CASE REPORT OLGU SUNUMU

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Osteomyelitis Developing in Fracture After Tooth Extraction in the Mandible of Patient with Osteopetrosis

Osteopetrozisli Hastanın Mandibulasında Diş Çekimi Sonrası Oluşan Kırıkta Gelişen Osteomiyelit

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ABSTRACT The case report aims to present a clinical and radiographic feature of osteomyelitis developing in a fracture line after tooth extraction in the mandible of a patient with osteopetrosis. A 49-year-old female patient was admitted to our clinic with complaint a pain and swelling in the right side of the face. It was learned that tooth extraction was performed about 3-year ago, then two fracture lines developed in the relevant region. During medical anamnesis, it was learned that the patient had osteopetrosis. Extraoral examination revealed a wide-diffuse swelling and an extraoral fistula in the right mandibular/submandibular region. Intraoral examination was shown a red and inflamed area. Radiographic examination was showed mostly a well-defined osteolytic lesion involving bone-sequestrum. The patient was consulted to the otolaryngology clinic for treatment and diagnosed with chronic osteomyelitis. Tooth extraction should be avoided in patients with osteopetrosis because of the risk of jaw fracture and osteomyelitis.

Keywords: Osteopetrosis; osteomyelitis; tooth extraction; mandibula; fracture

ÖZET Bu olgu sunumu, osteopetrozisli bir hastanın mandibulasında diş çekimi sonrası oluşan kırık hattında gelişen osteomiyelitin klinik ve radyografik özelliklerini sunmayı amaçlamaktadır. Kırk dokuz yaşındaki kadın hasta yüzün sağ tarafında ağrı ve şişlik şikâyeti ile kliniğimize başvurdu. Yaklaşık 3 yıl önce diş çekimi yapıldığı ardından ilgili bölgede iki kırık hattı geliştiği öğrenildi. Sistemik anamnez sırasında osteopetrozis hastası olduğu öğrenildi. Ekstraoral muayenede sağ mandibular/submandibular bölgede geniş-yaygın şişlik ve ekstraoral fistül saptandı. İntraoral muayenede kırmızı ve iltihaplı bir alan görüldü. Radyografik muayenede kemik sekestrumunu içeren, çoğunlukla iyi sınırlı bir osteolitik lezyon görüldü. Hasta tedavi için kulak burun boğaz kliniğine konsülte edildi ve kronik osteomiyelit tanısı aldı. Osteopetrozisli hastalarda çene kırığı ve osteomiyelit riski nedeniyle diş çekiminden kaçınılmalıdır.

Anahtar Kelimeler: Osteopetrozis; osteomyelit; diş çekimi; mandibula; kırık

Osteopetrosis is a hereditary disease characterized by increased bone density due to a defect in osteoclast function and malformed bone resorption.[1] Osteopetrosis was originally described in 1904 by the German radiologist Albers-Schönberg.[2] Three types of osteopetrosis have been classified as the malign autosomal recessive type, the intermediate autosomal recessive, and the benign autosomal dominant type.[3]

There are crucial clinical and radiological findings in the diagnosis of the osteopetrosis.[1] The clinical findings are commonly skeletal abnormalities such as fragile bone and osteosclerosis and hematologic diseases such as anemia or thrombocytopenia. In addition, vision defect and hearing impairment may occur in the patients. The radiological findings are generally diffuse osteosclerosis, increased cortical bone thickness, and decreased medullary canal diameter.[3] A "marble bone" appearance is usually common in the skeleton, a "bone-in-bone" appearance in the bones of the spine or the hand phalanges, and a "rugger-jersey spine" appearance in the vertebral endplates.[1]

Orofacial examination findings of patients with the osteopetrosis are generally unerupted tooth, malformed crowns and roots, enamel hypoplasia, increased dental caries, and tooth loss.[3,4] Furthermore,

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osteomyelitis of jaw is a severe complication in patients with the osteopetrosis.[3] The osteomyelitis is an inflammation of bone marrow involving the cortical plates and periosteal tissue.[5] Especially, tooth extraction procedures can cause osteomyelitis of the jaws because the procedure can induce a fracture risk, healing force, and irregular alveolar ridges.[3,4]

The case report aims to present the clinical and radiographic features of the osteomyelitis developing in the fracture line after the tooth extraction in the mandible of the patient with osteopetrosis.

