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A Pathological Fracture of the Mandible due to Osteomyelitis following a Full Dental Clearance

Abstract: Orofacial infections following dental extractions are a common referral to an oral and maxillofacial department as an emergency, especially when combined with swelling and limited mouth opening. The case presented demonstrates a rare complication of chronic suppurative osteomyelitis with bilateral pathological fractures of the mandible, which occurred following a staged dental clearance.

CPD/Clinical Relevance: Dental extractions are one of the most common treatments carried out by oral surgeons and general dental practitioners. This case highlights a rare but encountered complication of routine oral surgery and demonstrates when it is necessary to make an immediate referral to the local oral and maxillofacial surgery unit.

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Case report

A 58-year-old female patient presented at an oral and maxillofacial outpatient clinic with a nine-month history of pain, swelling and draining fistulas at the right angle of mandible and submental region. This was following a staged full dental clearance by her general dental practitioner. A 10-week course of oral antibiotics had already been prescribed without resolution of symptoms or signs. On presentation she was afebrile with a normal pulse rate and blood pressure. She had a swollen right face, trismus and suppurative infection draining from these fistulas (Figure 1); there was no palpable lymphadenopathy.

Intra-orally there were draining fistulas present on the alveolar mucosa of the right body of the mandible and mobility of the mandible was present at the right angle and symphysis. Radiographic examination was carried out with an OPT but, due to patient factors, a diagnostic image was unachievable and CT imaging was undertaken. This revealed gross destruction of the right ramus and symphysis of the mandible, associated pathological fractures and multiple sequestrae with pockets of gas in the soft tissues in keeping with infection. The coronoid and condylar processes, as well as the proximal body of mandible, were noted to be markedly sclerotic on the right-hand side with a lamellar type periosteal reaction. These features were felt to be consistent with a diagnosis of chronic suppurative osteomyelitis (Figures 2–5). A white cell count, CRP and bone profile were normal.

Her past medical history was significant and included severe rheumatoid arthritis, steroid-induced osteoporosis, major depression, hypertension, severe peripheral vascular disease with a left below

knee amputation and she was therefore wheelchair-bound, peptic ulcer disease, and continued MRSA colonized venous ulceration. Current medications included prednisolone, clopidogrel, omeprazole, mirtazapine, atenolol, furosemide and amiloripene. There was a history of allergy to penicillin and rifampicin. She was an ex-smoker with a 30-pack year history and she drank moderate levels of alcohol.

The patient was immediately admitted to clinic and commenced on intravenous vancomycin and metronidazole on the advice of microbiology clinicians and investigations performed to assess her suitability for surgical intervention. Her clopidogrel was stopped and she was placed on low molecular weight heparin. Whilst waiting for surgery, swabs from the draining fistulas revealed a growth of *Strep. milleri* sensitive to the prescribed antibiotics.

Following the initial management and surgical work-up, the patient underwent surgical debridement of the fracture sites via an extra-oral approach. Necrotic bone was debrided back to healthy bleeding

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bone (Figure 6), the fistulas were excised and rigid internal fixation was applied to the mandible in the form of a preformed 2.5 mm reconstruction plate with bicortical locking screws (Figures 7 and 8). The intra-oral fistulas were excised and closed primarily and a nasogastric tube was placed to enable enteral nutrition whilst the intra-oral wounds healed. Pathological and microbiology specimens taken at the time of surgery confirmed the clinical diagnosis and sensitivities. The antibiotics were continued for a further four

weeks intravenously and the patient was then discharged from hospital. Follow-up of the patient has demonstrated excellent wound and fracture healing with no further complications.

Discussion

This case demonstrates some of the typical features of chronic suppurative osteomyelitis (secondary chronic osteomyelitis), a rare but well documented potential complication of chronic odontogenic infections which general dental practitioners may encounter.

Osteomyelitis is an inflammatory process involving cortical and cancellous bone. In the maxillofacial skeleton the mandible is the most frequently affected bone.¹ It occurs most commonly in the mandible due to the dense, poorly vascularized cortical plates and a single blood supply from the inferior alveolar neurovascular bundle.²

The majority of the time a bacterial focus is identified as a source of infection, however, occasionally in some cases no clear aetiology is found. In modern day practice, osteomyelitis more often presents as a chronic condition and is more commonly associated with debilitated, immunosuppressed or medically compromised patients.³

Classification

Using the nomenclature discussed by Eyrich *et al*,¹ acute and chronic osteomyelitis are differentiated arbitrarily on time: the acute process occurs up to one month after the onset of symptoms and



Figure 1. Draining sinuses at the right submandibular and submental regions.

