# Recurrent Chronic Maxillary Osteomyelitis in a Child: A Rare Presentation

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Dr Nneka Chukwumah Department of Preventive Dentistry University of Benin Teaching Hospital Benin City, Edo State Nigeria. Email: nnechukwumah@yahoo.com.cor **Objective** Osteomyelitis in the craniofacial region could result from untreated dental conditions such as pulpal or periodontal infections or by direct inoculation from trauma or surgery. Its development is influenced by a decreased host defensive mechanism and could result in major complications such as infection of the brain and cranium, making prompt and correct diagnosis of maxillary osteomyelitis crucial in its management. The objective of this study was to report a rare case of recurrent maxillary chronic osteomyelitis in an 11-year-old.

Case report: An 11-year-old boy who presented to the Paediatric Dental Clinic of the University of Benin Teaching Hospital with left sided facial swelling, purulent discharge from his left eye and left preauricular region of one-week duration. Past history revealed patient had been managed with similar symptoms at age 6 and had a partial maxillectomy done 5 years prior. Following clinical examination, radiological assessment (CT scan) and laboratory investigation (culture yielded growth of Staphylococcus arlettae) a diagnosis of recurrent chronic suppurating osteomyelitis was made. Patient was severely anaemic with underlying malnutrition and had to be optimized for surgery which was surgical debridement and drainage of abscess under general anaesthesia and patient placed on antibiotics and patient responded well to treatment.

**Conclusion:** Increased suspicion of rare disease conditions such as maxillary chronic osteomyelitis in children is important and more so a recurrent case as this. Management should be multidisciplinary and follow a holistic approach.

**Keywords:** Recurrent chronic osteomyelitis, maxilla/maxillary, child, Staphylococcus arlettae.

### INTRODUCTION

Infections of the mandibular and maxillary bones pose difficulties for dentists and maxillofacial surgeons during management despite recent advances in diagnosis, surgical management and antimicrobial therapy (Zimmerman et al, 2020). In developing countries, a resurgence has been reported and adduced to the poor socioeconomic conditions, antibiotic resistance and poor healthcare systems (Bienvenue et al, 2021). The term "osteomyelitis" refers to an inflammatory disease of the bone that starts as an infection of the medullary cavity and Haversian system of the cortex and progresses to affect the periosteum of the infected area (Borle, 2014). Osteomyelitis typically develops in the craniofacial region as a result of untreated dental conditions via simple contiguous spread such as pulpal or periodontal infections

or by direct inoculation from trauma or surgery (exodontia, oral mucosal wounds and maxillofacial fractures) (Lucidarme et al, 2022).

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The development of osteomyelitis is influenced by a decreased host defensive mechanism, which could be local or systemic. Local causes include trauma, radiation, or certain medications (Goldblatt et al, 2017), specific bacteria that cause infection are Staphylococcus aureus or Mycobacteria. Systemic conditions such as diabetes, cancer, autoimmune disorders, and malnutrition have

been linked to osteomyelitis (Miloro and Peterson, 2012), other causes include steroids, chemotherapeutic drugs, bisphosphonates, and other cytotoxic therapeutic drugs (Yavuz et al, 2008).

Osteomyelitis may be categorized as acute or chronic. Acute osteomyelitis refers to a bone inflammation as a result of previous trauma, puncture wound, surgery, bone fracture, abscessed tooth or infection of the soft tissue, ear or sinus. Chronic osteomyelitis usually occurs after an acute osteomyelitis episode which was not totally cured or properly managed. Cases with greater than 4 weeks duration from onset of symptoms could be termed primary or secondary. Primary chronic osteomyelitis usually of unknown origin is a nonbacterial chronic inflammatory disease and is linked to autoimmune disorders and syndromes (Stern and Ferguson, 2013), while the secondary chronic osteomyelitis of the jaw is usually a bacterial infection of dental origin from pulpal, periodontal diseases, post tooth extraction or foreign bodies (Eyrich et al, 2003).

The incidence of osteomyelitis has reduced significantly with proper oral hygiene and improved antibiotic therapy (Barry et al, 2007). Jaw osteomyelitis is notably uncommon (Kushner and Alpert, 2003) and especially so in the maxilla (Habib et al, 2016). Maxillary osteomyelitis is particularly rare in children (Wehl et al, 2019), as infants and young children in their first decade of life are least susceptible (Mandrekar et al, 2022). The rarity of maxillary osteomyelitis may be attributed to its high vascularity, thin cortical plate and relative lack of medullary tissue which reduces its ability to undergo necrosis (Pattnaik et al, 2017). However, when it occurs, the patient may experience major complications such as infection of the brain and cranium (Habib et al, 2016; Senel et al, 2007), making prompt and correct diagnosis of maxillary osteomyelitis crucial (Pattnaik et al, 2017).

