Osteomyelitis of Maxilla involving Multiple Cranial Bones

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***Abstract* - Osteomyelitis of the jaws is rare in today era due to the advance of antimicrobial agents and osteomyelitis of the maxilla is even extremely rare. Osteomyelitis of the jaws is associated with a complex microbiota. The two prominent causes are odontogenic infection and sinusitis. Osteomyelitis is an acute or chronic inflammatory process that can involve cortical and trabecular aspects of bone or bone marrow. Cranial bones are infrequently involved, but spreading of inflammation with involvement of surrounding structures represent the important risks, as are cerebral abscess, encephalitis, or meningitis. This study presents a case of osteomyelitis of maxilla involving multiple cranial bones caused by spreading of contiguous inflammation sustained by *Acineto-bacter baumannii*. The resolution of infection was gained with a combination of surgical treatment and antibiotic therapy.**

Keywords; osteomyelitis, maxilla, cranial bones, Acinetobacter baumannii

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Introduction

Osteomyelitis can be defined as an inflammatory condition of the bone, which begins as an infection of the medullary cavity, rapidly involves the haversian systems, and extends to involve the periosteum of the affected area. Conditions altering the vascularity of the bone such as radiation, malignancy, osteoporosis, osteopetrosis, and Paget's disease pre-dispose to osteomyelitis. Systemic diseases like diabetes, anemia and malnutrition that cause concomitant alteration in host defenses profoundly influence the course of osteomyelitis. Necrosis of maxilla can be caused by fungal infection, trauma, irradiation, Herpes zoster, necrotizing sialometaplasia, midline lethal granuloma, Gaucher's disease, bisphosphonate related osteonecrosis of jaw (BRONJ), and bacterial infection.

In tooth-bearing bone, osteomyelitis is usually caused by polymicrobial odontogenic bacteria. Oral microflora which are most frequently involved in osteomyelitis are gram negative anaerobic rods and facultative anaerobic cocci of staphylococcus and enterococcus (Poonia *et al*., 2016). Specific infectious agents responsible for chronic osteomyelitis are difficult to identify. Nearly 80-90 % of chronic cases demonstrated *Staphylo-coccus aureus* and *Staphylococcus epidermidis*. Fungal organisms such as *Candida parapsilosis* and *Aspergillus* also have been reported causing craniofacial osteo-myelitis, especially in patients who are immuno-compromised (Elangovan & Srinivasa, 2013).

Acinetobacter baumannii is a gram-negative coccobacillus that is the etiological agent of nosocomial infections resulting in septicaemia, meningitis, endocarditis, pneumonia, and wound and urinary tract infection. It has also been identified as an ESKAPE pathogen (*Enterococcus faecium, Staphylococcus aureus, Klebsiella pneumoniae, Acineto-*

bacter baumannii, Pseudomonas aeruginosa, and Enterobacter species). Other symptoms are fever and chill, rash, confusion or altered mental state and muscle pain. A total of four strains had different isolation site (Zurawski *et al*., 2012). In 101 patients with osteomyelitis of jaw, the most frequently isolated agents were *Enterobacter species, Acinetobacter baumannii,* and *Pseudomonas aeruginosa* representing about 70% of all the isolates (Cordeiro *et al*., 2012).

Dealing with osteomyelitis in head and neck bones is not the same as in other bones of the body due to the nature of the bones, complex anatomy of the region, and aesthetics. The general lack of awareness of the prevalence of the disease and its features often leads to a misdiagnosis and delay in treatment. Early detection of this condition and prompt attention will prevent the need for a surgical intervention in an otherwise protracted course of illness.

Table 1. Jaw osteomyelitis classification (Lew & Waldvoger, 1997)

Suppurative osteomyelitis	Non-suppurative osteomyelitis
Suppurative acute osteomyelitis	Chronic osteomyelitis
Primary suppurative chronic	Diffuse sclerosing osteomyelitis
Osteomyelitis secondary preceding phase	Focal sclerosing osteomyelitis
Infant osteomyelitis	Proliferative periostitis
	Osteoradionecrosis

Case Report

A 50 year-old male with history of pain at the right upper molars and premolars region last 2 years ago.

