Refractory maxillary osteomyelitis with osteopetrosis: A case report[i]

Abstract

In osteopetrosis, refractory jawbone osteomyelitis often occurs because of the progression of bone sclerosis, which makes the bone brittle. Herein, we report a case of refractory osteopetrosis-related maxillary osteomyelitis for which conservative therapy and surgical intervention were performed along with imaging evaluation.

A 58-year-old woman had maxillary gingivitis for 2 years, for which her tooth was recently extracted. Subsequently, she developed sequestration and oroantral fistula and visited our department. Panoramic radiography, computed tomography, and magnetic resonance imaging (MRI) scans revealed left maxillary osteomyelitis, sinusitis, and sequestration. She had been diagnosed with osteopetrosis in childhood and exhibited sclerotic changes, which were likely caused by osteopetrosis. Bone scintigraphy using Tc-99m-HMDP revealed no increase in tracer uptake in the jawbone. Finally, the patient was diagnosed with osteopetrosis-related left maxillary osteomyelitis and sinusitis; hence, she underwent hyperbaric oxygen therapy, multiple surgeries, and antibiotic therapy. Hepatic dysfunction occurred during the treatment, and a mediastinal tumor was suspected. Thus, fluorodeoxyglucose-positron emission tomography was performed. Further, the patient was diagnosed with extramedullary hematopoietic foci associated with osteopetrosis and hepatic dysfunction due to hepatitis B, there was no Tc-99m accumulation in the maxilla, and hepatitis was treated concurrently. Six months after the final surgery for osteomyelitis, antibiotic therapy was discontinued. Notably, 2 years have passed without exacerbation of osteomyelitis. Moreover, bone scintigraphy using Tc-99m-HMDP did not reveal increased Tc-99m accumulation in the jawbone.

In cases of osteopetrosis-related osteomyelitis, interpretation of findings should be done cautiously because Tc-99m may accumulate in an area with active bone metabolism and osteomyelitis may be masked.

Refractory osteomyelitis with osteopetrosis requires careful interpretation of MRI and bone scintigraphy scans, and long-term follow-up is required.

Keywords

Osteopetrosis, Osteomyelitis, Maxilla, Oroantral fistula, Imaging evaluation

1. Introduction

Osteopetrosis (OP)—first reported by Albers-Schönberg in 1904 [1]—is characterized by impaired bone resorption due to osteoclast dysfunction, which impairs bone maturation and causes progressive osteosclerosis, making the bone brittle. Therefore, patients with OP often experience pathological fractures and osteomyelitis. In particular, OP is common in the jawbone and is often difficult to treat [2]. Some studies have reported the treatment course of patients with OP-induced osteomyelitis, and plain radiography, computed tomography (CT), and magnetic resonance imaging (MRI) have been used to diagnose osteomyelitis. The usefulness of nuclear medicine examinations, such as bone scintigraphy, for evaluating osteomyelitis activity is well known [3-7]. However, to the best of our knowledge, no study has reported the usefulness of these examinations during the treatment of such patients. Therefore, herein, we report the case of a patient with refractory OP-related maxillary osteomyelitis who underwent conservative therapy, including hyperbaric oxygen (HBO) therapy and surgical debridement, and imaging evaluations.

2. Case report

A 58-year-old woman visited our department for the first time in November 2018 with a chief complaint of nonhealing tooth extraction sockets. She had gingival swelling for 2 years for which she underwent drainage of the left maxillary posterior region at a dental clinic and received periodontal treatment. However, her dental symptoms did not improve; therefore, teeth 25 and 27 were extracted 3 months ago. Furthermore, sequestrum was removed at the dental clinic 1 month after tooth extraction. Gingival swelling and purulent drainage temporarily subsided; however, swelling and purulent drainage recurred 1 month later. Further, an oroantral fistula was found during irrigation of the socket, and the patient was referred to our department.

Regarding family history, the patient's father had cholangiocarcinoma. However, the details of her mother were unknown because she died when the patient was a child. The patient had no compatriots.

The patient was diagnosed with OP during elementary education but was not specifically followed up until she suffered a fracture as an adult. Further, her OP was followed up in the orthopedic department. Her medical history included a pathologic fracture in the right femur, plate fusion at the age of 38 years, total joint replacement of the left hip at the age of 56 years, cesarean section, uterine fibroids, hypertension, hepatitis B carrier status, benign paroxysmal positional vertigo.

The performance status of the patient was 1, and she did not have anemia or impaired liver or renal function. Moreover, she had no subjective symptoms, such as postnasal drip or dull headache, but exhibited mild swelling of the left zygoma. Furthermore, there was no sensory or motor impairment in her facial skin. In the oral cavity, a fistula was found in the area of teeth 25 and 27 of her left maxillary jaw, connecting with the maxillary sinus (Fig. 1A). The mucous membrane surrounding the fistula was swollen and red, and when the fistula was irrigated, a small amount of blood mixed with drainage was discharged from the nostril, confirming the opening of the spontaneous orifice.

Panoramic radiography revealed a bone defect on the floor of the left maxillary sinus, and CT scan revealed alveolar osteitis and osteomyelitis in the left maxillary molar region, left maxillary sinusitis, and sequestration (Fig. 1B–F). In addition, remarkable osteosclerotic changes in the facial and cerebral cranium were observed, which were probably caused by OP. Notably, the MRI scan revealed fluid retention in the left maxillary sinus. Meanwhile, the bilateral maxillary and mandibular bone and skull marrow exhibited low intensity on both T1-weighted and T2-weighted images with no enhancement effect, suggesting OP-induced

sclerotic changes. Bone scintigraphy using Tc-99m-HMDP did not reveal increased Tc-99m uptake in the mandible (Fig. 2).

Finally, a clinical diagnosis of left maxillary osteomyelitis and left maxillary sinusitis with OP was made.

Treatment and progress *(Table 1)*

Treatment with 400mg clarithromycin was initiated. Later we reduced the weight to 200mg (half dose) and administered it. Three months after the first visit, sequestrum was removed under local anesthesia, and HBO therapy was performed 20 times after surgery. Further, after consulting with an otorhinolaryngologist, clarithromycin therapy was continued for six months, expecting an immunoregulatory effect [8]; however, sinus mucosa thickening persisted (Fig. 3). This was probably attributed to apical periodontitis of tooth 24; thus, the tooth was extracted under local anesthesia, and the maxillary sinus–oral fistula was enlarged. When the patient consulted a physician for hepatic dysfunction, a mediastinal tumor was suspected, and fluorodeoxyglucose (FDG)-positron emission tomography (PET) was performed (Fig. 4). After PET, mediastinal tumor was ruled out, and the patient was diagnosed with extramedullary hematopoietic foci associated with OP and hepatitis B-related hepatic dysfunction, and treatment for hepatitis was started. Regarding maxillary osteomyelitis, self-irrigation was continued from the sinus–oral fistula. However, her acute symptoms recurred; therefore, antibiotic therapy was resumed, but it was difficult to control the symptoms. Thus, anti-inflammatory and maxillary sinus radical surgeries were performed (Fig. 5). Histopathological examination of the resected tissue revealed osteomyelitis and sequestration in the OP bone (Fig. 6). Six months after the final surgery, the symptoms were stable, and antibiotic therapy was discontinued. For 2 years (4 years since the first visit), the symptoms have not aggravated (Fig. 7).

