Unusual Case of Management of Osteomyelitis after Orthognathic Surgery: Mandibular Reconstruction with TMJ Prosthesis

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Case Report

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Abstract

This case report presents a rare case of osteomyelitis following bimaxillary orthognathic and geioplasty surgeries. The patient, a 41-year-old female, presented with facial pain, difficulty in opening her mouth, and mandibular deviation four months after the initial orthognathic surgery. During the four months, the patient had experienced two post-operative complications that required surgical intervention. Examination revealed mobility between osteotomy segments of the right mandibular body, and Computer Tomographic scan analysis revealed osteomyelitis, and non-union of the right osteotomy side of the mandible from the previous surgery. Due to the advanced state of the disease and significant bone resorption, treatment was planned in two stages, involving bone debridement and then reconstruction with a custom made extended temporomandibular joint prosthesis. The patient had no postoperative complications and achieved satisfactory functional and aesthetic outcomes. This case emphasizes the importance of careful monitoring and prompt management of postoperative complications following orthognathic surgery to prevent rare but serious complications such as osteomyelitis.

BACKGROUND/INTRODUCTION

Orthognathic surgery is a procedure performed to correct discrepancies in the jaws in order to provide a better function for the patients, including improved chewing, speech, and aesthetics. (1–3) Despite being a safe and effective procedure, some complications can occur in the postoperative period, sometimes requiring surgical re-intervention, such as postoperative infections after orthognathic surgery, which are however uncommon. (2,3) In a study out of 250 patients who underwent orthognathic surgery, only 6 (2.4%) presented with postoperative infection. (2) In another study, which analyzed 5,023 cases of orthognathic surgery, 60 cases progressed to postoperative infection. (1)

Osteomyelitis (OM) is a rare infectious disease that may occur after orthognathic surgery. Although rare, it is a serious complication that can lead to significant bone damage and compromise the patient’s quality of life. (3,4) This comorbidity is characterized by an infectious process that invades the bone marrow, leading to devitalization and resorption of the bone, potentially resulting in pathological fractures or sepsis. (5)

To the best of author knowledge, only six cases of osteomyelitis after orthognathic surgery has been reported. Except for two studies, all literature reports have briefly described about the occurrence of osteomyelitis in the patients after orthognathic surgery. (3,4,6,7)

The purpose of this paper is to report a case of a patient who underwent orthognathic surgery and had a history of two re-interventions before being treated by our team. Due to the advanced state of the disease and significant bone resorption with loss of continuity, a personalized mandibular prosthesis was used for reconstruction.

CASE REPORT
A 41-year-old Caucasian female patient with no previous medical conditions or allergies, and a previous history of alcohol and tobacco use 15 years back, presented to our clinic with complaints of facial pain, difficulty in opening her mouth, and mandibular deviation. She had undergone a bimaxillary orthognathic and mentoplasty surgeries 4 months prior to her visit but reported experiencing post-surgery complications which required two additional surgical interventions.

As per patient, first postoperative complication occurred after 1 month of the orthognathic surgery. Patient reported increased volume on the right side of the face, facial erythema, pain, and mandibular deviation to the right during the opening. A Computer tomographic (CT) scan of the face revealed bone resorption on the right side. Surgical debridement of the wound and removal of devitalized bone and fixation material, followed by steel wire fixation of bones (Fig. 1A-B). The patient was kept on antibiotic therapy for 7 days.

In the second postoperative complication which was after 2 months of the first surgical procedure, the patient presented with occlusal deflection to the right, increased volume on the right side of the face, intraoral suppuration, and temporomandibular joint (TMJ) pain. Another surgical debridement was performed, and the fixation material was replaced. The patient was kept on elastics intermaxillary fixation for 1 month and antibiotic therapy for 7 days postoperatively. During the postoperative period, the patient experienced persistent swelling and pain in the TMJ.

She presented to us after one month of the third surgery. Upon examination, the patient presented with the same symptoms as before, as well as a surgical wound dehiscence in the right buccal region with no pus discharge on the posterior right side of the mandible. During the examination, mobility was also detected between osteotomy segments of the right mandibular body.