Patient had undergone dental treatment at that region and after 1 week later, he was hospitalized at 1000-bedded Nay Pyi Taw Hospital for facial edema at the right side and high fever. And transferred to Yangon General Hospital for extremely high fever, neurological complication of 6^{th} and 8^{th} cranial nerve palsy and hearing loss at the right side. Six months after the onset, hearing loss also presented at the left side. The audiogram revealed right total sensorineural hearing loss and left partial sensorineural hearing loss. Halitosis and purulent discharge in the oral cavity were noticed. There was no relief from medication, the swelling persisted and he was referred to Eyes, Nose, Throat (ENT) hospital. It was diagnosed as basal meningitis with periodontal abscess due to pansinusitis and right parapharyngeal abscess according to MRI result. There was no evidence of pulmonary tuberculosis and no evidence of *Mycobacterium tuberculosis* on marrow biopsy. One year after the onset, punch biopsy was performed to exclude the paranasal sinus (PNS) tumor, but only negative biopsy for malignancy.

On general examination patient was moderately built, poorly nourished and anemic. The patient also presented marked deterioration of his general state of health and poor oral hygiene. On extraoral examination, there is slight swelling at the right side of cheek (Figure 1). Patient had no medical history of hypertension or diabetes mellitus. There was also total hearing loss on both sides. On examination of oral cavity, multiple missing teeth in the left and right upper posterior regions were noted. But, there was no history of tooth extraction at these regions and exfoliated gradually. A large area of brownish-discolored, loose, exposed necrotic bones (sequestrums) at both sides of right and left upper posterior region was found (Figure 2). There was tenderness on palpation of gingiva along the region of sequestrum.

Upper anterior teeth showed grade III mobility. Other intraoral findings included multiple carious teeth and chronic generalized periodontitis.

Patient was treated with oral co-amoxiclav 625 mg three times per day and metronidazole 200 mg three times per day for 2 weeks and followed by oral doxycycline 100 mg twice a day and dazolic 500 mg twice a day for further 4 weeks.