3. Discussion

This report presents the case of a patient with maxillary osteomyelitis who was diagnosed with OP during childhood. Plain radiography, CT, MRI, bone scintigraphy, and nuclear medicine examinations were performed during the treatment of the patient. Notably, the treatment for OP-related osteomyelitis of the jaw is the same as that for ordinary osteomyelitis of the jaw, and it includes antibiotic therapy and surgical anti-inflammatory procedures such as extraction of the causative tooth and bone sequestration. Among the 24 cases of OP-related maxillary osteomyelitis reported to date (Table 2) [3-7, 9-24], 11 occurred in men and 14 in women, with an average age at the first visit of 31.1 years and a median age of 27.0 (8–66) years. Overall, 17 patients had a history of tooth extraction before the first visit. In addition to radiographic findings, CT, MRI, and nuclear medicine examination data were available for 19, 1, and 5 cases, respectively. In addition to antibiotic therapy, surgical anti-inflammatory procedures were performed in all cases, except one, and HBO was employed in some patients. Regarding the outcomes of these therapies, 20 (83.3%) of 24 patients were found to have relatively good progression without recurrence of acute symptoms. In particular, one patient experienced repeated recurrences of acute symptoms without surgical intervention, whereas the remaining four patients underwent reoperation and other treatments, and their symptoms improved. The average follow-up period was 10.8 (median, 5) months, and recurrent cases were reported to be difficult to manage [11, 12]. In addition, some studies have reported cases of new-onset mandibular osteomyelitis several years after initial symptom remission [5, 22]. Given that a complete cure is difficult to achieve in such cases, continuous oral care is desired even after symptom resolution.

OP is a genetically heterogeneous disease with various onset times. This disease is broadly classified into malignant and benign types. Malignant OP is an autosomal recessive inheritance disease that is diagnosed at birth or in early childhood and is often associated with

poor prognosis. Benign fractures are further classified into types I and II, with type II having a higher rate of multiple fractures [25]. Notably, these fractures have been reported to occur in 1 in 10–50 million people [25]; however, given that only a small number of patients with OP are followed up, asymptomatic or mild cases may be diagnosed with osteomyelitis [4, 16, 18, 19]. Although the present case was diagnosed in childhood, it progressed without any specific treatment. In cases of late-onset OP, recurrent fractures or osteomyelitis may lead to the diagnosis of the disease in adulthood [2]. Therefore, dentists must consider OP given that inflammation may occur.

In cases of osteomyelitis of the jaw, in addition to CT and MRI, nuclear medicine examinations, such as bone scintigraphy, are useful for evaluating osteomyelitis activity and for early detection of lesions. In a previous report of OP-related osteomyelitis, the degrees of osteosclerosis, sequestration tendency, and soft tissue hyperplasia (including sinus mucosa) were evaluated by CT. Only one patient was referred for MRI; however, both T1-weighted and T2-weighted images showed low signals [4], indicating OP-induced sclerotic changes. Some studies conducted nuclear medicine examinations during diagnosis and after treatment [3-7]. However, to the best of our knowledge, no study has reported nuclear medicine examinations during treatment, and only Carvalho et al. used nuclear medicine examinations for evaluating therapeutic effects. Our patient had OP-related maxillary osteomyelitis, and we performed nuclear medicine examinations before, during, and after treatment, although the modalities were different. Bone scintigraphy before and after treatment revealed diffuse uptake in limb bones, knee joints, ankle joints, and skull, probably due to OP, but no uptake was noted in the left maxillary bone. FDG-PET scan during treatment revealed strong uptake of SUVmax 12.631 in the left maxillary sinus; however, there was no uptake in the left maxillary bone, suggesting that increased uptake was associated with maxillary sinusitis.

Nuclear medicine examination is useful for early detection and qualitative evaluation of lesions, osteomyelitis pathophysiology, presence or absence of activity, and evaluation of therapeutic effects. However, in OP-related osteomyelitis, the tracer accumulates in areas where bone metabolism is active, such as fracture sites and long bone metaphysis. Therefore, areas of osteomyelitis may be masked [26, 27], indicating that nuclear medicine studies should be cautiously conducted. In reports of OP-related maxillary osteomyelitis, nuclear medicine examination revealed accumulation in the osteomyelitis area; however, accumulations were also noted in other areas (Table 3) [3-7]. In a study by Carvalho et al., ^{99m}TC-MDP three-phase scintigraphy revealed nonspecific uptake in the left maxillary and zygomatic bones. Therefore, an additional ^{67}Ga scintigraphy was performed, which revealed increased accumulation in the left maxillary bone and zygoma, indicating osteomyelitis [7]. Accordingly, we believe that the addition of ^{67}Ga scintigraphy may have led to increased uptake in the affected maxillary bone, even in our case in which no increased uptake in the affected maxillary bone was observed throughout the course. Despite the high costs and radiation exposure involved, nuclear medicine examination may be useful for both diagnosis and evaluation of therapeutic effects.

4. Conclusion

We encountered a case of OP-related maxillary osteomyelitis and had the opportunity to perform imaging evaluation over time. Conservative therapy, including HBO, and surgical intervention have led to a chronic course of the disease. Given the possibility of a new onset of osteomyelitis, long-term follow-up is desirable. Although 2 years have elapsed since the last surgery in the present case, we plan to continue follow-up.

Ethical Approval

In this report, we anonymized personal information and ensured protection of privacy. The patient provided consent for the publication of this report.

Conflict of interest

The authors declare no conflicts of interest.

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Figure legends

Fig. 1(A) Intraoral photograph taken at the first visit. A fistula was detected in the left maxillary molar region.

(B) Panoramic radiography at the first visit. The floor of the upper left maxillary sinus had a bone defect, indicating loss of trabecular bone and sclerosis in the mandibular body.

Plain computed tomography at the first visit: (C) axial plane, soft tissue mode; (D) axial plane, hard tissue mode; (E) coronal plane, hard tissue mode; (F) sagittal plane, hard tissue mode.

Soft tissue with irregular osteolytic changes was found on the left maxillary jaw in the area of teeth 24, 25, 26, and 27. The cortical bone at the floor of the maxillary sinus disappeared in the area of tooth 26 on the left maxillary sinus into the left maxillary sinus in soft tissue mode. Left maxillary posterior alveolar osteomyelitis and left maxillary sinusitis occurred after separation of the sequestrum. Findings suggestive of sequestration were also found in the area of tooth 24 at the distal and buccal sides of tooth 27.