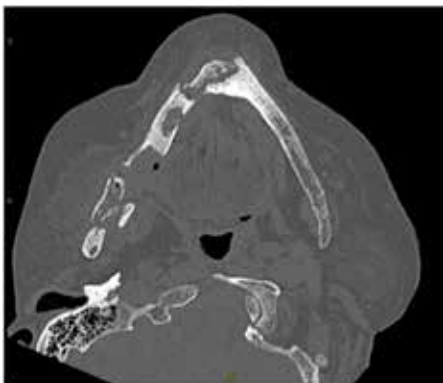


Figure 2. CT axial images demonstrating the pathological fractures and sequestration.



Figure 3. CT coronal reformat showing bony destruction of the right ramus of mandible.



Figure 4. CT coronal reformat showing bony destruction and sequestration at the mandibular symphysis.



Figure 5. 3D reconstructed image of the pathological fractures of the mandible.



Figure 6. Intra-operative view of the right ramus of mandible osteomyelitis.



Figure 7. Rigid internal fixation *in situ* following thorough surgical debridement and fracture reduction.

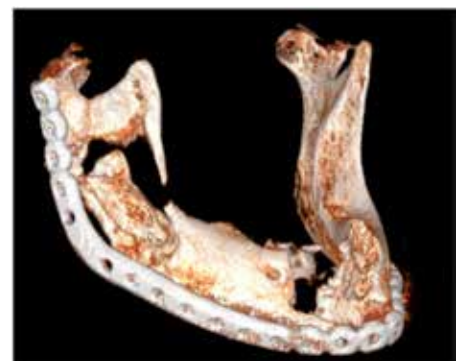


Figure 8. 3D reconstructed image of the post-operative CT following debridement and internal fixation.

chronicity is beyond this point.⁴

Chronic osteomyelitis is often further subdivided into primary chronic osteomyelitis (PCO) and secondary chronic osteomyelitis (SCO). PCO is a non-suppurative chronic inflammation of unknown aetiology which starts insidiously, with no acute phase. SCO is defined as chronic osteomyelitis that develops secondarily to an acute phase, with the presence of suppuration, sequestration, abscess and fistula formation at some stage during the disease process, occurring as a result of a defined, infectious aetiology.⁵

Aetiology

Osteomyelitis of the jaw is a relatively uncommon inflammatory disease in developed countries.⁶ It usually affects adults and is relatively rare in children. The incidence of the disease has dramatically decreased with modern day antimicrobials and improvements in medical and dental care.^{7,8} The primary cause of chronic osteomyelitis of the jaws is infection by odontogenic micro-organisms.⁷ It may also arise as a complication of dental extractions and surgery, maxillofacial trauma and the subsequent inadequate treatment of

a fracture, and/or irradiation to the mandible.⁹ Predisposing factors include autoimmune disease,¹⁰ trauma¹¹ and other conditions affecting the vascularity of the bone, such as radiation, malignancy, osteoporosis, osteopetrosis, and Paget's disease. Systemic factors that cause concomitant alteration in host defences, such as diabetes, steroids and malnutrition, profoundly influence the course of osteomyelitis.¹²

The jaws are unique from other bones in the body by the presence of teeth which create a direct pathway for infectious agents to invade bone by means of caries and

Differential Diagnosis	Clinical Features	Radiological Features	Diagnosis
Osteosarcoma	Pain, swelling, paraesthesia, loss of sensation, increased mobility of teeth, trismus – patients tend to be middle-aged	Irregular; 'sun-ray appearance' – trabeculae of bone radiate perpendicularly around the lesion	Imaging and biopsy
Osteoradionecrosis	Pain, non-healing tissue, history of radiotherapy or bisphosphonate use	Irregular, moth-eaten, minimal evidence of healing	Imaging and biopsy
Osteomyelitis	Intra-oral or extra-oral sinus, pain, swelling, pyrexia, malaise, trismus, paraesthesia of teeth, increased mobility of teeth Patients tend to be at extremes of age	Irregular; moth-eaten, sclerosis of surrounding bone, sequestra of bone	Imaging and biopsy
Squamous cell carcinoma	Non-healing tooth socket, indurated, rolled edges, swelling, increased mobility of teeth, paraesthesia, decreased mobility of adjacent tissues	Irregular, bony destruction and invasion, displacement of teeth	Imaging and biopsy
Ossifying fibroma (cemento-ossifying fibroma)	Painless swelling, slow-growing mass in bone	Radio-opaque associated with root of a tooth and a radiolucent margin	Biopsy – excisional for treatment and for confirmation of diagnosis
Ewing's sarcoma	Pain, swelling, mucosal ulcers, displacement and increased mobility of teeth	Non-specific features, may mimic an infectious process or a malignancy. Moth-eaten destructive radiolucency of the medullary bone with expansion and destruction of the cortex	Biopsy and imaging
Chondrosarcoma	Pain, swelling, increased mobility of teeth	Radiolucency – well or poorly circumscribed, varied appearance, may be multi-locular and contain calcifications	Biopsy – histological confirmation for diagnosis
Metastatic disease	Swelling, increased mobility of teeth, displacement of teeth, paraesthesia	Moth-eaten, ragged edges, radiolucent	Further imaging, biopsy – depending on disease progression

Table 1. Differential diagnosis of osteomyelitis.

periodontal disease.¹³ Oral bone appears to be particularly resistant to infection despite its exposure to oral flora.¹⁴ This further reiterates the rarity of the mandible developing osteomyelitis.

