgical treatment was planned, specifically the implan-

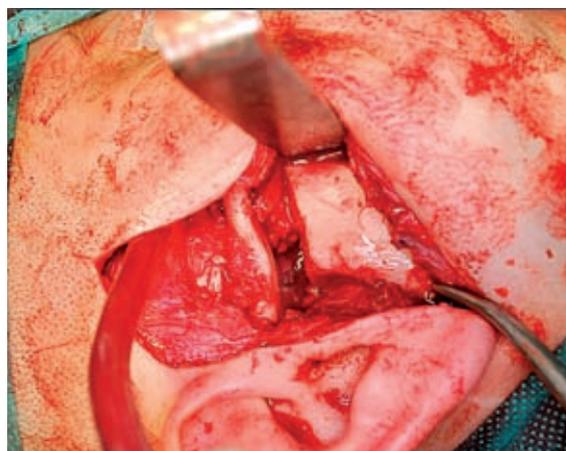


Fig. 3. Preauricular cut. Photo: K. Dowgierd

Ryc. 3. Dojście operacyjne przeduszne.
Fot.: K. Dowgierd



Fig. 4. Excision of ankylotic tumor from right temporomandibular region. Photo: K. Dowgierd

Ryc. 4. Usunięty guz ankiolityczny z okolicy stawu skroniowo-żuchwowego prawego. Fot.: K. Dowgierd

tation of the temporomandibular joint prosthesis. However, due to some difficulties in nasal intubation, the surgery was postponed. After a laryngological consult, a release of intranasal adhesion was recommended before the planned surgery. The patient complained of pain in the right temporomandibular joint (once every 2 weeks). After prolonged chewing, the girl felt pain in the right parotid area and redness of the right cheek – Frey's syndrome. There were some additional difficulties in breathing and snoring at night.

At the end of 2012, osteoplastic surgery of the jaw was performed under general anesthesia together with temporal bone osteoplasty on the right side and right temporomandibular joint reconstruction with the use of Biomet Microfixation® prosthesis (Figs. 6, 7) from the preauricu-



Fig. 5. Posterior-anterior X-ray projection of patient's head after excision of ankylotic tumor

Ryc. 5. Zdjęcie PA głowy pacjentki po usunięciu guza ankiolitycznego

lar approach. The patient was discharged in good condition, with a recommendation of mechanotherapy (exercising the jaws and tongue motor activity) and polarized light therapy with a Biop-