Fig. 2 Bone scintigraphy using Tc-99m-HMDP before treatment.

No increased uptake in the maxilla and mandible was noted in any arterial, blood pool, and bone phases. Conversely, diffuse and mild hyperuptake was observed in the bones of the extremities, especially those around the left and right knee joints, those around the left and right ankle joints, and the skull, suggesting changes due to osteopetrosis.

Fig. 3 Plain computed tomography during treatment: (A) axial hard tissue mode and (B) coronal hard tissue mode.

Soft tissue thickening from the area of teeth 25 and 27 on the left maxillary jaw to the maxillary sinus persisted. The sequestrum on the buccal side of the area of tooth 27 tended to separate but was not completely separated.

Fig. 4 Fluorodeoxyglucose-positron emission tomography: (A) axial image fused with CT scan of the maxillary sinus and (B) maximum intensity projection image.

Fluorodeoxyglucose-positron emission tomography was performed to investigate a suspected posterior mediastinal tumor. A strong accumulation of SUVmax 12.631 was observed in the left maxillary sinus. Mucosal thickening and partial calcification were also observed, suggesting sinusitis. Weak accumulations of approximately SUVmax 4 were observed in various parts of the vertebral body (Th9, 8, 4, and sacrum), and multiple weak accumulations were observed in the posterior mediastinum, suggesting extramedullary hematopoiesis. Diffuse sclerosis was observed in all bones, which is considered a finding of osteopetrosis, but there was no increase or decrease in fluorodeoxyglucose uptake.

Fig. 5 Intraoperative photograph of general anesthesia surgery.

The mucoperiosteum was incised, detached, and inverted, and the poor granulation and scar tissues were scraped off to achieve mobility of the sequestrum. The sequestra in the centrifugal part were removed using an ultrasonic cutting device, and bleeding from the bone surface was confirmed. The sinus mucosa was abraded and removed, pressure hemostasis was applied, and an absorbable tissue reinforcement material and physiological tissue adhesive were applied. The maxillary sinus was filled with baramycin ointment gauze, the mucoperiosteal flap was sutured, and a hemostatic

sheath was placed to complete the operation. The ointment gauze was removed on day 4 after surgery.

Fig. 6 Histopathological photograph:

(A) magnifying glass.

Bacterial masses were noted on the surface and in the marrow cavity, and the bone cavity was empty.

(B) ×200.

Osteocytes disappeared and many empty bone cavities were observed.

(C) ×100.

There was a tendency for the bone marrow cavity to disappear.

Fig. 7 (A) Intraoral photograph after treatment.

A sinus–oral fistula persisted in the left maxillary molar area.

(B) Plain computed tomography after treatment.

The soft tissue thickening in the left maxillary sinus remained, but pneumatic was maintained. Bone wall thickening in the left maxillary sinus increased over time.

(C) Bone scintigraphy using Tc-99m-HMDP after treatment.

There was a left-to-right difference in the accumulation of Tc-99m-HMDP in the maxilla, but no increased uptake was observed in the maxilla and mandible. The difference was considered to be caused by osteogenesis after anti-inflammatory and maxillary sinus radical surgery and the persistence of sinusitis owing to the 2-year-long period. Similar to bone scintigraphy findings before treatment, diffusely increased uptake was noted in the extremity bones, bones around the knee joints, bones around the ankle joints, and skull.

[i]**Abbreviations**

CT: computed tomography

FDG: fluorodeoxyglucose

MRI: magnetic resonance imaging

HBO: hyperbaric oxygen therapy

OP: osteopetrosis

PET: positron emission tomography

Table 1 The chronological progress report of the patient throughout her treatment

Date		Clinical course
2018	Nov	First visit (Fig. 1a)
		Panoramic radiograph (Fig. 1b)
		CT (Fig. 1c–f)
		MRI
		Technetium bone scan (Fig. 2)
		Clarithromycin (CAM) 400 mg
2019	Feb	Sequestrectomy with local anesthesia followed by 20 times HBO.
		Clarithromycin (CAM) 200 mg for 6 months
	Aug	CT (Fig. 3a, b)
		Extraction of teeth 24 and sinus−oral fistula enlargement was performed under local anesthesia.
		A detailed examination of hepatic dysfunction suggested a mediastinal tumor, and FDG-PET (Fig 4) was performed.
		She was diagnosed with extramedullary hematopoiesis associated with osteopetrosis and liver dysfunction due to hepatitis B.
2020	Apr	Symptoms became acute, and clindamycin (CLDM) therapy was started.
	Oct	Surgical anti-inflammation and maxillary sinus radical surgery were performed under general anesthesia (Figs. 5 and 6) followed by 10 times HBO.
2021	Apr	Ended antibiotic administration.
2022	Dec	Maintaining symptomatic remission. (Fig. 7a–e)

Table 2 List of case reports of osteopetrosis-related maxillary bone myelitis

	Author Year of reporting	Age /Sex	Side involved	History of tooth extraction before the first visit; time of tooth extraction	Imaging modality mentioned in the paper	Surgical treatment	Another management	Outcome; follow-up period
1	Sofferman 1971 [9]	27/F	Left maxilla	Maxillary teeth; 1 year ago	Simple X-ray photography	#Sequestrectomy	#Construction of maxillary prosthesis	no recurrence; 6 months
2	Crockett 1986 [3]	24/F	Left maxilla, zygoma, and pterygoid plates	Multiple teeth; ND	CT, technetium bone scan*	#Sequestrectomy	#Antibiotic therapy #Maxillary denture with an obturator	Recurrence; 3 months (After retreatment, she had no symptoms; for9 months)
3	Hanada 1996 [4]	41/F	Left maxilla	NP	CT, MRI, and technetium bone scan*	#Sequestrectomy	#Antibiotic therapy #Maxillary denture with an obturator	Uneventful; ND.
4	Long 2001 [10]	54/F	Left maxilla	ND (maxillary bone debridement and an attempt at closure of the maxillary oroantral fistula at age 49)	CT	#Extractions of the remaining maxillary teeth #Debridement of the necrotic bone #Sequestrectomy	#Antibiotic therapy	Recurrence; 8 months (After retreatment, he had no symptoms of infection; 1 month)
5	Barry 2003 [11]	28/F	Left maxilla	Several teeth; NM	Panoramic radiography and CT	NP	#Antibiotic therapy	Symptoms persist; 5 years

6	Junquera 2005 [5]	60/F	Left maxilla	Left maxillary and mandibular teeth; 6 months ago	CT and technetium bone scan*	#Sequestrectomy	#Antibiotic therapy	She was asymptomatic, showing complete cicatrisation of the exposed bone; 6 months
7	Hamdan 2006 [12]	12/M	Right maxilla	NP	CT	#Debridement of the necrotic bone.	#Antibiotic therapy	Symptoms recurred within a year. Retreatment improved his symptoms; ND
8	Er 2006 [13]	12/M	Left maxilla	Multiple teeth; 10 months ago	Panoramic radiography	#Removal of the necrotic bone.	#Antibiotic therapy #Topical phenytoin administration	Healing was uneventful; 3 months.
9	Barry 2007 [14]	27/F	Right maxilla	NP	Panoramic radiography and CT	#Debridement #The oroantral fistula process was closed with a buccal advancement flap.	#Antibiotic therapy	Chronic course; ND.
10	Vázquez 2009 [6]	23/F	Right maxilla	NP	Panoramic radiography, Gammagraphy*	#Dental extractions #Curettage of the necrotic bone.	#Antibiotic therapy for 3 weeks	The patient remains in a stable condition without complete healing.