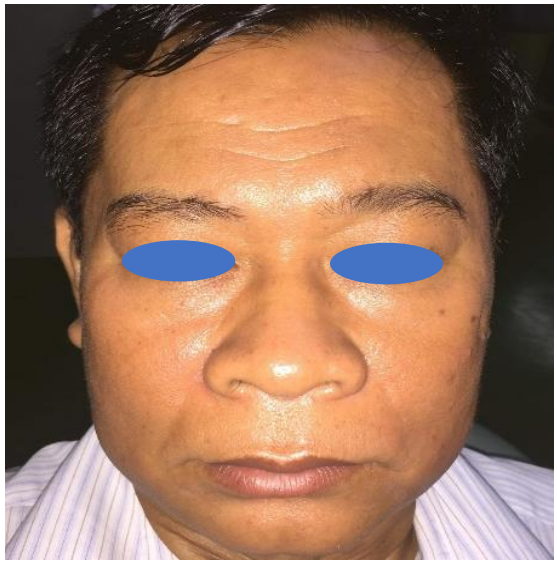

Figure 1. Preoperative photo showed facial swelling at the right side of cheek

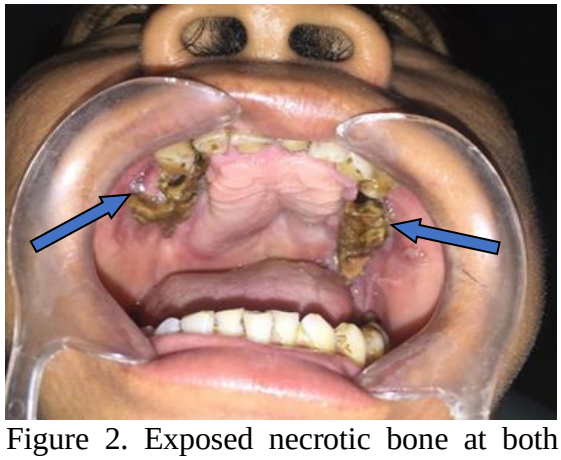

Figure 2. Exposed necrotic bone at both sides of maxillary posterior region

Investigations

The routine hematological investigation was normal except for leukocytosis and elevated ESR. Patient was seronegative for HIV and hepatitis. There was no history of steroid therapy. Panoramic radiograph demonstrated diffuse radiolucency extending from periapical region involving the palate. The patient was advised to get a computed tomography (CT) scan to know the nature and extent of the disease. CT scan showed chronic osteomyelitic changes of maxilla, floor of the temporal fossa and base of the skull. Bilateral maxillary sinusitis and left mastoiditis were also noted. Thickened and sclerosed left maxillary sinus lateral wall and left malar prominence indicate chronic osteomyelitic changes (Figure 3).

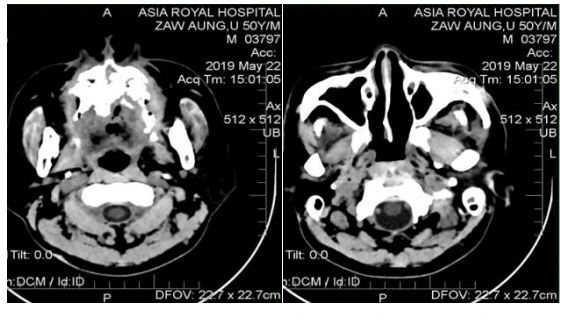

Figure 3. CT scan showed (A) chronic osteomyelitic changes of maxilla, floor of the temporal fossa and base of the skull. Bilateral maxillary sinusitis and left mastoiditis were also noted. (B) Thickened and sclerosed left maxillary sinus lateral wall and left malar prominence

Treatment

After the patient's infection was controlled, sequestrectomy and maxillectomy was planned under general anesthesia.

Crevicular incision was given on both buccal and palatal aspects. The flap was reflected. As operative finding, the whole maxilla detached from palatal bone as a

sequestrum. Fibrous tissue and small amount of bone covered the palatal bone (Figure 4). Sequestrectomy and curettage was performed along with total extraction of maxillary teeth and Type II subtotal maxillectomy (According to Cordeiro's maxillectomy classification) (Figure 5). Patient was kept under the antibiotic coverage. Specimen sent for histo-pathological examination and culture and sensitivity test. Report confirmed the lesion to be chronic osteomyelitis of maxillae which is a rare condition. Culture report showed *Acinetobacter baumannii* complex which causes osteomyelitis. According to antimicrobial sensitivity test, Tazobactam, Ceftriaxone, Sulbactam, Cefepime, Imipenem, Gentamycin, levofloxacin, Minocycline, Tigecycline and Colistin are sensitive. Postoperative period is uneventful (Figure 6). The patient was recalled after one month and showed improvement with resolution of the lesion. Prosthetic rehabilitation with removable maxillary denture was fabricated in Department of Prosthodontics (Figure 7).

Table 2. Cordeiro's maxillectomy classification (Cordeiro, 2012)

Type	To be resected
Type I. partial	1 – 2 maxillary walls
Type II. Subtotal	3 – 5 maxillary walls including palate
Type III. Total	All 6 maxillary walls a) Lacking orbital exenteration
Type IV. Radical	b) With orbital exenteration 5 maxillary walls, palate and orbit excepted