The diagnosis of osteomyelitis can be made using the patient's history, findings on clinical examination, and radiographic and pathologic assessments (Senel et al, 2007). Clinical history could suggest the presence of odontogenic infections or reveal a history of poorly healed fractures from facial trauma (Topazian et al, 2002). In chronic osteomyelitis, there is an underlying history of acute osteomyelitis, with the presence of old or active fistulae with purulent secretion, poorly vascularized, pigmented, atrophic skin adhering to the bone and susceptible to ulceration. Pain on palpation and soft tissue induration may be evident (Topazian et al, 2002). Other clinical features reported include fever, swelling, trismus and pathologic fracture (Baltensperger and Eyrich, 2009). Radiographic features show a "Moth-eaten" appearance due to the enlargement of medullary spaces and widening of Volkmann's canals as a result of bone lysis and replacement with granulation tissue. Sequestra which are islands of dead bone and involucrum (reactionary subperiosteal deposition of new bone) may be seen (Ressy et al, 2014). On histology features seen include

necrotic bone with irregular aggregates of bony trabeculae with empty lacunae due to the absence of osteocytes, absence of osteoblastic lining, and chronic inflammatory cells such as lymphocytes (Poonia et al, 2016). Organisms commonly seen on microbiology test include Gram-positive Gram-negative and microorganisms such as Staphylococcus aureus, Staphylococcus epidermidis, Peptostreptococcus, Pneumococci, Haemolytic streptococci, Escherichia coli, and Bacteroides and as well as secondary fungi infections such Mucormycosis (Gupta et al, 2017).

Treatment of osteomyelitis is both medical and surgical, it also depends on the severity of the infection, time of presentation as well as underlying medical conditions. Procedures could include removal of aetiological factor such as an infected tooth, incision and drainage, sequestrectomy, saucerization, decortication, jaw resection, antibiotic therapy, and hyperbaric oxygen therapy (Barry et al, 2007). This article reports a case of recurrent chronic osteomyelitis of the maxilla in an 11year boy who presented to the Dental centre of the University of Benin Teaching Hospital (UBTH).

#### **CASE REPORT**

An 11-year-old boy presented to the Paediatric dental clinic of the University of Benin Teaching Hospital (UBTH) with left sided facial swelling, purulent discharge from his left eye and left preauricular region of one-week duration. Patient's father gave a history of extraction of a right maxillary tooth about 5 years ago in a primary health centre following complaints of pain and abscess from the tooth. After the extraction, the extraction site continued to discharge pus and was referred to the ear, nose and throat (ENT) specialist where a diagnosis of chronic suppurative sinusitis was made and a maxillary antrostomy performed on him. After a few months of unsuccessful treatments and continuous pus discharge, associated inability to open the mouth and weight loss, the patient was subsequently referred to the Oral and Maxillofacial Surgery clinic UBTH where he was diagnosed with chronic suppurating osteomyelitis and a right partial maxillectomy with surgical debridement was done. There was no history of any underlying medical condition such as diabetes, cancer, autoimmune disorders or any syndromes but a suspicion of parental neglect and possible malnutrition was noted.

On general examination, child appeared small for age, malnourished, lethargic, had fluffy hair, very pale sclera, and a surgical scar on the right side of his mid face. On extra-oral examination, there was an obvious facial asymmetry evident by a diffused swelling on the left side of the face with a discharging sinus anterior to the left ear and pus discharge from the left eye at presentation (figure 1). On palpation, there was mild tenderness over the left maxilla, submandibular lymph nodes were enlarged, palpable, mobile but not tender. His mouth opening was inadequate, with an interincisal distance of 14mm,

halitosis was perceived, with poor oral hygiene evident by accumulated plaque and calculus. There was pus discharging intraorally from a fistula between the oral and nasal cavity in relation anterior maxilla and also an area of exposed bone with pus discharge in the maxillary right and anterior oral vestibule (Figure 2). The maxillary incisors (11, 22) were also mobile but not tender with displacement of 11. There were multiple missing teeth (12,13,14,15,16,21) in the maxillary arch (Figure 3).

Computed tomography scan imaging revealed areas of sclerosis and lytic bone changes involving the maxilla and left zygomatic arch with hypodense collections in the maxillary sinuses and mastoid air cells (Figure 4). A diagnosis of recurrent chronic suppurative osteomyelitis of the maxilla was made.