11	Trivellato 2009 [15]	25/F	Right maxilla and zygoma	The teeth 13, 32, 34, and 36; just before (He had a history of a traffic accident with facial abscess in the right zygomatic region the previous year)	Panoramic radiography and CT	#A marginal resection in the right maxilla #The oroantral fistula process was closed with Bichat's buccal fat pad.	#Antibiotic therapy for 1 month	Significant improvement in the healing process and no cutaneous fistula.
12	Krithika 2009 [16]	① 18/M ②8/M ③ 16/M	①Bilateral maxilla ②Bilateral maxilla ③Right maxilla	①Bilateral maxillary molars; 18 months ago. ②ND (Sequestrectomy for maxillary osteomyelitis 4 months ago) ③Right maxillary molar; 2 months ago	①ND ②CT ③Panoramic radiography	①Corticotomy ②Extractions of the maxillary right and left first deciduous molars ③ #Sequestrectomy	#Antibiotic therapy	①The healing was uneventful; 1 month ②The healing was satisfactory, and no recurrence; 4 months ③Uneventful with no recurrence of osteomyelitis; 6 months.
13	Öğütcen-Toller 2010 [17]	18/F	Left maxilla and mandibular anterior	ND	Panoramic radiography	#Extractions of all the malformed teeth and all partially impacted teeth in both jaws. #Sequestrectomy #Closure of oroantral opening with a buccal flap.	#Antibiotic therapy for 6 weeks before surgery and 8 weeks after surgery	Her symptoms subsided and no recurring infections occurred; 2 years.

14	Ambika 2010 [18]	28/M	Right maxilla	Right maxillary molars; 7 months ago. (He has a history of trauma 9 months ago)	Panoramic radiography, CT	#Local debridement and sequestrectomy (partial maxillectomy was planned, but the patient refused treatment).	#Antibiotic therapy for 15 days	Asymptomatic; 3 months (loss of further follow-up).
15	Khademi 2011 [19]	15/M	Bilateral maxilla	Multiple teeth; since 6 years ago (He received antibiotic therapy and three surgical interventions)	CT	#Surgical debridement, curettage, sequestrectomy, and sinus tract excision.	#Antibiotic therapy for 2 weeks	His condition improved, oral mucosa healed and drainage decreased but did not completely subside; 9 weeks.
16	Adachi 2013 [20]	44/F	Left maxilla	Left maxillary 2nd and 3rd molars; 3 years ago	Panoramic radiography and CT	#Curettage of the sequestrum.	#Antibiotic therapy for 7 days	Healed satisfactorily; 4 months.
17	Infante-Cossio 2014 [21]	40/F	Right maxilla and mandible	Multiple; ND (She had sequestrectomies 2 and 5 years ago for osteomyelitis of the jaw)	Panoramic radiography and CT	#Necrotic bone debridement.	#Antibiotic therapy for 4 months	No recurrent episodes of severe infections; 3 years.
18	Kulyapina 2016 [22]	66/M	Right maxilla	Right maxillary third molar; 2 weeks ago	Panoramic radiography and CT	#The oroantral fistula process was closed with Bichat's buccal fat pad #Sequestrectomy #Curettage	#Antibiotic therapy for 1 month #topical decongestive	Recurrence; 3 months (After retreatment, he had no symptoms of infection; 7 years)

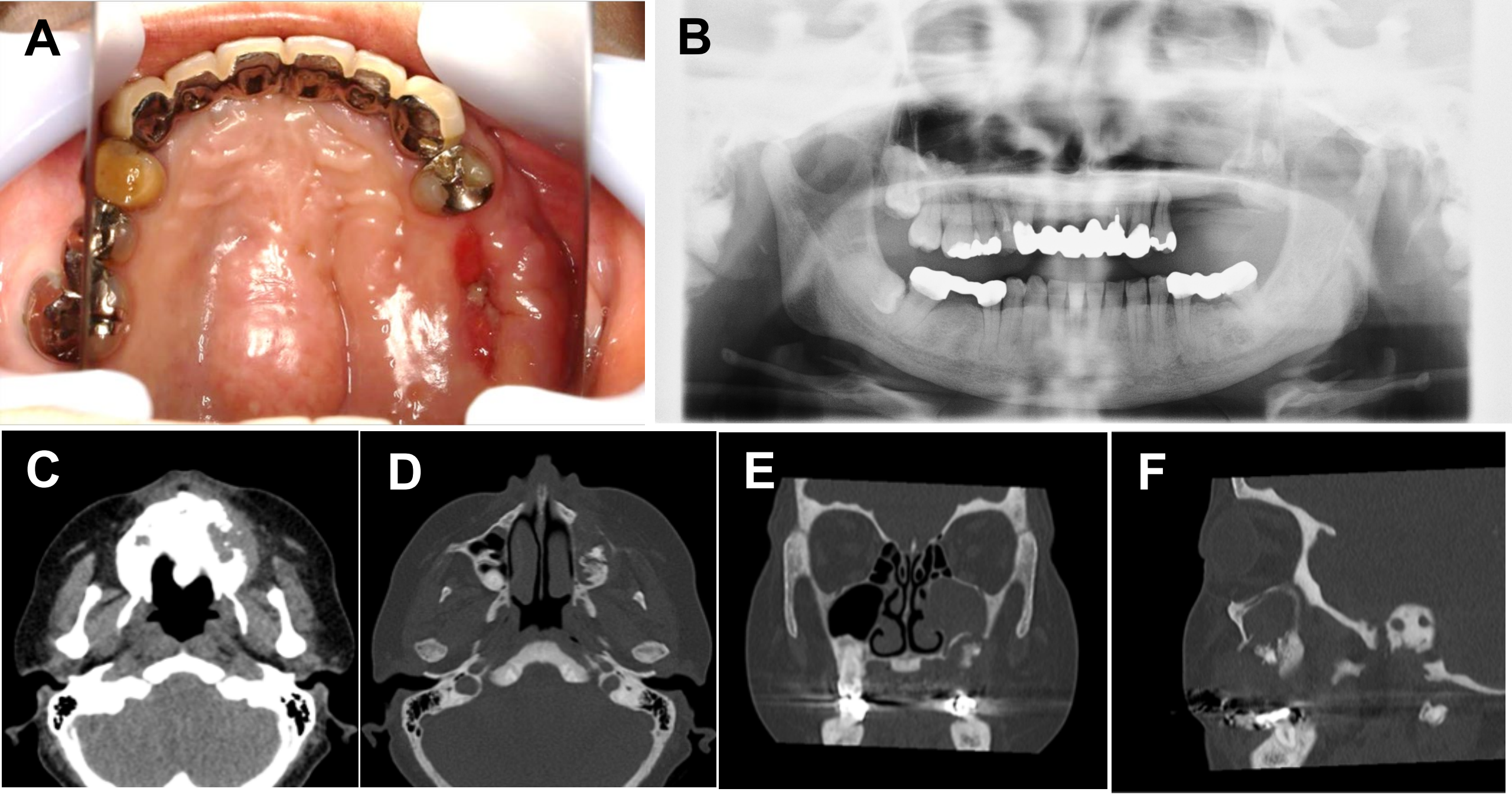
19	Liu 2016 [23]	50/F	Left maxilla	Left maxillary molar; 3 years ago	CT	#Sequestra removal and debridement of the necrotic region were performed via nasal endoscopy.	#Antibiotic therapy for 1 week #The fistula was flushed and drained of pus via nasal endoscopy every 2 weeks	Chronic osteomyelitis was resolved, and the fistula was replaced with fresh granulation; 2 months.
20	Carvalho 2018 [7]	40/M	Left maxilla and zygoma	ND (He had been treated 6 years before with marginal resection for ADO-related mandibular)	CT, technetium, and gallium bone scan*	#Drainage #Sequestrectomy #Extraction of teeth 22, 23, and 24. #Partial resection of the left maxilla and zygomatic bone.	#Antibiotic therapy for 6 months #HBO	Healing was uneventful; 6 months.
21	Sallies 2020 [24]	25/M	Right maxilla	Right maxillary molar; 6 months ago	Panoramic radiography and cone-beam CT	#Extraction of the maxillary first molar #Incision and drainage #Surgical debridement #Advancement flap for closing the wound.	#Antibiotic therapy #Pentoxifylline and tocopherol #Acrylic obturator	ND

