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Case Report

An unusual case of total maxillary sequestration in a diabetic patient

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Summary
Osteomyelitis of the jaw is of relatively low incidence. Maxillary Osteomyelitis is however rare compared with mandibular osteomyelitis. The extensive blood supply and the strut-like bones of the maxilla make it less prone to chronic infections. Systemic conditions that alter the host's resistance such as diabetes mellitus, autoimmune disorders, agranulocytosis, anaemia, especially sickle cell anaemia are predisposing factors for the development of this condition. An unusual case of chronic maxillary osteomyelitis induced by trauma in a diabetic, with subsequent atypical necrotising ulceration of palatal mucosa resulting in total maxillary sequestration is presented.

Keywords: *Maxillary osteomyelitis, sequestration, diabetes mellitus*

Résumé
L'ostéomyelite de la joue a une incidence relativement faible. L'ostéomyelite du maxillaire est cependant rare comparé á l'ostéomyelite mandibulaire. L'innervation en sang et la nature des os du maxillaire font d'elles la réduction en infections chronique. Les conditions systémique qui alternent la résistance de l'organisme telsque le diabete mellite et les désordres auto-immunitaire, l'agranulocytose, l'anémie specialement l'anémie drépanocytaires sont les facteurs prédisposant le developement de cette condition. Un cas non habituel d'ostéomyelite chronique maxillaire induit par le traumatisme chez un diabétique avec une ulcération non typique de la mucose pallate résultant á une sequestration maxillaire totale est presentée.

Introduction

By strict definition: Osteomyelitis is an inflammation of the medullary portion of the bone. However, the process is rarely confined to the endosteum and usually affects the cortical bone and periosteum. Therefore, osteomyelitis may be considered an inflammatory condition of bone that usually begins as an infection of the medullary cavity which rapidly involves the Haversian system and quickly extends to the periosteum.

The incidence of osteomyelitis of the jaws is relatively low. This is remarkable, considering the high frequency and severity of odontogenic infections. This low incidence is a result of a fine balance between the host resistance and the virulence of the micro-organisms. The virulence of the micro-organisms in addition to any condition altering the host defence mechanism and alteration of jaw vascularity are important in the onset and severity of osteomyelitis. Systemic conditions that alter the host's resistance and influence the course of the disease include diabetes mellitus, autoimmune disorders, agranulocytosis, and anaemia, especially sickle cell anaemia. Others are leukaemia, AIDS, malnutrition, anticancer chemotherapy and steroid drug use. The importance of controlling these conditions in order to achieve proper response from the treatment of osteomyelitis cannot be over emphasised.

Here we report our experience in the management of a case of unusual chronic maxillary osteomyelitis in a diabetic patient. The atypical presentations of the condition as well as the challenges of diagnosis and treatment are highlighted.

Case summary

A 43- year old female was referred to our hospital following the discovery of a painful, persistent mid-facial swelling associated with an area of anterior palatal mucosal necrosis resulting in oro-nasal communication.

The patient was said to have been involved in a road traffic accident 5 weeks earlier. She was observed to have sustained a blunt soft tissue injury to the face and an open fracture of the proximal aspect of the left tibia. She was being managed in a private hospital where an infection of the open tibia fracture was noticed 2 weeks post injury. A week later (3 weeks post injury), a progressive and painful facial swelling was observed which persisted inspite of parenteral antibiotics. An associated necrotizing ulcerative process of the palatal mucogingival was similarly noticed which further progressed to anterior palatal perforation with a resulting oro-nasal

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communication. The patient was consequently referred to our hospital for further management.

On admission in our hospital, clinical examination revealed a chronically ill-looking middle age woman, fully conscious and alert, pale, dehydrated, afebrile and anicteric. Essential findings in the oral and maxillofacial region were diffuse mid facial swelling, severe halitosis with a wide area of an irregular ulcer in the anterior two-third of the palate associated with mucosa necrosis and fistulous communication with the nasal cavity. The necrotizing process also extended to involve the nasal mucosa with pus drainage through the nostrils. There was also an area of exposed palatal bone which appeared necrotic.

Examination of the open tibia fracture revealed a granulating wound extending from the distal third of the left femur to the proximal third of the tibia anteriorly. There was a small area of exposed tibia measuring 4cm in length. All other systems appeared clinically normal.

A background immunosuppression resulting in a fulminant oral infection was suspected. The base line investigations conducted revealed anaemia (PVC 26%), E & U values were within normal range. FBC showed mild granulcytosis while retroviral screening was non reactive. Random Plasma Glucose of 500mg/dl was obtained. Craniofacial CT scan revealed fluid congestion of the maxillary and ethmoidal sinuses. A swab of the oral wound was obtained for microscopy, culture and sensitivity (M/C/S). Incisional biopsy of the oral ulcer was performed and empirical intravenous antibiotics (Ceftriaxone 1g 12 hourly, Metronidazole 500mg 8 hourly) were commenced.

During the course of treatment, progressive loosening of maxillary dentition was noted. This later resulted in spontaneous exfoliation of teeth (Fig. 1). The patient was reviewed by the endocrinologists and was commenced on mixed-split insulin therapy with subsequent attainment of euglycemia. Patient was rehydrated with isotonic saline and transfused with 2 units of whole blood to correct anaemia. The leg wound was reviewed by the plastic surgeons and regular honey dressing was instituted. The patient was also prepared for debridement of the necrotic oro-nasal wound under general anaesthesia.

