

MANAGEMENT OF MAXILLARY OSTEOMYELITIS IN PATIENTS WITH UNCONTROLLED DIABETES AND RAMSEY HUNT SYNDROME: A RARE CASE

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ABSTRACT

Osteomyelitis is an infectious disease which occurs in jaws and one of the main factors in its aggravation is the impairment of microcirculation. Microangiopathy that develops in long term uncontrolled diabetes patients without predisposing factors is thought to change the course of osteomyelitis in those patients. It is aimed to improve recovery by controlling diabetes in this case report.

The patient with diabetes referred to our clinic with swelling and pain at the right maxillary region. In the clinical examination, the right side of the maxilla was mobile. The biopsy result was osteomyelitis. Treatment plan was the excision of jawbone lesions and subsequent hyperbaric oxygen therapy.

Diabetic control is important to reduce the risk of osteomyelitis in patients with uncontrolled diabetes. In the present case, treatment of osteomyelitis related to uncontrolled diabetes with surgery and hyperbaric oxygen therapy was demonstrated.

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Keywords: Craniofacial Bone, Diabetes, Osteomyelitis, Osteonecrosis

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KONTROLSÜZ DİYABET VE RAMSEY HUNT SENDROMLU HASTADA MAKSİLLER OSTEOMİYELİT TEDAVİSİ: NADİR BİR OLGU

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ÖZ

Osteomyelit kemik iliğinin enfektif hastalığıdır. Osteomyelitin şiddetini arttıran faktörlerden birisi kemik içi mikrosirkülasyonun bozulmasıdır. Uzun süreli kontrolsüz diabeti olan hastalarda gelişen mikroanjiyopatinin osteomyelitin seyrini olumsuz yönde değiştirdiği düşünülmektedir. Bu olgu sunumunda uzun süredir kontrolsüz diabeti olan hastada gelişen maksiller osteomyelitin tedavisi sunulmuştur.

Kliniğimize sağ üst çenede ağrı ve şişlik şikayeti ile başvuran hastanın yapılan ağız içi klinik muayenesinde sağ maksillanın bütün olarak hareketli olduğu görülmüştür. İnsizyonel biopsi sonucu osteomyelit olarak raporlanmıştır. Hastaya kombine antibiyoterapi başlandıktan sonra glisemik regülasyonun sağlanması için endokrinoloji konsültasyonu yapılmıştır. Genel anestezi altında nekrotik alanların eksizyonu ve sonrasında hiperbarik oksijen tedavisi uygulanmıştır.

Kontrolsüz diyabetli hastalarda osteomyelit riskini azaltmak için diyabetik kontrol önemlidir. Bu olguda kontrolsüz diyabet ile ilişkili osteomyelitin klinik özellikleri, cerrahisi ve ek olarak hiperbarik oksijen terapisi ile tedavisi sunulmuştur.

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INTRODUCTION

Osteomyelitis is an infectious bone disease which begins in the medullar cavity, rapidly involving haversian canals with an extension to the periosteum.¹ Osteomyelitis of the facial bones was common in pre-antibiotics era but today it is a rare condition. Discovery of antimicrobial agents, advanced surgical treatment options and adjunctive therapy like hyperbaric oxygen have altered the incidence and prognosis of this disease.² Osteomyelitis occurs more frequently in the mandible than in the maxilla because of the blood supply. Maxilla has an extensive blood supply and thin cortical plates and bone marrow with struts.³ The main etiological factors of osteomyelitis of facial bones are trauma, dental and rhinogenic infections. The systemic factors which compromise the host immunity, such as diabetes, viral infections, HIV, malnutrition, increase the incidence of osteomyelitis. Also it negatively affects the progression and prognosis of the disease.⁴ Microvasculopathy and insufficient inflammatory response can develop in patients with long term uncontrolled diabetes mellitus. Diminished inflammatory response and vascularity can increase the risk of osteomyelitis of the bone because of slower healing rate due to reduced tissue perfusion. Also, it can worsen the course of the disease, causing widespread necrosis of the bones.

A rare, severe complication of the reactivation of Varicella Zoster Virus in the geniculate ganglion of the facial nerve is known as Ramsay Hunt syndrome (RHS). RHS is characterized by otalgia, vesicles in the auditory canal, and ipsilateral facial paralysis.⁵

In this paper we present osteomyelitis of the craniofacial bones in a female patient who has dental infections, poor oral hygiene, long term uncontrolled diabetes and Ramsay Hunt Syndrome.