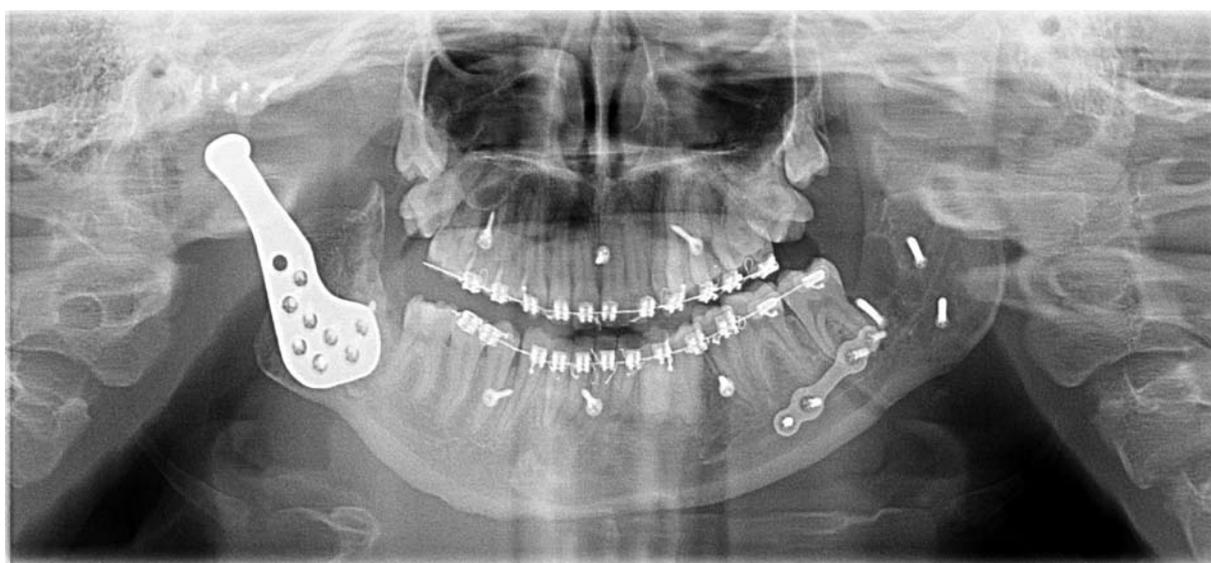


Fig. 6. Panoramic X-ray after implantation of right artificial temporomandibular joint

Ryc. 6. Zdjęcie pantomograficzne po zabiegu wszczepienia stawu skroniowo-żuchwowego prawego

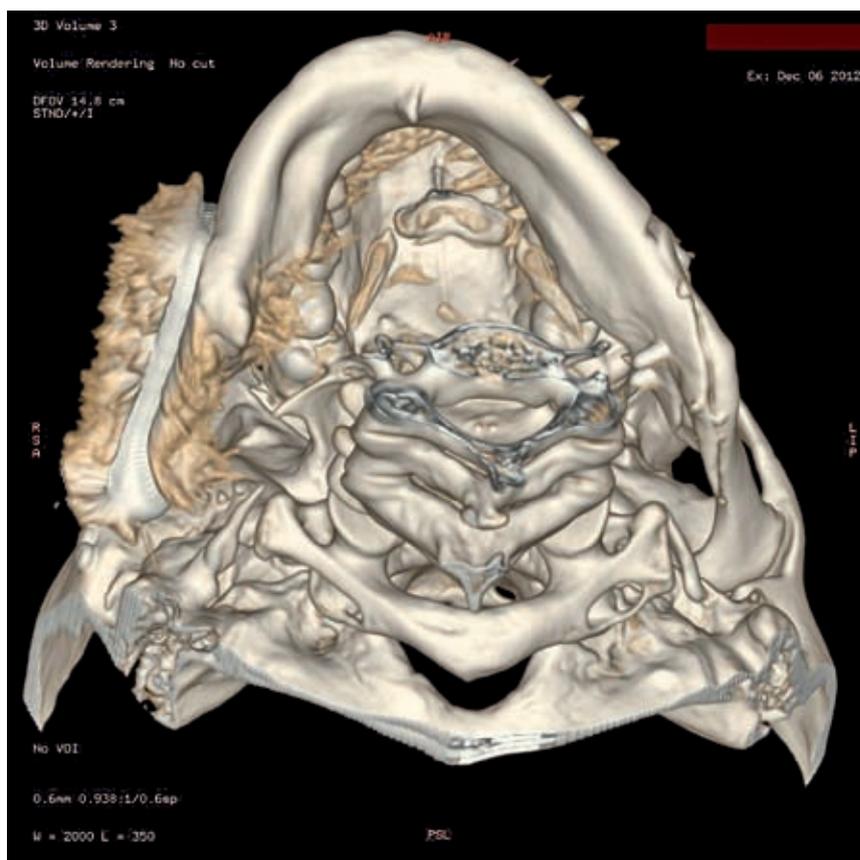


Fig. 7. CT of patient's head after implantation of prosthetic TMJ, bottom view of the mandible

Ryc. 7. TK głowy pacjentki po zabiegu wszczepienia protezy s.s.ż., widok od dołu żuchwy

tron® lamp. Increased oral hygiene was also recommended – rinsing with Eludril® or Octanisept® 4 times a day. A follow-up visit was suggested to take place in two weeks.

The next stage was *genioplastic* corrective surgery after about 6 months of implantation of the temporomandibular joint prosthesis. Its aim was surgical mentum protrusion (Fig. 8). An orthodontist handled the patient's further treatment (Fig. 9).