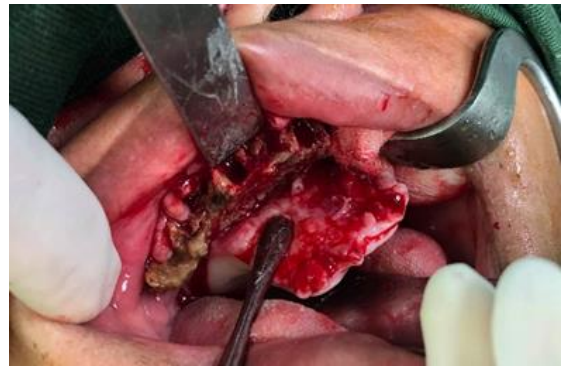

Figure 4. Intraoperative finding as the whole maxilla necrotized

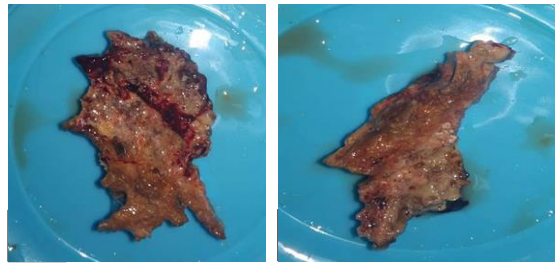

Figure 5. Excised Necrotic Maxilla at both sides of maxillary posterior region (A) Left side (B) Right side

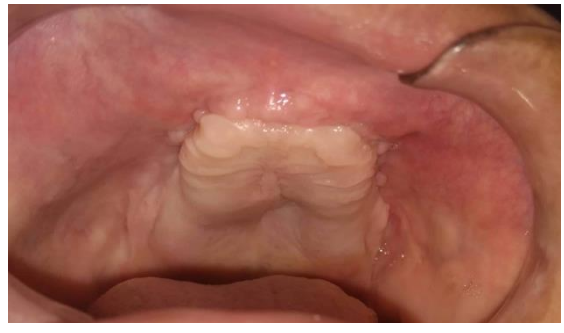

Figure 6. Postoperative healing

Figure 7. Upper removable prosthesis rehabilitation (Courtesy of Department of Prosthodontics)

Discussion

Osteomyelitis was a common disease before the advent of antibiotics. Osteomyelitis, once a dreaded condition, has lost its ability to instill fear predominantly due to advances in current resources for accurate diagnosis, surgical treatment, and potent antibiotic therapy resulting in better outcomes (Habib *et al*., 2016). The incidence of maxillary osteomyelitis is low as the bone has thin cortical plates associated with high degree of vascularity. Cranial bones are infrequently involved, but infection spread to surrounding structures can lead to cerebral abscess, encephalitis, or meningitis (Sasindran *et al*., 2015).

Osteomyelitis of maxilla was originally described by Rees in 1847. There has been another case reported with osteomyelitis of maxilla that led to meningitis by direct extension (Elangovan & Srinivasa, 2013). Osteomyelitis of the maxilla is more frequently seen in younger patients, infants, and at the age range of 40-60 years. Lesions are normally quite extended, often with undistinguished borders. The most common presenting signs and symptoms of maxillary osteomyelitis include fever, headache, facial pain and swelling, mucosal ulceration and necrosis. Progressive bony destruction and formation of sequestrations are hallmarks of osteomyelitis (Sharma *et al*., 2014).

Maxillary osteomyelitis can be classified based on the following causes; traumatic, rhinogenic, and odontogenic. Lew and Waldvoger (1997) classified osteomyelitis of the jaw into suppurative and non-suppurative, depending on its infectious character or hematogenous origin (Table 1).

Radiographically spotty osteolytic changes are more frequently observed in chronic osteomyelitis of the maxilla. Cross sectional imaging modalities such as computed tomography (CT) scanning and magnetic resonance imaging (MRI) are now considered standard for diagnosing osteomyelitis. That assists the surgeon to plan optimal surgical management, avoiding morbidity and complications to adjacent critical structures. CT is superior to MRI in detecting sequestrum, cloaca, involucre (Narsapur *et al*., 2016).