CASE REPORT

In January 2022, a 49-year-old female patient was admitted to the Department of Oral and Maxillofacial Radiology, Faculty of Dentistry, Gazi University, with pain complaint, swelling in the right side of the face, and fistula under the mandible. The patient stated that the swelling had started 1 month ago and gradually increased, and a yellowish pus flew into the mouth. Then the fistula formed in the last week, and she was relieved when pus flowed from there. Occasionally, there was a paresthesia in the right lip. It was learned that the first right premolar tooth was extracted in 2019 at a local dental clinic, and two fractures developed in the tooth extraction and angle mandible region in 2020. At that time, the patient reported that she did not go to treatment due to the coronavirus disease-2019 pandemic.

Medical anamnesis revealed that the intermediate autosomal recessive type osteopetrosis was diagnosed when she was at 3-year-old. It was learned that bilateral proximal femur fractured due to a moderate intensity trauma 10-year ago, and right proximal tibia and bilateral ankle fractured spontaneously about 3-year ago. The fractures were treated with prosthesis. The patient was said that her parents were consanguineous marriages, and there were no other family members with osteopetrosis disease.

Extraoral examination revealed a wide-diffuse swelling in the cheek and masseteric region of the right side of the face. The region was indurate and tender with an evident sign of inflammation such as

FIGURE 1: Images the findings of the extraoral and intraoral examination (**A1:** Swelling in the right side of the face, **A2:** Fistula under the mandible, **B:** Partially erupted molar tooth and red, inflame region).

pain, edema, and loss of function in palpation. In the submandibular region, an extraoral fistula with a slightly crusted over, yellowish center, red surrounding, and intermittent pus drainage discharge was observed. Trismus was noted. Intraoral examination revealed a partially erupted molar tooth and a red-inflamed area in the right mandibular molar region related to the extraoral edema (Figure 1).

In 2022, panoramic radiography showed mostly a well-defined osteolytic lesion involving bone sequestrum, association with mandibular canal, and reaching from alveolar process between the second and third molar teeth to the right mandible ramus. It demonstrated diffuse generalized radiopaque image, and osteosclerosis on the maxillary and mandibular alveolar bones, and increased mandibular cortical bone thickness (Figure 2). Cone-beam computed tomography confirmed the bone sequestrum, periosteal reaction, and osteosclerosis (Figure 3). The archives records of the patient's panoramic radiographs taken in 2019 and 2020 were accessed. The radiograph of 2019 showed only an extraction socket of the mandibular right second premolar tooth. The radiograph of 2020 showed two fracture lines inferior of the extraction socket and between the second and third molars (Figure 2).

The patient was preliminarily diagnosed with chronic osteomyelitis according to the clinical and radiographic findings. Due to the wideness of the osteolytic lesion, the patient was consulted to the