Case 2

A 17-year-old male patient came to the Children's Center for Craniofacial Anomalies and Oral and Maxillofacial Surgery in Olsztyn for a two-jaw osteoplasty and inserting a vertical distractor of the ramus of the mandible on the left.

When he turned 8 he was treated multidisciplinary by an orthodontist with a fixed and movable apparatus. Jaw abduction was then around 15 mm. A significant asymmetry of the jaw could be seen extra-orally, as well as a mentum and nose shift to the left, and a mentum protrusion outside the biometric field (slant sagittal profile) (Figs. 10, 11).

There were additional acoustic symptoms in the temporomandibular joint on the right and left. The child was then diagnosed with morphological retrusive occlusion (the mother was also diagnosed with disocclusion), fibrous ankylosis of the temporomandibular joint, narrowing of the upper and



Fig. 8. Face photo after the treatment. Photo: K. Dowgierd

Ryc. 8. Zdjęcie twarzy po zakończonym leczeniu.
Fot.: K. Dowgierd

lower dental arch and tooth crowding. In 2010, the orthodontist referred the patient for surgical tooth extraction of no. 38, 48 and 15, under general an-



Fig. 9. Occlusal situation after the surgical and orthodontic treatment

Ryc. 9. Zgryz po zakończonym leczeniu chirurgicznym i ortodontycznym



Fig. 10. Face before the treatment. The picture shows mandibular range of motion approximately 15 mm. Photo: K. Dowgierd

Ryc. 10. Wygląd twarzy przed leczeniem chirurgicznym, widoczny zakres odwodzenia żuchwy rzędu 15 mm. Fot.: K. Dowgierd

esthesia. Due to jaw deformation and small abduction range, there were some complications with intubation. Furthermore, during orthodontic treatment the following teeth were extracted – 24, 34 and 54. Mouth opening and closing exercises (jaw

abduction width was 15 mm) and jaw protrusion were recommended, as well as an apparatus to exercise the temporomandibular joint and a surgical procedure in the future (Fig. 12).

During his stay at the Children's Center for Craniofacial Anomalies and Oral and Maxillofacial Surgery in Olsztyn, a head computed tomography was performed. Shortening of the ramus and body of the mandible on the left was observed. The joint surfaces of the *mandibular fossa* and mandible head were distorted and oblate, and do not create a proper joint surface. The condylar process was distorted and a false joint with the zygomatic arch had formed in the left temporomandibular joint. On the right, the mandible head and a joint lump were slightly oblate. On the side outline of the condylar process a surplus of bone structure was observed. When the mouth was open both sides of the mandible head moved slightly to the front towards the poorly formed lumps (Fig. 13).

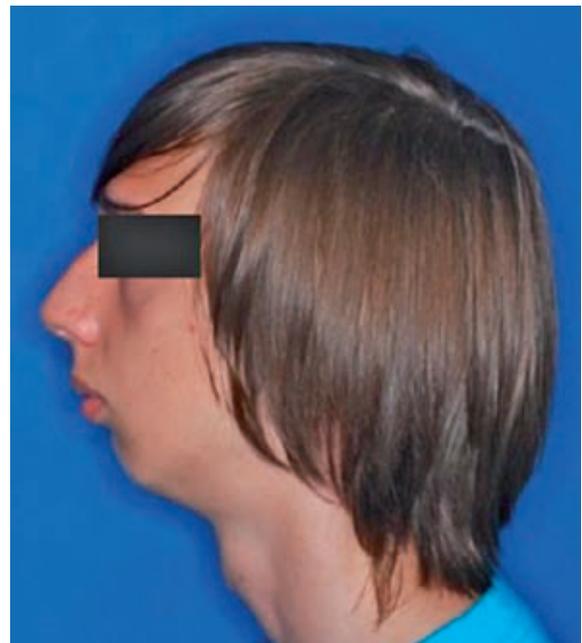


Fig. 11. Patient pretreatment profile photo. Photo: K. Dowgierd

Ryc. 11. Profil twarzy pacjenta przed zastosowaniem leczenia chirurgicznym. Fot.: K. Dowgierd

Furthermore, the patient complained of breathing difficulties, shortness of breath and chewing and speaking disorders (lisp). It is worth mentioning that the patient had had a broken jaw at the age of 4. Pain in the temporomandibular joint area increased during eating and yawning.