Upon analysis of the CT scan, we identified several issues including abnormal position of the right mandibular condyle as compared to the left side, extensive osteosclerotic reaction, bone sequestration, poorly defined cortical borders findings suggestive of osteomyelitis, inadequate synthesis material and non-union of the right osteotomy side of the mandible from the previous orthognathic surgery. (Fig. 2A-B)

With provisional diagnosis of OM based on clinical and radiological findings, treatment was planned in two stages i.e. resection and then reconstruction. We proceeded with a right mandibular bone debridement procedure, which involved removing the fixation material, performing a bone fragment biopsy, and analyzing the culture and antibiotic sensitivity of the fragment and plates. The patient remained on intermaxillary fixation for a prolonged period while closely monitored by our oral and maxillofacial team for oral hygiene and care. (Fig. 3)

To ensure adequate nutrition, a nutritionist supervised patient’s intake through a nasogastric tube. The infectologist evaluated the biopsy and culture results after the procedure but could not identify the organisms. Following the recommendation of the infectologist, the patient was started on a 6-month course of oral antibiotic therapy with sulfamethoxazole-trimethoprim. Hyperbaric oxygen therapy (HBOT) was also initiated for 30 sessions.
After the surgical debridement, a large bone defect of approximately 4mm was present, extending from the right mandibular body in the second premolar region to the subcondylar region. Due to the lack of bone in the proximal stump, it was not feasible to perform a reconstruction using reconstruction plates. Therefore, our team opted for a custom-made TMJ prosthesis, which required the removal of the remaining condyle. The patient was closely monitored during this period and remained intermaxillary blocked until the completion of the prosthesis fabrication.

After the availability of custom-made TMJ prosthesis patient underwent surgery for reconstruction. A right submandibular approach was used in conjunction with a pre-auricular approach for the replacement of remnant condyle and fixation of the custom made TMJ prosthesis. A left side mandibular body synthesis was replaced by reconstruction plate and bone grafting was performed in the maxilla to improve the bone volume.

Currently, the patient is undergoing a 1-year postoperative follow-up clinically and radiologically. She has experienced significant improvement in facial aesthetics, with good mouth opening and no joint pain. The recent tomographic examination of the face has revealed good fixation of the osteosynthesis material and proper positioning of the bone segments (Fig. 4). However, the patient still suffers from some esthetic sequelae, such as skeletonization of the right side due to multiple surgical procedures. Resolving these remaining issues will be a significant for the patient and our team.

**DISCUSSION**

Osteomyelitis of the jaws is a severe disease characterized by an inflammatory process in the medullary portion of infectious origin, which can lead to bone devitalization and subsequent resorption, creating structural defects(5,8). The main cause of osteomyelitis is bone trauma, including dental extractions, fractures, and surgeries.

Despite Orthognathic being a planned and safe surgery, there is still a possibility of complications. In the retrospective study, a complication rate of 18.4% was obtained for intraoperative and postoperative complications in orthognathic surgery.(2) In another study, it was found that post-orthognathic surgery infections are uncommon, with an incidence rate of 8%.(7) During our literature review, we came across only 5 cases, reporting the occurrence of osteomyelitis following orthognathic surgery. (3,4,7)

The patient profile mostly affected by this disease is male, caucasians, over 40 years of age. Our patient was also elderly which is similar to literature.(3,7) has also suggested to follow the patients closely after orthognathic surgery with the age of above 35, smoking habits and immuno-compromised. Our patient had a previous history of smoking 15 years prior to orthognathic surgery and was medically fit.

Normally, patients with jaw osteomyelitis might present with pain, swelling, lymphadenopathy, fever, paresthesia involving inferior alveolar nerve, difficulty in mouth opening and fistula. OM might be classified as acute or chronic depending upon the day of presentation since bony infection.(8) In the study of Salman et al(2011) and Nelke et al(2015), patient presented with the infectious symptoms
ranging from swelling, purulent discharge and wound dehiscence after 7–14 days of the orthognathic surgery which were recurring and not responding to the treatment and eventually progressing to chronic OM. In contrast to above studies where patient infectious complication was managed by the same team who performed orthognathic surgery, our patient presented to us after four months of the unsuccessful management of the suspected infectious complication of the orthognathic surgery by the team who performed orthognathic surgery primarily. In line with literature, our patient presented with right side erythema, swelling and intra-oral open wound in the buccal region with no suppuration and bad taste.

Osteomyelitis is usually associated with the mandible, due to its dense cortical structure, thin medulla and decreased vascularity in comparison to maxilla(9). Our patient also developed osteomyelitis of the mandible which is in accordance to reported cases of osteomyelitis after orthognathic surgery.

Diagnosis of osteomyelitis (OM) is based on clinical, microbiological, and imaging findings.(8) CT imaging can be used to diagnose osteomyelitis of the mandible. According to Schulze et al (2015) study, two tomographic signs along with clinical examination and history may help to diagnose osteomyelitis. OM findings in CT scan might consist of osteolytic lesions (irregular hypodensities), poorly defined cortical borders, bone sequestration, associated periosteal reaction, and osteosclerotic changes.(8) In our study, initial diagnosis of chronic OM was based on history, clinical and CT scan presentation. In the computed tomography (CT) scan performed by the patient, signs of extensive osteosclerotic reaction, osteolysis, bone sequestration, and poorly defined cortical borders were present. Similar to Nelke et al (2015), no organism could be identified. In both the studies of Salma et al (2011) and Nelke et al (2015), cases were initially diagnosed on the basis of Cone Bean CT findings of osteomyelitis.