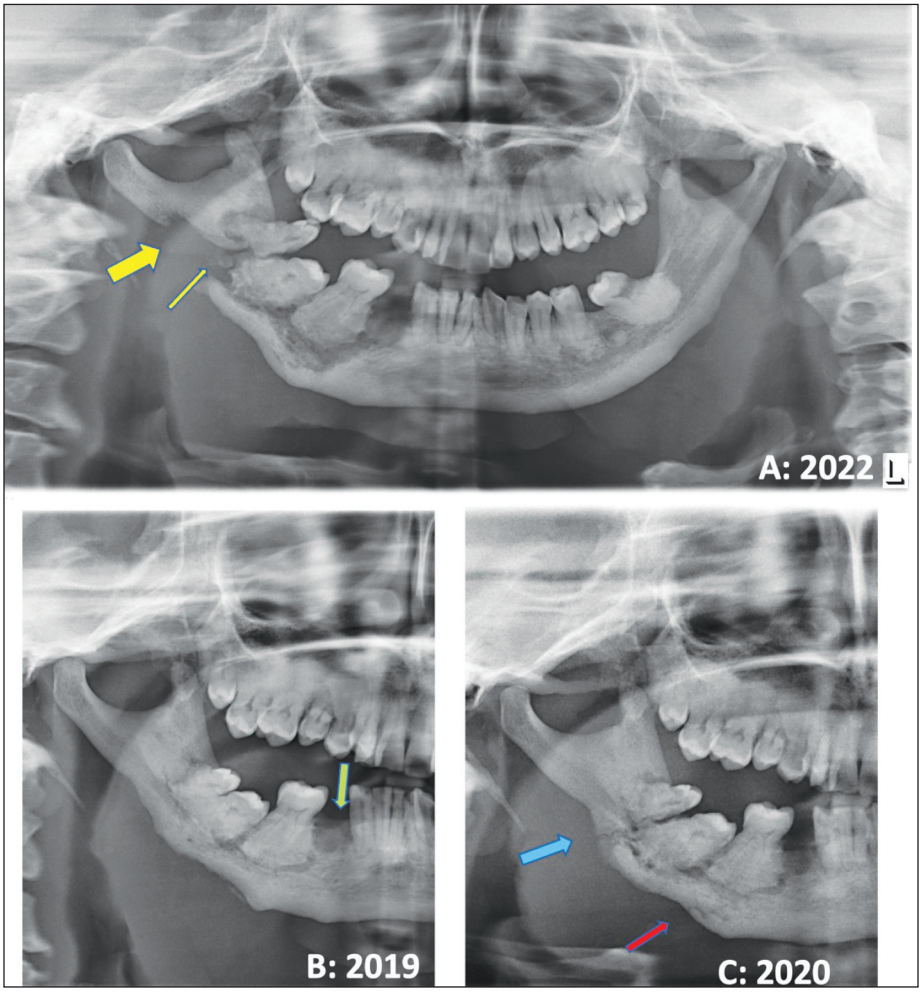

FIGURE 2: Image of panoramic radiographies (**A:** The panoramic radiography in January-2022, **B:** The cropped panoramic radiography in 2019, **C:** The cropped panoramic radiography in 2020, thick yellow arrow: osteolytic lesion, thin yellow arrow: bone sequestrum, green arrow: the socket of the extraction tooth, blue and pink arrows: fracture lines).

