Sagittal mandibular osteotomy in a patient with Eisenmenger's syndrome: A case report

Strzałkowa osteotomia żuchwy u pacjenta z zespołem Eisenmengera – opis przypadku

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Abstract

The various abnormalities of occlusion cause significant discomfort to the patient suffering from them. Currently, the surgical treatment of malocclusion in healthy patients is a routine process. The situation is completely different when the patient has a serious disease of the respiratory or cardiovascular system – a condition which may contraindicate such treatment.

A 30-year-old female patient, with a class III skeletal defect (open bite and progeny) and Eisenmenger's syndrome was chosen as a clinical case. The DDS-Pro software was selected to plan the operation. The bilateral sagittal split osteotomy of the mandible was selected as the method of surgery. At the time of the initial examination, the patient had been denied surgical treatment several times in several other clinics. Before the treatment began, the patient underwent intensive preparation in the cardiac surgery unit for 2.5 months. Using the software, a surgical intervention was planned with the production of a surgical template. The operation was then performed; the treatment period was unremarkable. The resulting occlusion and changes in the shape of the face fully met the patient's psychological and esthetic expectations.

In conclusion, a complicated cardiovascular pathology does not always deprive patients with malocclusion of the possibility to undergo surgical treatment.

Key words: orthognathic surgery, malocclusion, progeny, Eisenmenger's syndrome, osteotomy of the mandible

Słowa kluczowe: chirurgia ortognatyczna, wada zgryzu, progenia, zespół Eisenmengera, osteotomia żuchwy

Introduction

The human face has always received special consideration as a social signal which performs both informative and communicative functions. Unfortunately, the human face may be distorted by the presence of various congenital skeletal deformities, such as bite anomalies. The appearance of congenital abnormalities, especially in the maxillofacial area, is caused by different risk factors or genetic disorders during prenatal development.¹ These disorders may be the source of esthetic and psychological discomfort.

However, the presence of concomitant – most often congenital – somatic pathologies in orthognathic patients greatly reduces their chances of receiving any necessary surgical treatment in general, and the correction of facial defects in particular. These are mostly the diseases of the respiratory and cardiovascular systems. The presence of one of these pathologies increases to a great extent the risk of nonsurgical bleeding and complications during general anesthesia.² Moreover, these are high-risk patients for small clinics and they require treatment in large hospitals, equipped with full-sized resuscitation departments. The situation is further complicated by the fact that orthognathic surgery is considered a medical procedure, and not urgent care. It therefore depends on the implementation of the scheduled interventions and may be delayed indefinitely.

Eisenmenger's syndrome is a life-threatening condition, requiring careful medical monitoring. It is a congenital cardiac defect, commonly caused by a shunt between the 2 ventricles of the patient's heart. Eisenmenger's syndrome occurs when the increased pressure of the blood flow in the lung becomes so great that the direction of the blood flow through the shunt reverses. Oxygen-poor blood from the right side of the heart flows into the left side of the heart and is pumped throughout the patient's body, causing insufficient oxygen supply to all organs and tissues.³

Today, thanks to the development of medical science, surgical treatment to correct the facial shape and size has become a routine process. Modern orthognathic surgery enables complex interventions, consisting of diagnostic and planning measures, surgery, and patient rehabilitation. Currently, the standard interventions for bite correction include the Le Fort I osteotomy of the maxilla, the bilateral sagittal osteotomy of the mandible and the compression-distraction techniques.⁴ It should also be noted that osteotomy is less costly for the patient than the compression-distraction techniques, being at the same time equally effective; due to the intraoral surgical approach, it does not distort the patient's face and is less time-consuming than the compression-distraction techniques. The bilateral osteotomy of the mandible has been recognized as an excellent technique for the correction of severe open-bite cases when the patient has

a normally developed mandible and no indication for bi-maxillary osteotomy.^{4,5}

Planning is an important and precise part of such interventions. After this stage, the surgeon knows exactly which bones to remodel and how to proceed in order to achieve the best results.^{6,7}

Taking into account all of the above, this article presents our experience using a multidisciplinary approach to the orthognathic treatment of patients with maxillofacial malformations in combination with a severe concomitant pathology.