The treatment of mandibular osteomyelitis usually involves debridement of devitalized bone, good soft tissue coverage, and long-term antibiotic use. However, in cases where bone involvement is in an advanced stage, more aggressive procedures should be used, such as bone decortication and resections followed by reconstruction.(10) In the retrospective review of complications after orthognathic surgery, Teltzrow et al (2005) briefly mentioned about the successful management of the patient diagnosed with OM after sagittal osteotomy by decortication and prolonged antibiotic treatment. In the study of Salman et al (2011), initial infectious complication was managed by irrigation, antibiotic therapy, and removal of the fixation material. However, later with the diagnosis of OM, patient was then subjected to surgical debridement of the affected area, extraction of the tooth related to the lesion, and venous and oral antibiotic therapy for a period of 3 months.

Our case was a unique and challenging scenario as compared to the other published reports as our patient initially presented with history of repeated surgeries and an advanced stage of osteomyelitis involving a large region of the mandible. Treatment was divided in two stages consisting of resection followed by reconstruction. Extensive resection of the devitalized bone had to be performed due to extensive spread of OM resulting in a significant mandibular defect. Hence, due to given the extent of the bone loss and the involvement of the subcondylar region, the use of traditional reconstruction plates for bone fixation was not feasible. Patient was kept on intermaxillary fixation and long-term antibiotics as
indicated by the infectious disease specialist. Patient underwent hyperbaric oxygen therapy (HBOT) which as per the review conducted by Rendina et al (2018), helps in the neoformation of blood vessels in supporting bone tissue, in improving the effectiveness of pro-inflammatory cells to combat the infectious process, and nutrition for bone healing. After adequate clinical and radiological follow up with no recurrence of OM, patient underwent reconstruction using a custom made TMJ prosthesis. A custom made TMJ prosthesis has been used previously for mandibular reconstruction in cases where there is significant mandibular structural compromise involving the condylar process.(11) After the procedure, the patient returned to HBOT and antibiotic therapy. Currently, the patient has been followed up for one year after the reconstruction clinically and radiologically with good bone healing and preserved mandibular functions without any recurrence of OM. However, patient will require further cosmetic surgeries for her facial esthetic defect due to the loss of bone support due to numerous surgical procedures performed in the management of OM.

FINAL CONSIDERATIONS

OM is a rare occurrence after OS with only few case reports published in the literature. However, surgeons should be highly vigilant during OS and then while managing post operative infectious complications initially in order to prevent the progression to OM might lead to significant morbidity for the patient.

Abbreviations

1. OM…………………………………………………………………… Osteomyelitis
2. CT…………………………………………………………………..... Computer Tomographic
3. TMJ……………………………………………………………………Temporomandibular Joint
4. HBOT……………………………………………………………….....Hyperbaric oxygen therapy

Declarations

Informed Consent:

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. The patient signed the consent to allow us to publish the case. A copy of the written consent is available for review by the editor of this journal.

Ethical Approval:

Not applicable.

Authors' contributions:

Conception and design of study: LS, JL, ON, TR, MC. Acquisition, analysis or interpretation of data: LS, JL, ON, TR , MC. Drafting of the manuscript: LS, JL, ON, TR, MC. Revising of the manuscript: LS, JL, ON, TR ,
MC. All authors approved the version submitted for publication.

**Availability of data and materials:**

All the data available has been shared within the manuscript.

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**References**


Figures
Figure 1

A: Panoramic radiography of the patient after the first intervention of orthognathic surgery. Fixation of the right side using steel wire, fixation of the left side with 1 plate 2.0 and 2 bicortical screws. B: Axial CT scan showing signs of bone reactive process on the right side.
Figure 2

CT scan performed during the first evaluation by the team. A: 3D reconstruction of the CT scan. White arrow indicating an area of significant bone involvement. B: Sagittal CT scan showing suggestive signs of osteomyelitis.
Figure 3

CT scan performed after surgical debridement procedure, removal of fixation material, and intermaxillary fixation.

Figure 4

CT scan performed with 1-year follow-up after mandibular reconstruction using a total ATM prosthesis on the right side, 2.4 reconstruction plate on the left side, and bone grafts in the maxillary region.