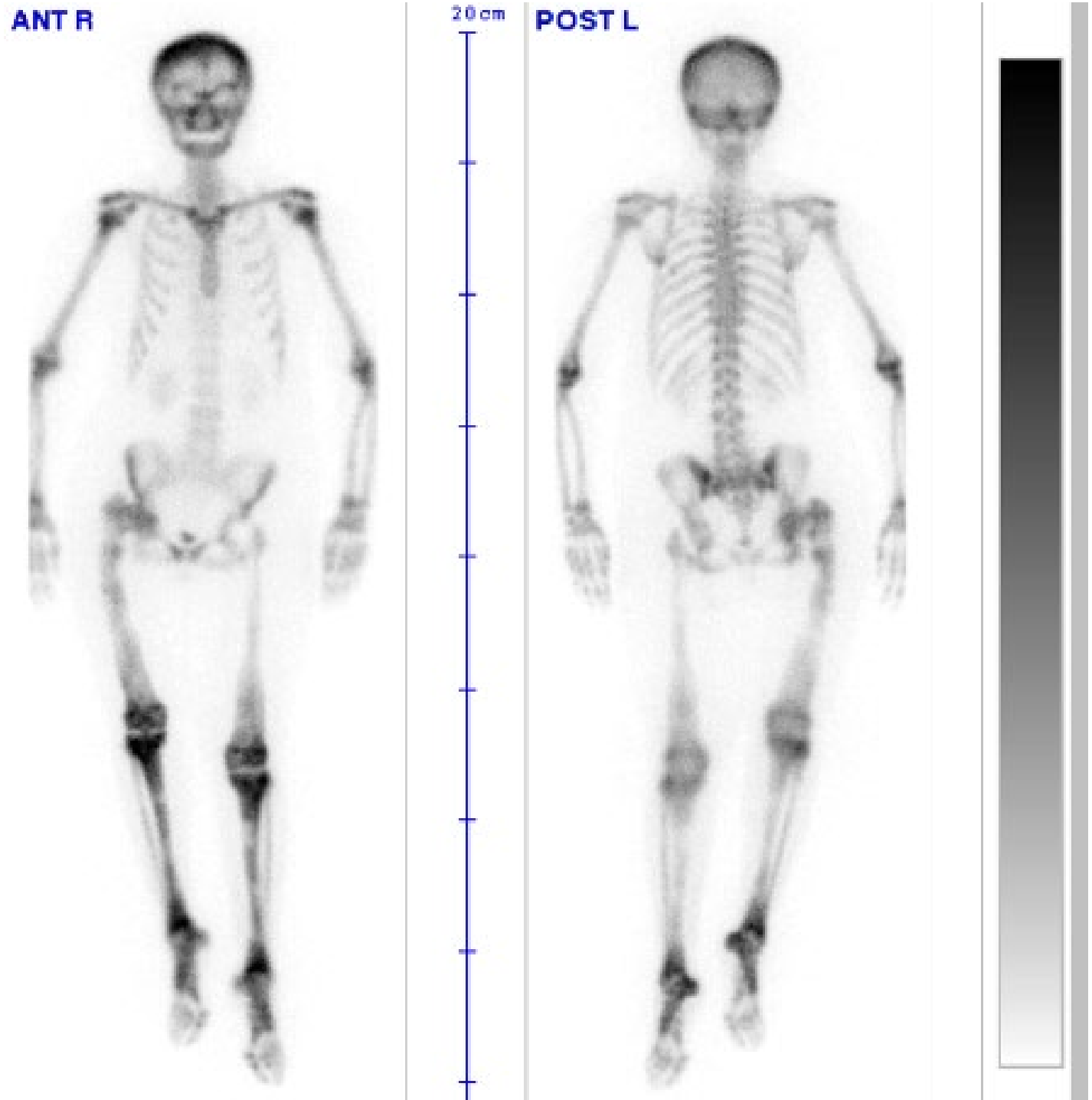
22	Nilesh 2020 [25]	45/F	Left maxilla	Multiple left maxillary teeth; 3 weeks ago	CT	#Surgical debridement.	#Antibiotic therapy	Satisfactory healing with complete resolution of symptoms; 4 years.