Skull base osteomyelitis represents a life-threatening complication of external ear canal infection. Malignant otitis externa usually begins as an external auditory canal infection followed by osteomyelitis of the temporal bone. Typically, skull base osteomyelitis infection begins in the external ear canal, but spread of infection from paranasal sinuses has also been occurred. Manifestations are severe otalgia, discharge, and cranial nerve palsy (VI, VII, VIII, IX, X, XI, XII). Currently, a cultured-based antibiotic therapy remains the main treatment option in skull base and temporal bone osteomyelitis (Fish *et al*., 2012). In this case, the patient also suffered complications of external ear infection and lost his sensorineural hearing loss and cranial nerve palsy, followed by osteomyelitis of floor of temporal fossa.

Systemic diseases like diabetes, anemia, and malnutrition that cause concomitant alteration in host defenses profoundly influence the course of osteomyelitis. Some studies have found that up to 68% of cases, associated with hyperglycemia secondary to uncontrolled diabetes (Gill & Pulcini, 2019). But, this patient did not have any previously known risk factors, including diabetes, malignancy irradiation, chemotherapy, autoimmune disease, steroid use, hepatitis or HIV infection.

Acinetobacter baumannii is an opportunistic pathogen that has become increasingly important over recent years as

a cause of nosocomial infections. *Acinetobacter baumannii* is associated with invasive infections, including osteomyelitis. It had a high degree of antimicrobial resistance, particularly to carbapenems. Few therapeutic options are available for treating pan-resistant strains, colistin and tigecycline has been used, but resistance to these options frequently emerges in clinical practice. In this case, patient was prescribed with co-amoxiclav and followed by doxycycline and this was already same according to sensitivity test.

Osteomyelitis of maxilla is difficult to treat because of anatomic and esthetics considerations. The goal of the treatment is eradication of the infection and restoration of the function. The treatments for the maxillary osteomyelitis range from a noninvasive approach to a more invasive radical treatment. The main treatment consists of stabilizing the systemic conditions of immuno-compromised patient, culture of the associated microorganism, sensitivity tests, image assessment to determine the lesion extension, empiric administration of the antimicrobial drugs to combat predominant microorganism, removal of septic foci such as teeth and bone sequestrations, debridement, decortications or resection according to the case (Burduk *et al*., 2010).

A combination of antibiotic treatment with surgery has shown to be effective in treating the condition. Wide spectrum antibiotic should be given initially as osteomyelitis usually present in association with an ulcer and soft tissue infection. Finally, proper antibiotic should be administered based on microbial examination of bone cultures (Nanwani *et al*., 2019). Surgical treatment involves removal of loose teeth and sequestrum, debridement, decortication, resection and reconstruction. Saucerization implies freeing of the upper cortical section to expose medullary cavity and debride necrotic tissues. Decortication implies removal of infected bone cortex. Resection is useful for low degree or refractory stages according to Cordeiro's classification (Table 2). Resection options for non-viable or infected bone of the maxilla can range from a simple excision to a radical maxillectomy (Pattnaik *et al*., 2017). The relapse rate can be as high as 20 %. The resultant maxillary defect can be managed with either prosthetic rehabilitation alone or reconstruction with subsequent dental or facial prosthesis (Sasindran *et al*., 2015).

Oxygen content of the tissue is crucial in the recovery from osteomyelitis. In addition to conventional methods such as intensive antibiotic therapy, surgical curettage, and drainage and removal of cause, hyperbaric oxygen (HBO) can beneficially influence the courses of persistent sinuses in chronic osteomyelitis. It is an adjunct in osteomyelitis therapy since it can elicit tissue hyper-oxygenation effect, antimicrobial activity, fibroblastic proliferation, neovascularization and bone matrix (Lentrodt *et al*., 2007).

Conclusion

Infection of the maxilla can cause serious complications for the patient such as infection of the cranial cavity and brain. Thus, it is essential that any maxillary osteomyelitis should be treated aggressively by the surgeon to avoid subsequent dreaded consequences. Timely treatment of this disease is paramount. Secondary osteomyelitis of the jaw due to odontogenic infections is now decreasing in prevalence due to the broad-spectrum antibiotic coverage during dental procedures. Despite this, it still remains a challenging clinical entity in developing countries.

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The authors declare there is no potential conflict of interest.

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