CASE REPORT

A 61-year-old female patient referred to our clinic had the complaints of pain and swelling in right maxilla on October 2016. There was hypertension, diabetes mellitus and Ramsay Hunt Syndrome in her medical history. Her initial glycosylated hemoglobin A (HbA1c) was approximately 10.5.

Right peripheral facial paralyze was seen in extra oral examination. (Figure 1a) Intraorally there was mobility in the right hemi maxilla. Also, poor oral hygiene was seen (Figure1b). The cone beam computed tomography (CBCT)

scan showed us moth eaten, undemarcated, lytic bone areas in ascending ramous, corpus of right mandible, right temporal, sphenoid bone and maxilla (Figure 2). The incisional biopsy was taken from right maxillary bone, and



Figure 1. a. Right peripheral facial paralysis due to Ramsay Hunt syndrome. b. Erythema in the right maxillary gingival tissue and poor oral hygen.

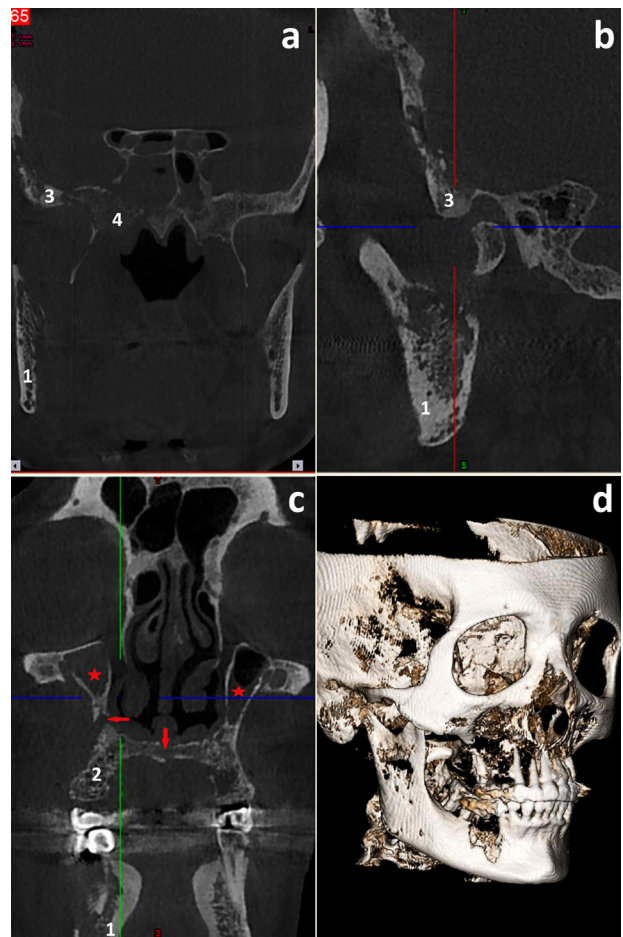


Figure 2. Undemarcated, moth eaten, destructive bone lesion in ramous and corpus of right mandible, right temporal, sphenoid bone and maxilla can be seen in CBCT. Sequester of right maxilla (red arrows) and mucosal thickening in maxillary sinuses (*). (a. Coronal view of mandible, temporal and sphenoid bones; b. Saggital view of mandible and temporal bones; c. Coronal view of maxilla and mandible; d. 3D view)

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the result was compatible with osteomyelitis. Based on the biopsy result and uncontrolled diabetes mellitus, the treatment was planned in three steps. These are improving glycemic control, surgical treatment, and hyperbaric oxygen therapy.