During initial examination, physiotherapy and myotherapy, and installing a fixed apparatus, mandibular distraction and vertical left ramus distraction were planned.



Fig. 12. Pretreatment panoramic X-ray

Ryc. 12. Zdjęcie pantomograficzne przed zastosowanym leczeniem



Fig. 13. Pretreatment posterior-anterior X-ray projection

Ryc. 13. Zdjęcie PA głowy pacjenta przed leczeniem



Fig. 14. Excision of the ankylotic tumor from the left temporomandibular joint. Picture shows preauricular cut. Photo: K. Dowgierd

Ryc. 14. Usunięcie guza ankiolitycznego ze s.s.ż. po stronie lewej, zastosowano cięcie przeduszne. Fot.: K. Dowgierd

During the first surgical procedure in September 2012, a two-jaw osteoplasty was performed under general anesthesia, as well as an osteoplasty of the temporal bone with the extraction of the ankylotic block (Fig. 14). Ramus osteoplasty on the left side was also performed together with the extraction of the ankylotic block and a coronoid process. A vertical distractor of the left ramus was installed (Figs. 15–17). The patient was discharged in a general good condition. A follow-up one month af-



Fig. 15. Establishment of a distraction in the left TMJ area. Photo: K. Dowgierd

Ryc. 15. Założenie dystraktora w okolicę s.s.ż. lewego. Fot.: K. Dowgierd



Fig. 16. Assessment of mandibular mobility after the surgical treatment. The picture shows a visible increase in the range of mouth opening to 35 mm. Photo: K. Dowgierd

Ryc. 16. Kontrola ruchomości żuchwy po zabiegu. Widoczne zwiększenie zakresu otwarcia ust do 35 mm. Fot.: K. Dowgierd

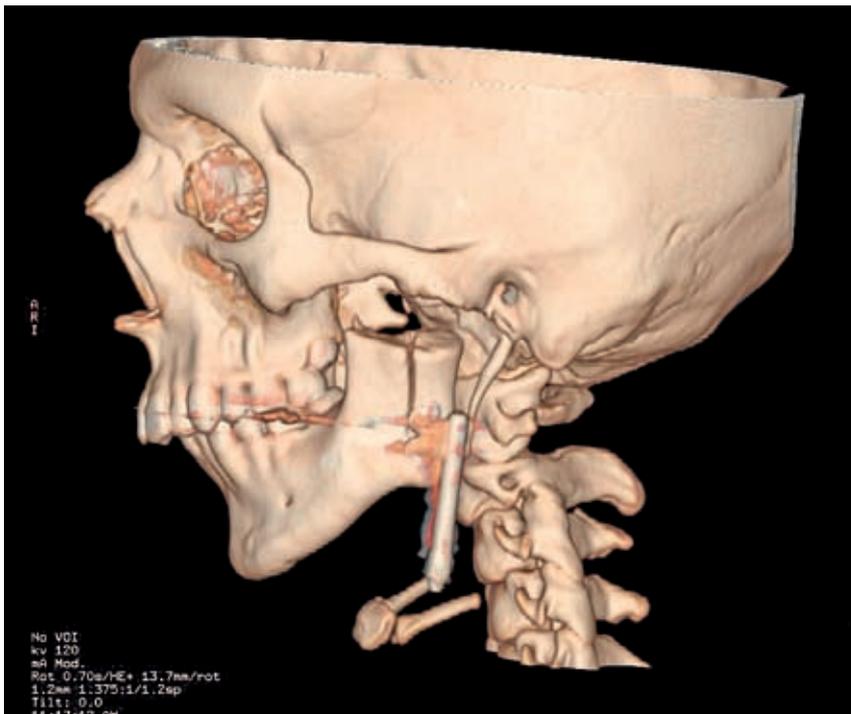


Fig. 17. CT of the head of the patient with the distractor in the temporomandibular region

Ryc. 17. TK głowy pacjenta z uwidocznionym dystraktorem w okolicy s.s.ż.