ND: not described, CT: computed tomography, MRI: magnetic resonance imaging, HBO: hyperbaric oxygen therapy
*Nuclear medicine examination

Table 3 List of case reports of osteopetrosis-related maxillary bone myelitis with nuclear medicine examination

Author; year of reporting	Nuclear medicine examination	Findings
Crockett; 1986 [3]	Technetium bone scan	Increased uptake was shown in the affected left maxilla and asymptomatic area, left hemimandibe, and both mastoid areas.
Hanada; 1996 [4]	Technetium bone scan	Increased uptake was shown in the affected left maxilla and all long bones, tibia, femur, and humerus.
Junquera; 2005 [5]	Technetium bone scan	Increased uptake was shown in the cranial base, skullcap, left mandibular ramus, and vertebrae, but not in the affected left maxilla.
Vázquez; 2009 [6]	Gammagraphy	Increased uptake was shown in the affected left maxilla and mandible.
Carvalho; 2018 [7]	Scintigraphy Tc & ^{67}Ga	A 99mTc-methylene diphosphonate (MDP) triphasic scintigraphy showed a nonspecific higher tracer uptake in the left maxilla and zygomatic bone. A ^{67}Ga scintigraphy showed uptake in the left maxillary and zygomatic bones.


A
B
C
D
E
F


ANT R
20 cm
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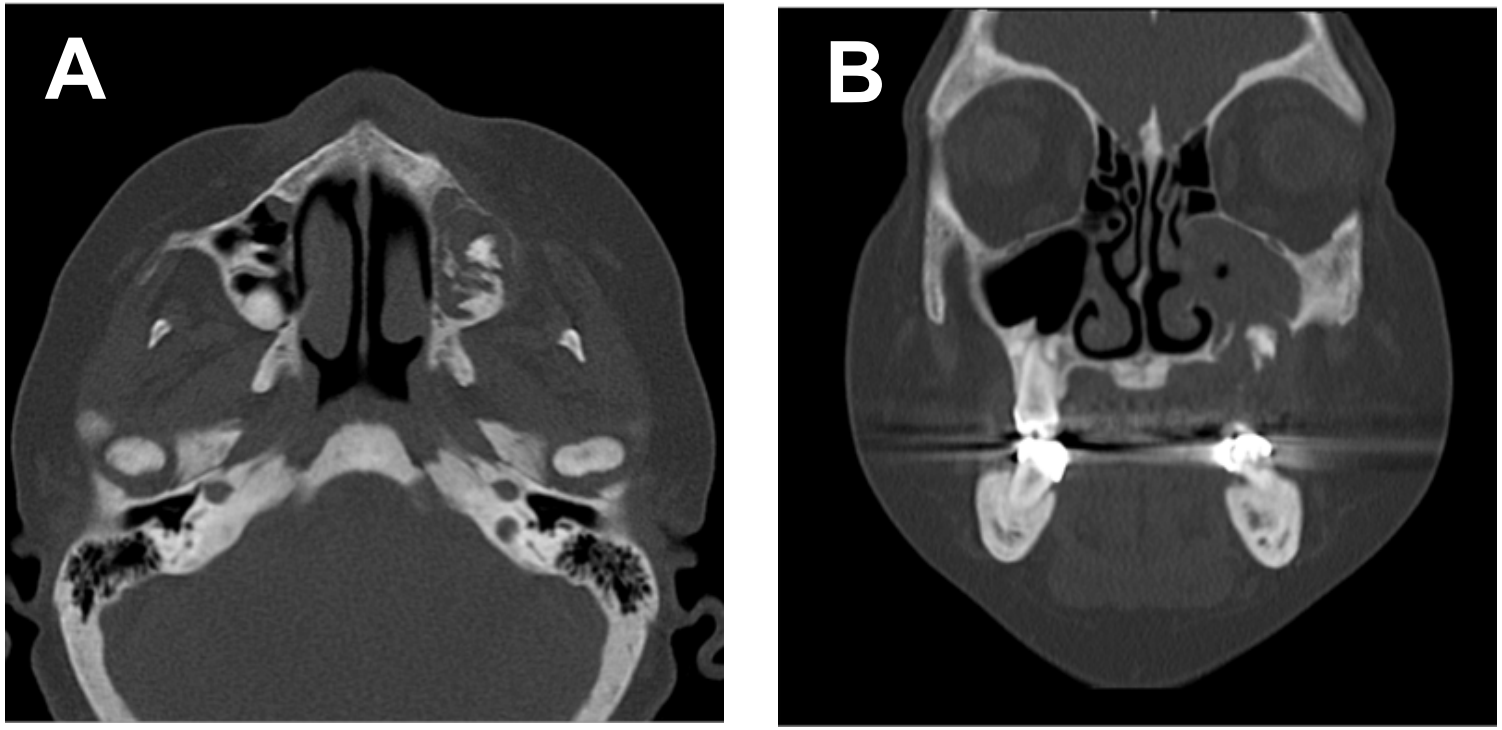
A
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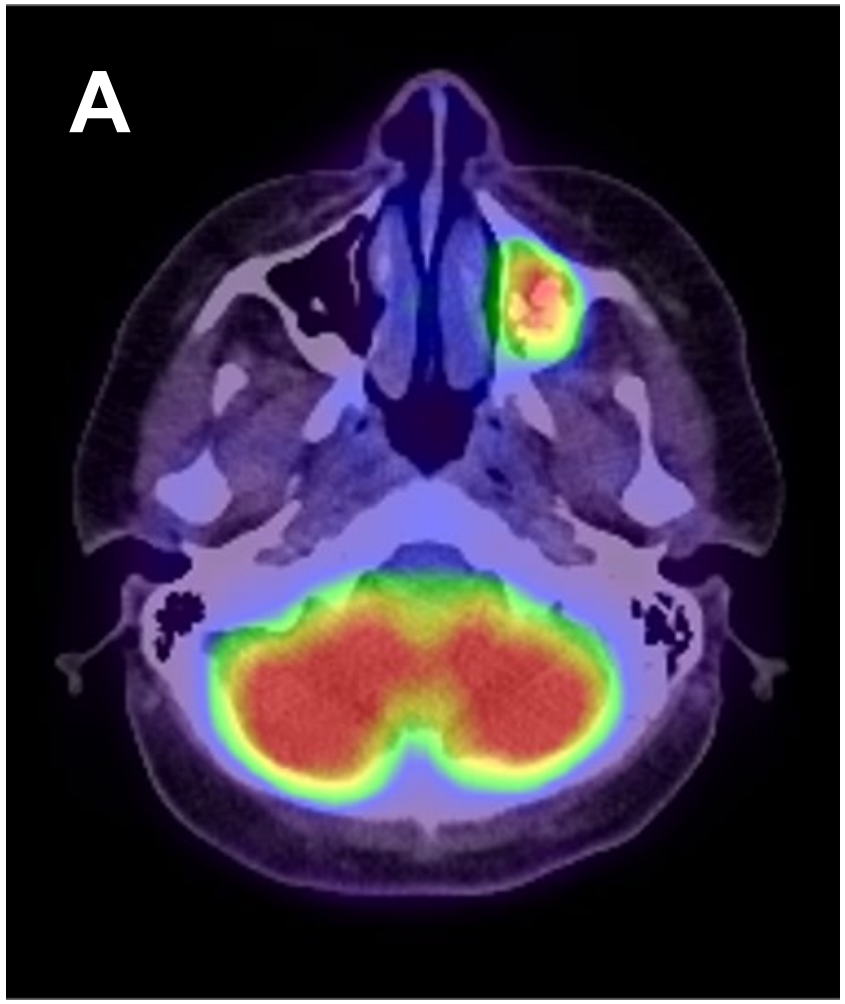
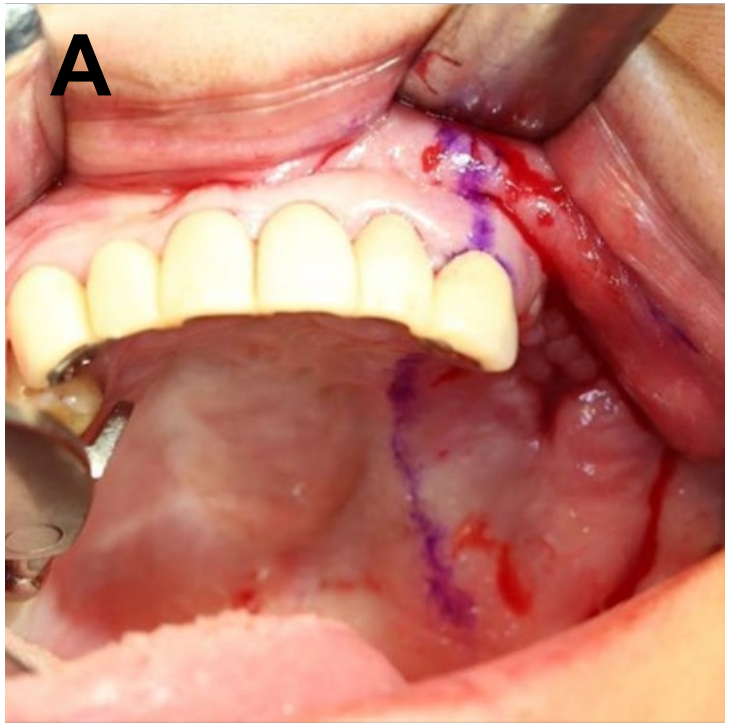
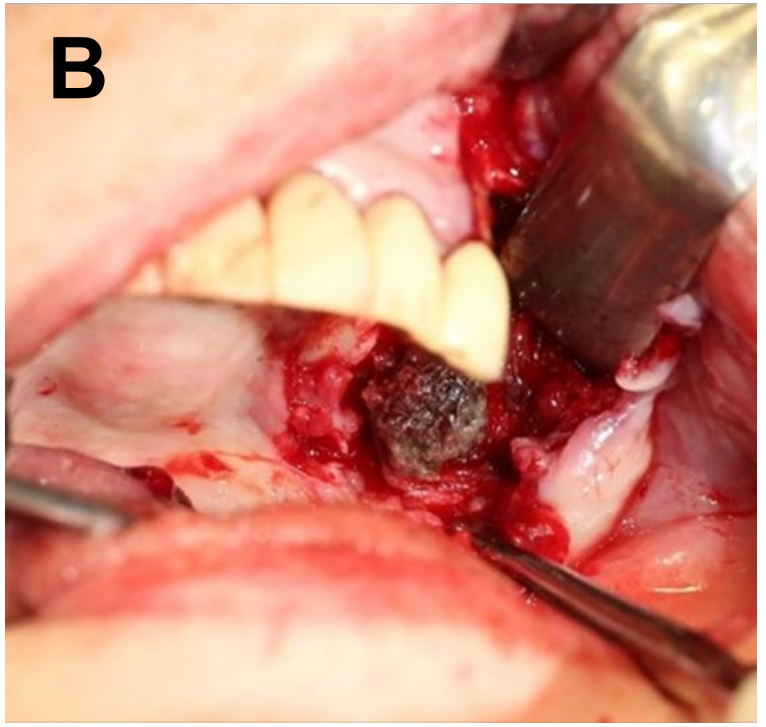
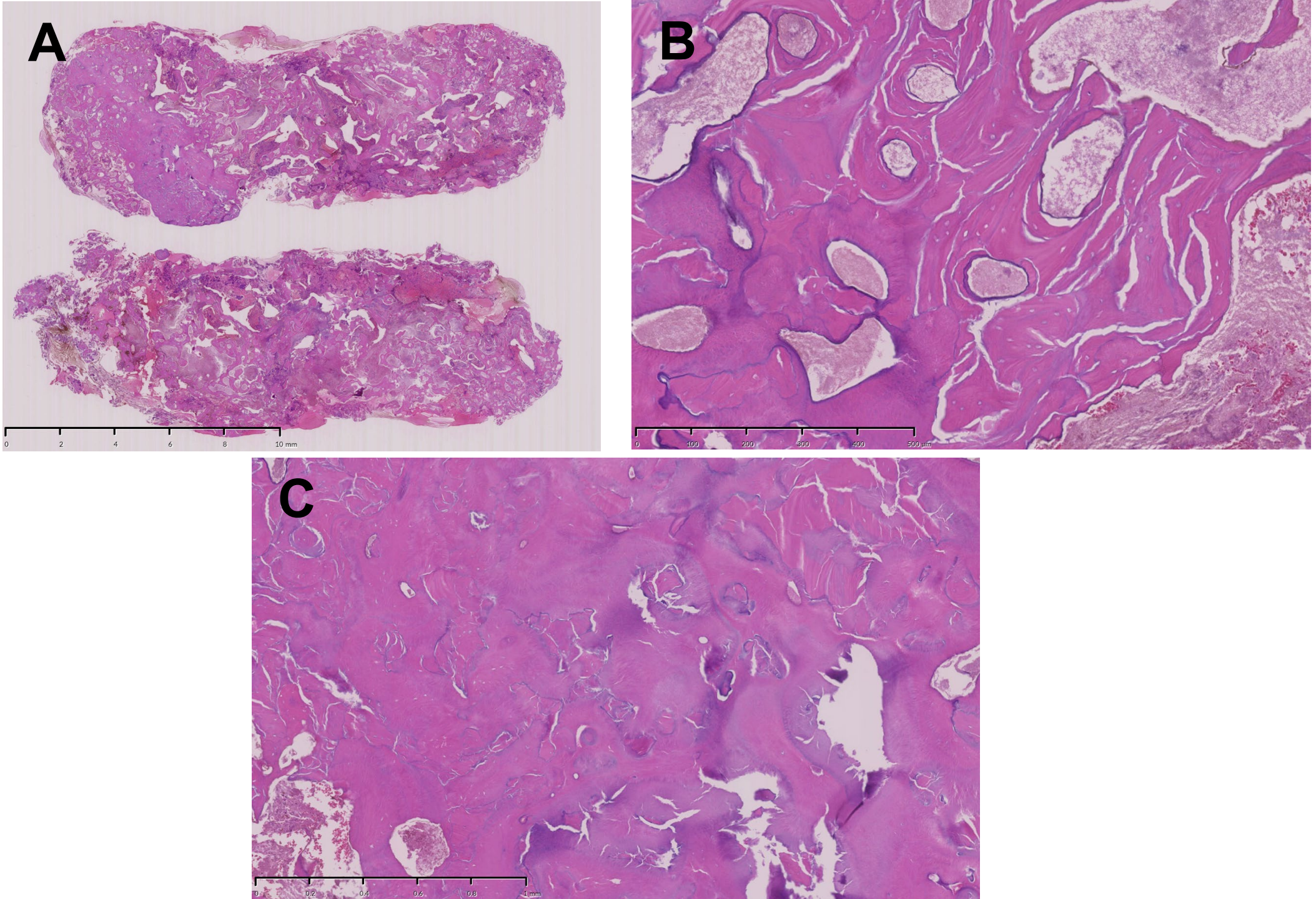