Laboratory investigation revealed patient was negative for retroviral disease, had AA-genotype (not a sickle-cell patient) to rule out other underlying medical conditions. Blood electrolytes and urea were within normal range; however, his blood panel showed a hemoglobin concentration of 2.6g/dl. Microscopy culture and sensitivity result yielded growth of *Staphylococcus arlettae* which was resistant to Clindamycin and Tetracycline (Figure 5).

The patient was immediately admitted and comanaged by the pediatricians, pediatric dentists and the oral and maxillofacial surgeons. Patient was optimized through blood transfusion (on account of severe anaemia), high protein-energy diet, proper oral hygiene. Considering the age of the patient, level of bone destruction and the previous partial maxillectomy a more conservative surgical approach was decided. Surgical debridement and drainage of abscess was done under general anaesthesia after patient was fully optimized for surgery. Surgical access was through an upper vestibular approach to drain right temporal abscess collection, remove bony sequestrum with copious saline irrigation. Packing of the surgical defect with ribbon gauze impregnated with tincture of benzoin compound (TBC) was done and vestibular mucosa restored with local intra-oral flap. Intravenous Clindamycin and Metronidazole were administered prophylactically but changed to intravenous Cefuroxime/Sulbactam and Metronidazole following the result of the microscopy culture and sensitivity test. Patient was converted to oral cefixime clavulanic acid on discharge. Patient has been closely monitored after surgery as shown in clinical photographs (Figure 6, 7 and





Figure 1. A clinical photograph showing pus discharge from the left eye with a discharging sinus anterior to the left ear. Discharging sinus shown by arrow.





Figure 2. Clinical photographs showing pus discharging intraorally from a fistula between the oral and nasal cavity in relation anterior maxilla as well as around the maxillary incisors and also an area of exposed bone in the maxillary right and anterior oral vestibule. Also shown is the inadequate mouth opening with an interincisal distance of 14mm.



Figure 3: Clinical intraoral photograph showing maxillary incisors (11, 22) which were mobile There were multiple missing teeth (12,13,14,15,16,21) in the maxillary arch.

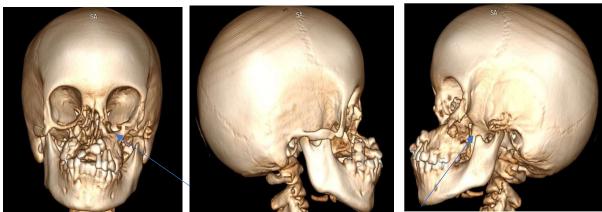


Figure 4: A 3-dimensional computed tomography which shows osteolytic changes in the midface extending from the alveolus below to the infraorbital rim and nasion above, unerupted and displaced tooth buds, malocclusion and denuded roots of maxillary teeth. The mid-face is retruded with absence of teeth in the maxilla. There is also a bony defect from previous right partial maxillectomy. The left side of the head show osteolytic changes in the midface and absence of the left zygomatic arch.

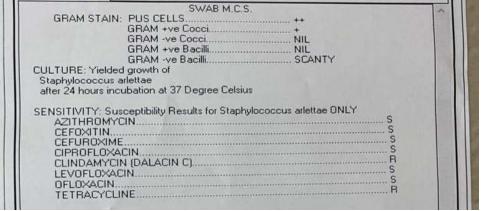


Figure 5: Microbiology Culture & Sensitivity test yielded Staphylococcus arlettae



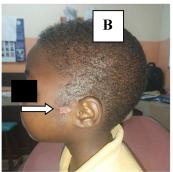




Figure 6: Two-weeks post operative photograph of patient. Healing left preauricular sinus (Arrow 9B).

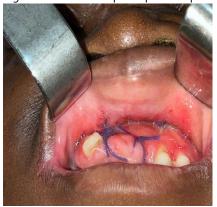




Figure 7: Two weeks post-operative intraoral radiograph which shows the repair of the intra-oral fistula and vestibules. Oral prophylaxis and hygiene were also ensured.



Figure 8: Two months post-operative photograph showing resolved abscess, healed operation scars and deviation of face to the left from previous maxillectomy.