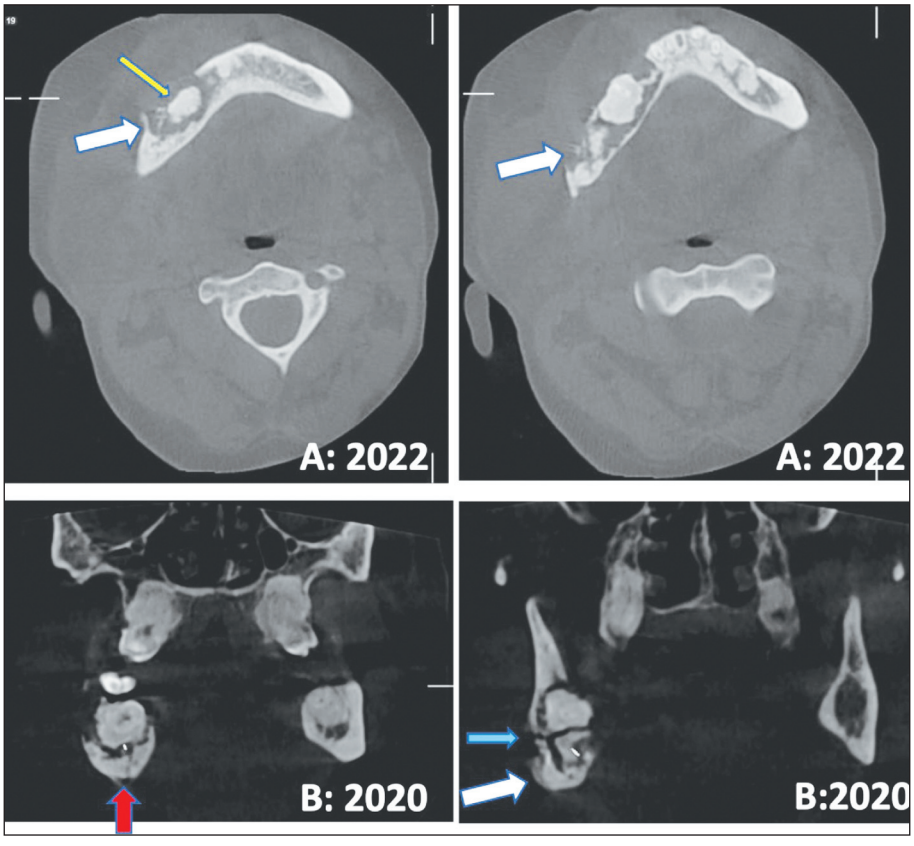

FIGURE 3: Image of cone-beam computed tomography (**A:** Axial sections in January-2022, **B:** Coronal sections in 2020, yellow arrow: bone sequestrum, white arrows: periosteal reaction and osteosclerosis, blue and pink arrows: fracture lines.

otolaryngology clinic for treatment. The patient was diagnosed with chronic osteomyelitis. Extraoral drainage of the abscess from the fistula and antibiotic treatment (14 days-intravenous; ampicillin/sulbactam, piperacillin-tazobactam, then 8-days-oral; ampicillin, sulbactam) were administered in hospital conditions. During the treatment, the third molar tooth in the relevant region spontaneously fell out. The healing was achieved after one month. The patient was followed up with 2-month intervals. The patient is under follow-up, and the treatment of the fracture area will be decided according to the healing process. The patient was informed. An informed consent form was signed by the patient.

DISCUSSION

Osteomyelitis is a serious complication associated with osteopetrosis due to its hypo-vascular nature of the bone.[6]

Bone vascularization is one of the determining factors for the healing process. Especially in the osteopetrosis, the vascularization is poor, and the bones become susceptible to infection due to increased bone density and decreased bone marrow cavities. Accordingly, prevention and control of infection is difficult and serious in the osteopetrosis.[7] Considering the jaws, osteomyelitis generally occurs in the mandible rather than in the maxilla, which has a spongious bone structure and rich vascular supply.[8] Osteomyelitis commonly occurs after the tooth extraction or the bone exposure in the patient.[6] Although osteomyelitis is increased in osteopetrosis, pathological fractures are also a typical manifestation of the disease.[9] These fractures may be the result of poorly organized bone, structural weakness of the bones, and increased bone density. Bones in these patients may have reduced resistance to torsional force. Therefore, long bones such as femur and tibia may

be at high risk for pathological fractures.[10] It has been suggested that pathological fracture may occur in the mandible after tooth extraction.[3] In addition, to the best of our knowledge, a case of pathological fracture of the mandible after tooth extraction is presented for the first time in this case report. In the present patient, osteomyelitis occurred following the bone fracture associated with tooth extraction.

Patients with osteopetrosis can visit the dentist for routine control or any complaints. Especially, good dental care and oral hygiene are crucial. Periodic dental examination, fluoride treatment, and professional dental cleaning are beneficial.[11] A tooth caries should be treated as endodontic or conservative.[12] If the extraction is essential, it should be performed under optimal bone protection measures by a specialist dentist.[11] The patient should be followed up the post-operation due to necrosis development.

Many treatment methods are utilized for jaw osteomyelitis in the patients with osteopetrosis. The methods generally are applied incision and drainage, antibiotic therapy, sequestrectomy, decortication, jaw resection, and hyperbaric oxygen.[13] In the patient, pus from the fistula was flushed and drained, and antibiotic therapy was performed by intravenous and orally. In the literature, the treatment method commonly uses the antibiotic therapy for jaw osteomyelitis in the patients with osteopetrosis.[3,14,15]

In conclusion, the case report presents the clinical and radiographic features of osteomyelitis occurring in the fracture line after the tooth extraction in the mandible of the patient with osteopetrosis. Tooth extraction should be limited in patients with osteopetrosis because the extraction can be inducing the complications such as jaw fracture and osteomyelitis. Supportive of dental care and oral hygiene are very effective and crucial in these patients.

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No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

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