Fig. 1: Palatal mucosa necrosis with spontaneous exfloliation of anterior teeth

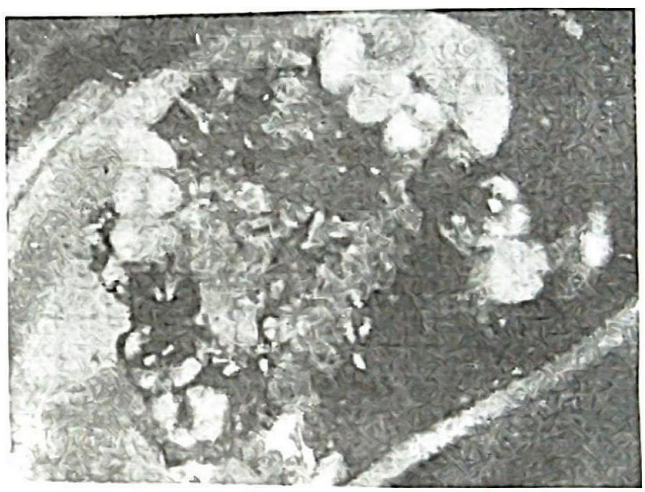

Fig. 2: Sequestrated maxilla

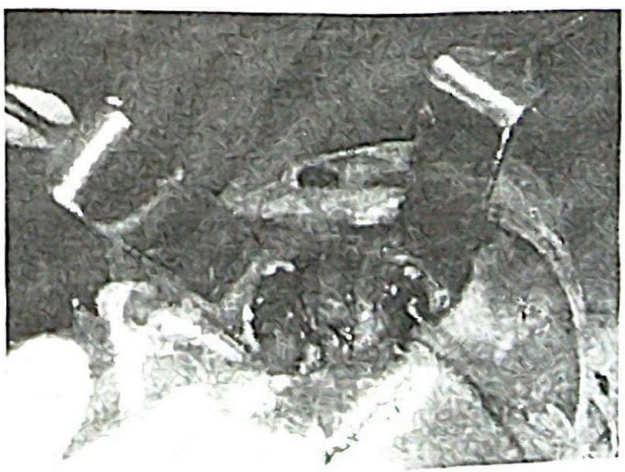

Fig. 3: Cavity after maxillectomy

Intraoperatively, complete sequestration of the entire maxilla was observed, consequently total maxillectomy at Le fort ii level was performed (Fig. 2 and Fig. 3)and affected nasal and antral mucosa excised and sent for histopathological examination. A mixed culture of organisms was obtained from the oral wound swab and these were sensitive to our choice of antibiotics, making further investigations into nature of specific organisms unnecessary. The histopathology was reported as chronic inflammatory cells in granulation tissues with associated tissue necrosis.

Peri and intraoperatively, patients had glucose-insulin infusion with close monitoring of blood sugar. She was recommenced on subcutaneous bolus insulin as soon as she resumed normal oral feeding. Glycaemic control was maintained, adequate nutrition, haematinics and rigorous antibiotic therapy were ensured. Anaemia was corrected to a PCV of 36% at the time of discharge from the hospital. The leg wound granulated well and healed satisfactorily after skin grafting. The general health condition of the patient improved. The resulting palatal defect was obturated with the use of an interim feeding plate and finally definitive obturator was fabricated.

We concluded that the condition was a case of unusual maxillary chronic osteomyelitis initiated by trauma and exaggerated by the undiagnosed background diabetes mellitus. The final treatment was satisfactory and patient was discharged home on oral hypoglycaemic agents. She has subsequently been reviewed in our outpatient clinic and appropriate diabetes related care is also being ensured.

Discussion

Chronic osteomyelitis of the jaws, especially the maxilla, is not very common. It occurs in about 1 in 5,000 people in the world literature. Some of the most common symptoms are pain in the bone, local swelling, redness, and warmth, fever, abscess at the site of infection and formation of skin sinuses. The extensive blood supply of the maxilla makes it less prone to osteomyelitis when compared to the mandible, while the thin cortical plates and porosity of the medullary portion preclude infections from becoming contained in the bone and facilitate spread of oedema and purulent discharge into the adjacent tissues.

The present case presented initially with a necrotising lesion, but the typical cratered ulcers of interdental papillae with yellowish grey slough and plaque accumulation usual of acute necrotising ulcerative gingivitis (ANUG) were not seen. The spread of the necrotising ulcer in necrotising ulcerative gingivitis is normally along the gingival margin with extensive destruction of interdental tissues, this was not the case with this patient. These findings prevented specific investigations diagnostic of ANUG.

The role of trauma as a predisposing factor to the osteomyelitis was exemplified. A patient who had sustained only blunt trauma to the midface however would not normally progress to osteomyelitis of the underlying bone. The extensive vascular compromise that resulted in complete sequestration of the entire maxilla in this case was unusual. The maxilla being a richly vascularized bone possesses an inherent resistance to infection, the previously undiagnosed diabetes in our patient might explain the rapid progression of the disease. The quick response to treatment following glycaemic control was an indication of the major role played by diabetes mellitus in this condition.

The striking observation of rapidly progressive necrotizing palatal mucogingival ulceration, perforation into the nasal cavity and nasal mucosa necrosis suggested a more lethal disease process. These are not features typically associated with chronic osteomyelitis of the jaws, thus making definitive diagnosis challenging. However, it is our opinion that local microangiopathy secondary to diabetes mellitus, the associated anaemia and vascular thrombosis secondary to infection significantly compromised the local microcirculation resulting in ischaemic necrosis of both soft tissues and bone in the region. This might have predisposed to the necrotizing ulcerative process and bone infarction which led to the oro-nasal communication. The rapid response of the patient to treatment adopted supported the fact that the disease was an inflammatory process exercabated by undiagnosed diabetes mellitus.

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