Fig. 18. Panoramic X-ray after implantation of the distractor in the left temporomandibular region

Ryc. 18. Zdjęcie pantomograficzne po zabiegu wszczępienia dystraktora w okolicę s.s.ż. lewego



Fig. 19. Blockage of the distractor. The picture shows the maximum untwist screw of the distractor

Ryc. 19. Blokada dystraktora, widoczne znaczne rozkręcenie śruby dystraktora

ter the surgery, increased oral hygiene and manual therapy of the jaw opening were recommended, as well as an everyday unscrewing of the distractor by twisting it once 2 times a day, which produced a distraction rate of 1 mm a day. This made it possible to move the jaw down in order to even the facial disproportion.

After a month of using the distractor (Fig. 18), it was blocked for about 2 weeks (Fig. 19). After this period, the patient had to undergo further orthodontic treatment with a fixed apparatus. The

treatment was successful because the patient could open his mouth as wide as 35 mm.

The final stage of his treatment was a two-jaw osteoplasty. It was performed through jaw osteoplasty together with the placement of a palatine distractor and jaw osteotomy together with the placement of a mid-sagittal distractor (August 2013). The patient continues treatment in the orthodontic clinic (Fig. 20).

The aforementioned surgical cases of ankylosis of the temporomandibular joint prove that



Fig. 20. Panoramic X-ray after the final operation

Ryc. 20. Zdjęcie pantomograficzne po ostatecznym zaopatrzeniu chirurgicznym

there is an effective way to decrease the extensiveness of this illness. Further prognosis is promising and may serve as an example of model treatment in cases of such diseases.

Discussion

The issue presented in the article is an example of difficult and complex treatment of temporomandibular joint dysfunction. Ankylosis of the temporomandibular joint is usually the result of certain disorders in the gestation period between weeks 10 and 11 [2]. It may also be secondary to an inflammation of the joint or adjacent organs e.g. inner or middle ear, or a different degenerative disease. The most common cause of such illness is purulent dermatitis and inflammation of the middle ear contracted in childhood, which result in hematogenous mandible inflammation or other joints (e.g. femoral joint) [3]. Another cause of this disease is a perinatal injury caused by e.g. forceps delivery [1, 4]. The disorder may cover one or both joints [2, 4].

The scope of jaw abduction in a young person is 40–52 mm from the edges of the upper and lower incisors. A decrease of this scope may be evidence of temporomandibular joint ankylosis. For instance, in the case of one-sided ankylosis, movement up and down is significantly limited and almost completely moved towards a healthy joint. In the case of two-sided ankylosis, there is a significant limitation in the hinge movement, which eliminates movement to the side and to the front [2].

Image diagnostics were performed based on functional images of the temporomandibular joint according to Schüller, which were taken when the mouth was closed and open, together with computed tomography. Posterior-anterior head images may also help in the diagnosis. A contrasting agent may also be injected in the joint and an X-ray may be taken. Temporomandibular joint arthroscopy may also be performed. However, the most reliable method of soft tissue imaging is magnetic resonance imaging [1, 2, 5].

In previous years, intensive mechanotherapy was the most common method of treatment in the first stages of life, and if such method was ineffective, then surgical treatment was recommended. In many cases, functional treatment gave satisfactory results (e.g. increasing the scope of jaw abduction) [3]. This method of treatment was implemented very early (in the first 1.5 years after the child was born), so the mother's assistance in exercising facial muscles and massaging was necessary. Only later did the child start to participate actively in the functional treatment of ankylosis. Jaw abduction could be then performed with the use of a wooden tongue-depressor. Such activities could be performed by an older child. Different mechanical devices were then used to support the exercises of the temporomandibular joint. Fanti-Krauze[®] was one of the devices prepared in the Oral and Maxillofacial Surgery Clinic at the Medical University of Lodz. There are now other devices available on the market, such as: Translator[®] by Great Lakes Orthodontics, Thera-Pacer 2000[®] by T Prime Technologies Inc., used in the first Thera-bite case presented by Therabite Corporation, and

a pneumatic Fischer dilator [3]. These functional methods were also effective, however the treatment presented in the topic of this paper significantly increases the therapy's effectiveness.