Infection spreads into the marrow spaces of the mandible, resulting in an inflammatory reaction. There is a decrease in the blood supply to the area as there is tissue necrosis and suppuration leading to the development of pus within the bone marrow spaces. The infection can extend through cortical bone, involving the periosteum and resulting in the creation of a collection of necrotic bone submerged in pus. Chronic suppuration will lead to the formation of intra-oral or extra-oral fistulas.

Presentation

Acute osteomyelitis is characterized by a virulent infection with intense pain, tenderness, inflammation, swelling, erythema and limited mouth opening. If, however, the bacteria are less virulent, the symptoms can differ and mimic an acute and prolonged alveolar osteitis, making it difficult to diagnose and thus treat. In its chronic form, features include fistula tract formation, bony sequestration and even pathological fracture.

Imaging

Imaging modalities in investigating this disease process include plain radiography in the form of a dental panoramic radiograph, however, due to the number of differential diagnoses, further imaging should be sought. This is usually in the form of conventional or cone beam CT, which will reveal in detail the extent of bone and soft tissue involvement. Bone resorption is the prominent feature with an osteolytic pattern. Sclerotic areas may be seen around the osteolytic area, particularly in long-standing lesions. There may be a periosteal reaction, which is usually lamellated, and appears as a thin, faint, radio-opaque line adjacent to the surface of the bone. A radiolucent band separates the new periosteal bone from the bone surface. If the process occurs repeatedly, an 'onionskin' appearance is observed.¹⁵ In the early stages of the disease process, MRI T1 weighted images are more useful at detecting marrow changes as inflamed tissue creates low signal intensity in the normally bright signal of fat contained in the marrow.³ MRI

may also be helpful in disease monitoring.¹⁶

Differential diagnosis

The differential diagnoses in cases of insidious onset should include malignant and benign entities, as discussed by Eyrich *et al.*¹ The benign lesions should include ossifying and non-ossifying fibroma. The malignant causes which should be considered are Ewing's sarcoma, osteosarcoma, chondrosarcoma, primary oral malignancies and metastatic disease (Table 1).

Pathogenesis

The widely documented difficulty in managing this condition reflects the pathogenesis, which is thought to be due to the relatively avascular and ischaemic nature of the infected area and sequestrum, thus producing an area of lowered oxygen tension that antibiotics cannot penetrate. The lowered oxygen tension impairs the bacteriocidal activities of polymorphonuclear cells and converts a previously aerobic infection into one that is anaerobic. The diffusion rate of antibiotics into the dead bone is so low that, regardless of the external concentration, adequate penetration is impossible. This may lead to ineffective antibiotic concentrations at the site of infection, despite adequate therapeutic serum concentrations.¹⁷ Early diagnosis of this destructive infection can reduce the need for surgical therapy and allow for the prevention of major complications. These include sepsis, gross mandibular destruction, pathological fractures and, in its chronic form, recurrence.

Treatment

Management in this case entailed a course of antibiotics in combination with aggressive surgical debridement. This is consistent with published protocols.^{7,18,19} It has been suggested that the minimum duration of antibiotic therapy to treat CSO is two weeks.²⁰ However, it has also been suggested by Bamberger²¹ that a minimum of four weeks is indicated. Fracture treatment requires rigid fixation; traditionally methods of treatment for pathological fractures include external fixation and inter-maxillary fixation. However, there is increasing evidence for the use of rigid internal fixation in the treatment of infected fractures.^{22,23} Adjuvant treatments available include ozone gas or hyperbaric oxygen

therapy,³ but these are reserved for persistent cases where standard treatment has failed, or in cases with already impaired vascularity.

Conclusion

This case is an example of when simple dental treatment can result in significant complications and morbidity. Early diagnosis of this destructive infection can reduce the need for surgical therapy and allow for the prevention of major complications. These include sepsis, gross mandibular destruction, pathological fractures and, in its chronic form, recurrence. The main reason for delayed diagnosis is as a result of a general lack of awareness of this condition, its risk factors and its presenting features.

This case had significant risk factors for the development of osteomyelitis as well as a characteristic clinical presentation. The diagnosis was missed, unfortunately, and treated with repeated courses of antibiotics for what was thought to be a simple 'slow to respond' post-operative infection. This led to significant progression in the disease process and significant morbidity.

Long-term close follow-up will be necessary to ensure that there is no recurrence of the infection or complications arising from the treatment carried out.

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