Case report

A young woman, aged 30, visited our clinic in October 2017. She had been diagnosed with congenital open bite (a class III skeletal facial deformity, characterized by a significant esthetic and functional defect) as well as Eisenmenger's syndrome with functional class II–III pulmonary hypertension. The patient had been denied surgical treatment several times in other clinics due to the complexity of the deformity and the presence of severe concomitant diseases. A thorough examination by our team revealed that it was possible to perform an orthognathic operation after proper preparation and planning. Written consent was given. The patient was examined by a psychologist and her psychological state was defined as satisfactory. Throughout the preparation phase, she remained under the supervision of a cardiologist and a cardiac surgeon. She underwent oxygen therapy and was treated with sildenafil (Strondex®; Microkhim Pharmaceuticals Ltd., Rubizhne, Ukraine). Warfarin, which she ingested daily, was discontinued in order to increase blood coagulation 3 weeks before surgery and 5 days after. At the beginning of the preparation phase, the thromboplastin sensitivity, measured as the International Normalized Ratio (INR), was 4.1. A re-examination was carried out 2.5 months later. As INR was found to be 1.8, we decided that the patient was ready for surgery (Fig. 1,2).

Fig. 1. Appearance of the patient's face before osteotomy

Fig. 2. The patient's orthopantomogram (OPG) before osteotomy

For virtual intervention planning, we used the DDS-Pro software from Digital Dental Services® (London, UK), which simulates operations with a high degree of accuracy and creates surgical templates that can be printed on a three-dimensional (3D) printer. The choice of the planning software was not random. Our team has successfully used the DDS-Pro software for 3 years.⁸ The 1st step consisted of scanning the cast models of the

patient's upper and lower jaws with a 3D scanner. These files, along with pre-operation spiral computed tomography (CT) with a slice thickness of 0.5 mm, were loaded into the DDS-Pro software, where a 3D skull model was created (Fig. 3). The simulation of bilateral sagittal split osteotomy was performed according to the above-mentioned technique, moving the mandible backward – 7 mm to the right, 4 mm to the left – with a simultaneous left rotation of 5° and 2° upward. The resulting occlusion was verified (Fig. 4). All of the fragments were fixed together and a virtual 3D bite template was created (Fig. 5). A new preoperative 3D skull model was created for the patient and the surgical template was checked virtually (Fig. 6). The procedure was printed out, and the template was tested on the cast models (Fig. 7) and in the patient's oral cavity, on both the maxilla and mandible.

Subsequently, the operation – the bilateral sagittal split osteotomy of the mandible (using the Obwegeser II technique) with the single-stage removal of teeth 38 and 48 – was performed in the cardiac surgery unit under endotracheal anesthesia using sevoflurane gas. After the mandible was incised, the tooth row in each jaw was reduced to the expected occlusion position using

Fig. 3. The patient's initial three-dimensional (3D) skull model (right, top and left views)

Fig. 4. Simulation of the surgical intervention (right, top and left views)

Fig. 5. Surgical template

Fig. 6. Overlaying the surgical template on the 3D model

Fig. 7. Surgical template on preoperative models

the prepared template. In our case, mandibular fixation was performed according to Champy et al., using 2.0-mm miniplates and screws produced within the IRENE® OsteoMed system (Tianjin Zhengtian Medical Instruments Co., Ltd., Tianjin, China).⁹ The IRENE intermaxillary fixation screws with orthodontic chain elastics were used for maxillo-mandibular fixation in the postoperative period.

The postoperative treatment period was unremarkable. The patient was under strict observation in the intensive care unit for the first 10 days following the surgery. On day 20, the patient was discharged and assigned post-hospital follow-up. The postoperative orthopantomogram (OPG) and photos of the patient's face were taken (Fig. 8,9). The resulting occlusion and changes in the facial shape fully met the patient's psychological and esthetic expectations.