In the first step of the treatment plan, the patient was hospitalized, and combined antibiotic therapy was started. Combined antibiotic therapy (amoxicillin and clavulanic acid+metronidazole) was used during 1 month-period before the surgery. Also, endocrinology consultation was done and the glisemic control regulated. After the regulation of blood sugar level, surgical therapy was done under general anesthesia. Necrotic bone between the right maxillary tuberosity-right upper central incisor and left upper central incisor -left second molar was removed. Upper right first molar, canine, incisor teeth and left incisor, and premolar teeth were extracted. The infected tissues in the maxillary sinuses were removed. Buccal fat pad was used for primary wound closure because of poor quality of gingiva and exposing maxillary sinus bilaterally (Figure 3). Molar teeth in the right mandible were extracted and necrotic bones in the ascending ramus and corpus of mandible were debrided with preserving inferior alveolar nerve. Necrotic bones were sent for pathologic examination and the result was again reported as osteomyelitis. Postoperative antibiotic (amoxicillin and clavulanic acid+ metronidazole) was used for 1 month after the surgery. Conservative treatment with medication and hyperbaric oxygen therapy was chosen to treat osteonecrosis of temporal and sphenoid bones because of limited access and potential complications of surgery. In the last step of the treatment plan, hyperbaric oxygen therapy was started after one week postoperatively. It consisted of 40 sessions and continued for 2 months. The patient was followed up, and uneventful healing was seen 12 months postoperatively. In this follow-up period, the blood sugar value was controlled every three months. There were no signs of recurrence during the 2 years of follow-up in maxilla and mandible (Figure 4a). The patient underwent prosthodontic treatment. Maxillary and mandibular removable partial denture prosthesis was done 12 months postoperatively (Figure 4b). The recovery of lytic bone areas in the ramus and corpus of right mandible, right temporal and sphenoid bone was observed in CBCT scan 6 months postoperatively (Figure 4c). The HbA1c value was approximately 8,5 in post-op follow up period.

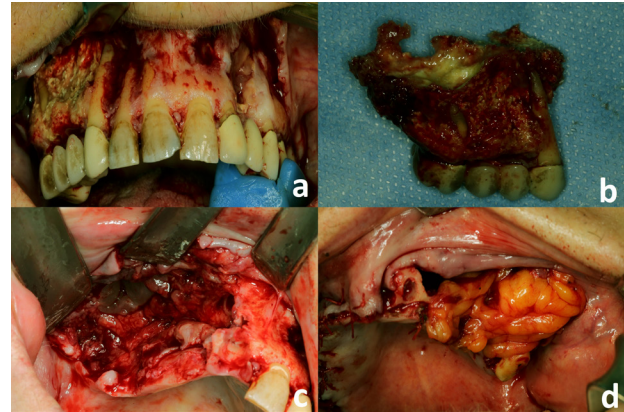


Figure 3. Intraoperative photographs; a. Widespread bilateral necrotic destruction of maxilla. b. Excised sequester of right maxilla. c. Exposed right maxillary sinus. d. Buccal fat pad used to reconstruction and primary wound closure.

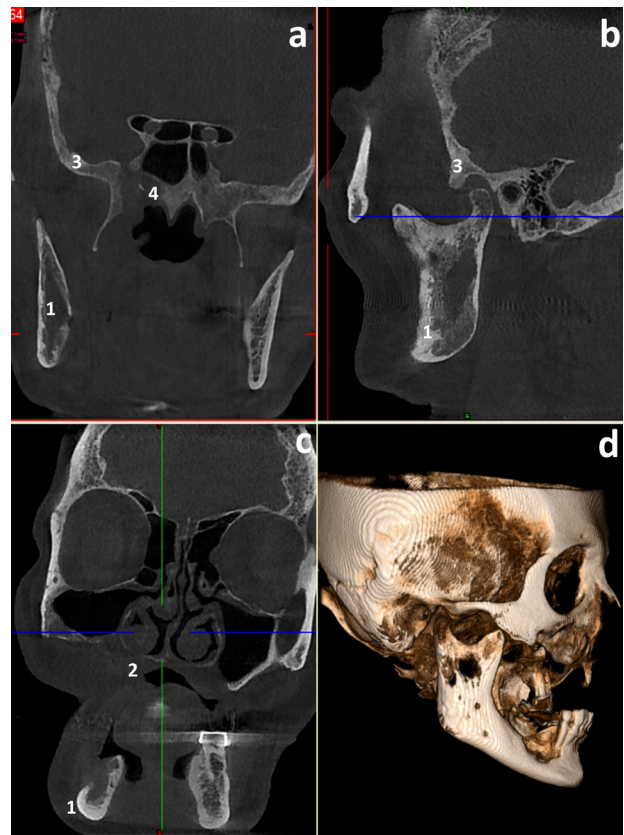


Figure 4. The recovery of destructive bone lesion ramus and corpus of right mandible, right temporal, sphenoid bone and maxilla can be seen in postoperative CBCT. (a. Coronal view of mandible, temporal and sphenoid bones; b. Saggital view of mandible and temporal bones; c. Coronal view of maxilla and mandible; d. 3D view)

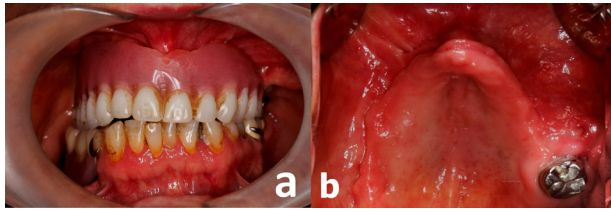


Figure 5. a. Maxillary and mandibular removable partial denture. b. No signs of recurrence at 12th month of the follow-up.