A

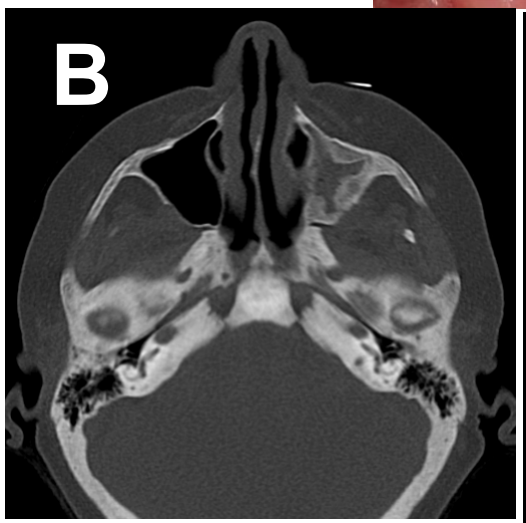

B


A


B


A
0
2
4
6
8
10 mm
B
0
100
200
300
400
500 µm
C
0
0.2
0.4
0.6
0.8
1 mm


A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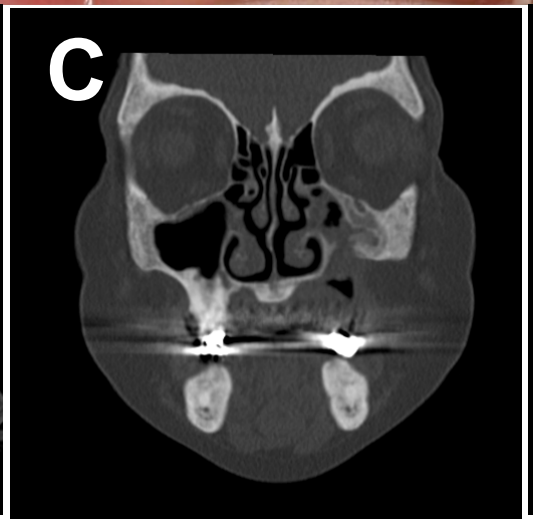
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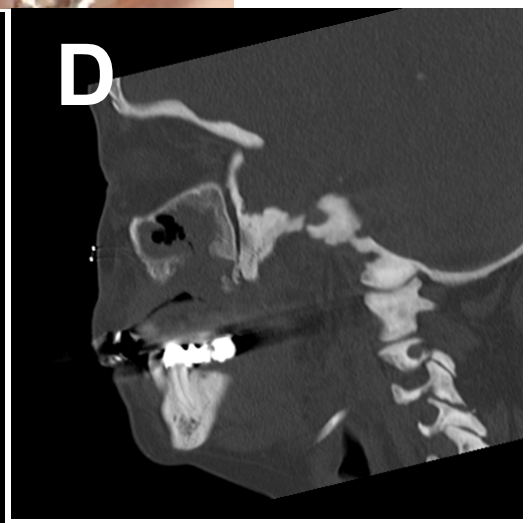
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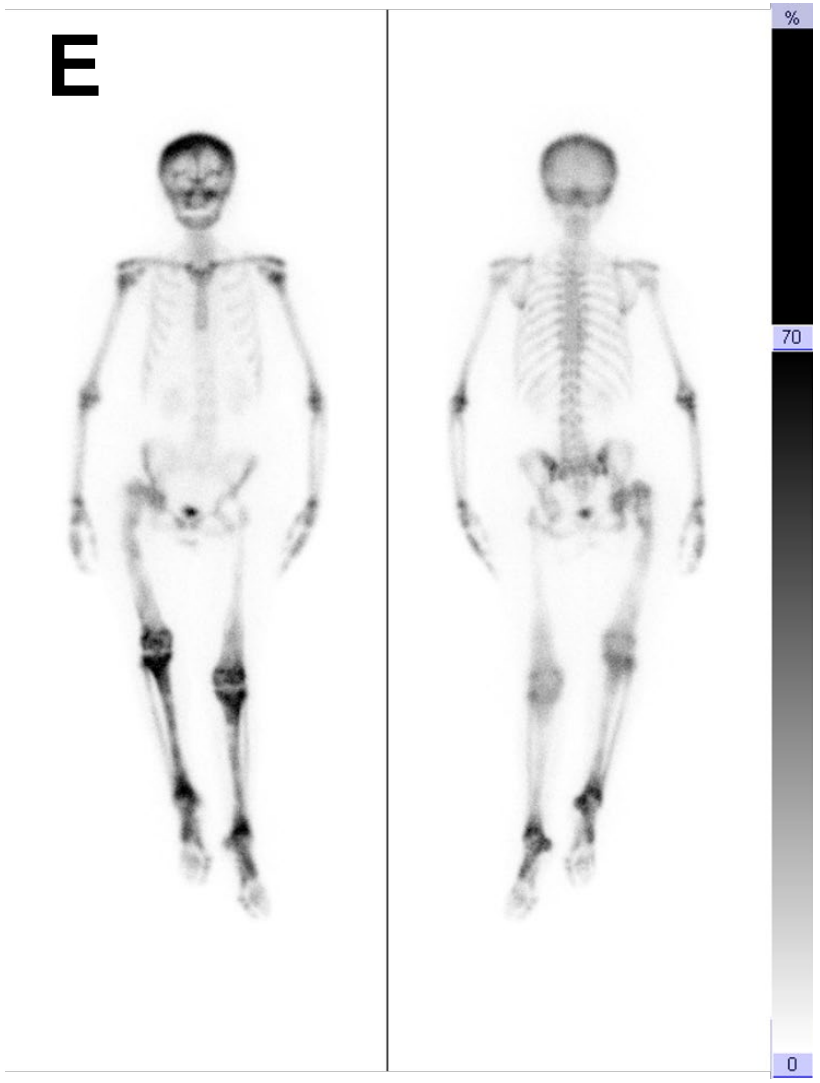
E
%
70
0