Fig. 8. The patient's postoperative OPG

Fig. 9. Appearance of the patient's face (frontal and profile views) 3 weeks after surgery

The 1st follow-up examination of the results was carried out 4 months after the orthognathic surgery. No deviations or complications were detected (Fig. 10). The final follow-up was conducted in May 2019 (15 months after the surgery). The patient did not notice any changes in bite. She was satisfied with the esthetic and functional outcome (Fig. 11). The clinical examination showed stable fixed occlusion throughout the dental arch. No pathological mobility was observed in the osteotomy area. The subsequent OPG showed healing at the osteotomy sites; miniplates and screws were fixed and stable (Fig. 12). The patient's follow-up continues.

Our study was conducted in accordance with the principles of the Declaration of Helsinki and the relevant laws of Ukraine, and it was approved by the Commission of Ethics of Danylo Halytsky Lviv National Medical University in Ukraine (Protocol No. 6/2019).

Discussion

Before the examination, we conducted a literature search for reports on the combined effects of maxillofacial and cardiovascular pathologies. As expected, there was not much information. We found only a few articles, but none were devoted to orthognathic patients with Eisenmenger's syndrome. Hupp presented a report on the successful treatment of a maxillary hemangioma using embolization in a patient with Eisenmenger's syndrome.¹⁰

Fig. 10. The patient's face (frontal and profile views) and bite 4 months after surgery

Fig. 11. The patient's face (frontal and profile views) and bite 15 months after surgery

Fig. 12. The patient's OPG on the 15th-month follow-up visit

The author mentioned that in this particular case, the presence of Eisenmenger's syndrome added an challenge to the administration of anesthesia, but the operation and anesthesia were performed without complications.¹⁰ Raines et al. presented a report on the successful outcomes of various non-cardiac surgeries performed on 12 patients

with Eisenmenger's syndrome using different types of anesthesia.¹¹ The authors emphasize that anesthesia can be safe when different types of anesthesia and medication are applied. In their opinion, the expected mortality rate for patients undergoing non-cardiac surgery may be up to 10%, which is lower than the mortality rate for parturient women after birth or cesarean section.¹¹ Another group of surgeons presented the case of a 38-year-old female who was admitted with cardiac failure and Eisenmenger's syndrome, and underwent total abdominal hysterectomy.¹² She was stabilized preoperatively with digoxin and diuretics. She was successfully managed with general anesthesia, the neuraxial blockade for pain relief, and perioperative bronchodilators and oxygen therapy.¹² This information was good news for our team. After the careful analysis of the abovementioned studies, we reached a consensus on surgical planning and organization.

The issue of which surgical technique to use was easier to resolve: we decided on the standard technique. The advantages of the selected method are described below.

For example, in their study of 2 groups of patients treated with osteotomy and distraction, Van Strijen et al. convincingly proved that distraction osteogenesis required a longer operation time (by 37% on average), but 1 day less of hospital stay than bilateral sagittal split osteotomy.¹³ The surgical cost of distraction osteogenesis was 36% higher than in the case of conventional sagittal split osteotomy.¹³ Baas et al. mentioned that patients experienced more pain after distraction osteogenesis, required more analgesics in the postoperative period and had more infections than the bilateral sagittal split osteotomy group.¹⁴ Moreover, Vos et al. reported a lack of significant differences in mandibular stability a year after treatment between 2 groups of patients where remodeling was performed – with bilateral sagittal osteotomy in one group and using the distraction technique in the other.¹⁵

It is clear that osteotomy requires surgeons to damage jaw integrity, i.e., to create an artificial fracture; it is extremely important to choose the appropriate technique and tool for fixation. This aspect becomes even more important when bilateral osteotomy is performed; 2 fractures require equally reliable fixation. In their study, Collins et al. compared standard 2.0-mm monocortical plates with 2.0-mm locking plates in the treatment of mandible fractures.¹⁶ They concluded that the mandible fractures treated with 2.0-mm locking plates and standard 2.0-mm plates presented similar short-term complication rates. Accordingly, our decision on the choice of the surgical technique was simple and definite.

Summarizing all of the above, we can say that the presence of complicated cardiovascular pathologies should not deprive patients with malocclusion of the possibility of undergoing surgical treatment. A multidisciplinary approach to the preoperative preparation of patients with cardiovascular diseases and dentoalveolar anomalies is effective, and can reduce the number of patients who are refused for orthognathic surgery.

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