DISCUSSION

Osteomyelitis of the maxilla is a rare entity with the widespread use of antibiotics, early diagnosis, and intervention guided by new imaging modalities.^{1,3} It has been reported extensively in literature, primarily in the form of case reports.^{5,7} It is important to consider the diagnosis in immunocompromised patients as it remains one of the infectious diseases most difficult to treat. In the past, osteomyelitis was encountered frequently and dreaded given its prolonged course, uncertainty of outcome, and possible disfigurement resulting from loss of teeth and bone.⁷ Osteomyelitis occurs more often in mandible than in the maxilla because of the blood supply. Maxilla has extensive blood supply and thin cortical plates, bone marrow with struts.³ Factors predisposing to osteomyelitis of the maxilla include dental infections, maxillary sinusitis, trauma, and radiation. Two main causes are dental infections and sinusitis.⁵ On the other hand, according to Peravali et al.,⁵ a higher incidence of osteomyelitis in maxilla compared to mandible i.e. 16 out of 31 patients (51.7%). 20% of the cases of mandibular osteomyelitis and 68% of the cases of maxillary osteomyelitis were related to uncontrolled diabetes mellitus in their study. Defective glucose utilization disrupts immune system efficiency by altering the blood flow distribution of facial bones.⁵ In this case, the main risk factor is poorly controlled diabetes mellitus and the odontogenic infections.

In the literature, multiple complications including post-herpetic trismus, meningitis, myelitis urinary retention due to reduced bladder tone, and malignant degeneration of basal cell carcinoma of cutaneous lesions have been reported to occur following herpes zoster infection.⁸ A rare complication of Varicella Zoster infection was reported as post-zoster osteonecrosis of the jaw in the literature.^{9, 10} The exact mechanism of osteonecrosis induced by herpes zoster is controversial. Varicella Zoster Infection-induced vasculitis or

inflammation of the local tissue and periosteum is thought to be the possible mechanism of postherpetic osteonecrosis.^{9,11} A case of severe mandibular osteonecrosis and Rumsey Hunt Syndrome was also reported secondary to herpes zoster infection in the literature.¹⁰ In the cases reported in the literature, it was observed that the trigeminal nerve had varicella zoster infection and the underlying systemic conditions. In this case, there was Ramsay Hunt Syndrome with only facial nerve involvement without trigeminal nerve involvement by Varicella Zoster. However, Ramsey Hunt Syndrome may have contributed to the rapid progression of osteomyelitis, causing it to impair systemic and local host resistance.

There are many treatment modalities for osteomyelitis of the jaws ranging from a non-invasive approach to a more invasive radical treatment.¹² Combination of antibiotic treatment with surgery has been demonstrated to be effective in treating the osteomyelitis. Surgical treatment involves removal of loose teeth and sequestrs debridement, decortications, resection, and reconstruction.⁷ Adjuvant hyperbaric oxygen therapy was successful in the treatment of patients with chronic recurrent osteomyelitis of the jaws.¹³ In our case, the patient was treated with a combined antibiotic therapy (amoxicillin and clavulanic acid+ metronidazole), surgery and hyper baric oxygen therapy.

CONCLUSION

Clinicians should carefully evaluate all underlying systemic conditions, particularly in patients with impaired host resistance such as diabetic patient, viral infection. Uncontrolled diabetes, herpes zoster infection, poor oral hygiene should be considered as a risk factor for development osteomyelitis of the jaw. Diabetic control is important to reduce the risk of osteomyelitis development in patients with uncontrolled diabetes. In case of diabetes mellitus related osteonecrosis of the jaw, improvement of glycemic control and combined antibiotic therapy before surgery is essential. Finally, adjuvant hyperbaric oxygen therapy can be beneficial.

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CONFLICT OF INTEREST

The authors declare that there are no conflicts of interest in regard to this work.

DATA PRESENTATION

This article was not submitted to or published in any other journal previously. This case was presented during 11th International Congress of Oral and Maxillofacial Surgery Society (AÇBİD), Antalya, Turkey, 2017.

INFORMED CONSENT

Informed consent from the patient was obtained after detailed information of the necessary treatment. Furthermore, informed consent was obtained also for publication of the procedure and follow-up treatment.

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