There is another belief which advocates for early surgical intervention. Surgery should be performed between the ages of 8 months and 1 year. This method prevents any secondary disorders in facial and mandibular development (e.g. bird-like profile, disocclusion) and it is less extensive. Włoch and Krajnik (acc. 3) believe that if there is a partial outline of joint gap visible in the X-ray, intensive physiotherapy is recommended in place of surgical treatment. Thus, mechanotherapy is recommended before and after a surgical procedure. A patient should perform active (with the use of their own muscles) and passive moves (with supporting devices) with as little force as possible so as not to injure the soft tissue [3].

Ankylosis is an example of team treatment, where surgical intervention follows an orthodontic one and starts even 2–3 years before a planned surgery but after the body stops growing. Fixed orthodontic appliances are then used. They are also often used after the surgery because they serve as a surgical splint that stabilizes the healing process. Preoperative treatment should even the shape and width of dental arches. Before the surgery, an acrylic splint is often prepared on plaster models in order to recreate the bite after the surgery. This splint helps the surgeon to recreate specific jaw relations during the surgery and is an element that fixes the condylar process after the ramus of the mandible is cut [2].

In the first clinical case, the use of a Biomet Microfixation® joint prosthesis was recommended. It is a procedure selected in the majority of clinics worldwide. This increases the scope of mouth opening and decreases post-surgical pain [6, 7]. This graft contains a molybdenum-chromo-cobalt (Co-Cr-Mo) prosthesis of the condylar process covered with titanium plasma for a better osseointegration and a joint cavity made from polyethylene (UH-MW-PE). The system may be used in three condylar process and joint cavity sizes. A complete set is sterile and may be disposable [8]. Reconstruction of the temporomandibular joint may also be performed through a rib or *fascia lata* graft, or even a coronoid process graft if there are no adhesions [9, 10].

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Partial or bilateral jaw hypoplasia may also be treated with the use of a distractor. It is a screw that is fixed to two cut bones, in this case to a partially cut ramus of the mandible. An incision of the cortical bone is made. The cancellous bone remains intact because of its tendency to stretch. The patients untwist the screw of the distractor by themselves once a day, which causes the bone to extend and the disproportion in the jaw structure to decrease. Usually the screw pitch is 0.5 mm or 1 mm with one twist. This activity should be repeated every day [2].

With surgical intervention, there is risk of complications. Very often, even the incision may cause problems. A preauricular approach may damage facial nerves, while a postauricular incision or incision from the mandibular angle do not clear the surgical field. The complicated structure of the temporomandibular joint causes many problems for the surgeon, thus extra-articular or intra-articular intervention is most common. Undoubtedly, surgical treatment is recommended for patients who have turned 10, when functional treatment is no longer possible. Opinions about ankylosis of the temporomandibular joint vary. Some say that surgical procedures interfere with mandibular growth and should be performed when patients turn 15 or in the third decade – when the stomatognathic system is fully developed [3]. Rehabilitation after the surgery is also controversial. One theory promotes complete rest while the other advocates active rehabilitation with full opening of the mouth. Everyone agrees that it is crucial to start rehabilitation immediately after the surgery [1, 11].

Arthroscopy and low-level laser therapy from the per cutaneous and mid-auricular approach may be the preferred future method of treating this illness, with positive results and no unnecessary trauma to the tissues [1, 12]. There is an interesting temporomandibular joint treatment through intra-articular injections of anti-inflammatory and analgesic agents. Medicines such as Diprophos® (Betamethasonum), Depo-Medrol® (Methylprednisoloni acetate) and Hyalgan® (Natrii hyaluronas) are used in this kind of therapy [13].

The above-mentioned cases show that there is effective treatment for temporomandibular joint dysfunction in extremely difficult cases like ankylosis.

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