## **DISCUSSION**

In the post-antibiotic age, osteomyelitis of the jaw, and particularly of the maxilla, is a rare occurrence (Habib et al, 2016), and even less common in children. However, osteomyelitis of the jaws is more common in developing countries when compared to developed countries (Kumar et al, 2013). This may due to the poor access to basic and

specialized oral healthcare (Kumar et al, 2013), and the low socioeconomic status of many patients in developing countries such ours have resulted in poor knowledge of oral health and lack of the financial means hence fail to seek medical attention early or even when needed. Exodontia is an established risk factor for the

Exodontia is an established risk factor for the development of osteomyelitis of the jaws (Bathoorn et al,

2013). This patient had an extraction for an abscessed tooth 5 years prior to presentation at a primary healthcare facility. Odontogenic infections are the main causes of chronic suppurative osteomyelitis (CSO) of the maxillofacial region (Kumar et al, 2013), and could develop as a complication of tooth extractions, poorly managed facial trauma/fracture and radiation to the jaws (Kumar et al, 2013). Certain predisposing factors have been associated with chronic osteomyelitis, which include; Diabetes, cancer, autoimmune disorders and malnutrition (Miloro and Petersen, 2012). Our patient was severely anaemic and malnourished at presentation. These underlying conditions would have contributed ultimately in reducing the supply of oxygen and nutrients required for healing to the marrow space. In addition, the transport of the body's blood-borne immune system to the marrow space is impaired (Hupp et al, 2009). The history of child neglect and malnourishment at presentation, probably explains the severity of the infection and its added complication of difficulty feeding caused by trismus and dental infection.

The clinical features of suppurative chronic osteomyelitis are localized pain, soreness, fever, painful or painless swelling, purulent discharge, intraoral fistula, cutaneous fistula, trismus, hypoesthesia of the inferior dental nerve, and pathologic fracture (Gudmundsson et al, 2017). This patient had a trismus, with exposed intraoral maxillary bone with pus discharges from oral and preauricular cutaneous fistula. Classic radiographic features include "Moth-eaten" appearance and widening of Volkmann's canals with sequestra and involucrum seen occasionally (Reddy et al, 2014). The patient's computed tomography scan imaging revealed areas of osteolytic changes in the midface extending from the alveolus below to the infraorbital rim and nasion on the left and absence of the left zygomatic arch. There were unerupted and displaced tooth buds, malocclusion and denuded roots of maxillary teeth. The mid-face was retruded with absence of some maxillary teeth. There was also a bony defect from previous right partial maxillectomy.

Both gram-positive and gram-negative organisms have been implicated it the microbiological aetiopathogenesis of osteomyelitis (Gupta et al, 2017). Interestingly, Staphylococcus arlettae was the only organism cultured from the specimen. It is a Coagulase Negative Staphylococci (CoNS) and although, not a common cause of osteomyelitis, is now becoming more widely acknowledged as a significant source of infections in both humans and animals (Lavecchia et al, 2019). There is also very little clinical relevance and annotation of genes that might be connected to pathogenicity and/or antibiotic resistance in the CoNS species Staphylococcus arlettae (Lavecchia et al, 2019). The blaZ gene was the only known -lactamase gene in staphylococci until recently when further putative -lactamases were discovered, one of which was a brand-new functional -lactamase encoded by blaARL in Staphylococcus arlettae, which interestingly

limits its therapeutic possibilities (Andreis et al, 2017). This explains the fact that coagulase-negative staphylococci (CoNS), which make up a sizable portion of the skin's and mucosa's microbiota in warm-blooded hosts, are also known to be classical opportunistic pathogens and a major source of a number of diseases linked to healthcare (Asaad et al, 2016). More research in the area of identifying microbiological agents involved the aetiology of chronic osteomyelitis especially in children is recommended as well as, an understanding of the mechanisms of action concerning treatment drug options. This patient's antibiotic sensitivity test was resistant some antibiotics commonly used for treating osteomyelitis such as clindamycin, penicillin and tetracycline.

A multidisciplinary approach may be required as found in this case with the involvement of the paediatric dentists, paediatricians, oral and maxillofacial surgeons, oral pathologists, radiologists and social workers. Chronic osteomyelitis is managed medically and surgically by use of long-term antibiotic medication and surgical techniques such as debridement of necrotic tissue, extraction the affected teeth, decortication, sequestrectomy, and saucerization (Gudmundsson et al 2017). There is no widely accepted duration of antibiotic cover in the treatment of paediatric osteomyelitis, however, three to six weeks of antibiotic therapy and an early changeover to oral medication have only largely been suggested (Wehl et al, 2019). In order to prevent post-operative infection in our patient and considering it was a recurrent condition, antibiotic medication was started intravenously before the surgery and continued afterward for 6weeks. The patient is still being closely monitored.

## CONCLUSION

Chronic maxillary osteomyelitis though uncommon in children may be on the rise, with the increasing rate of poverty and malnutrition, its social predisposing factors and consequent implications should be a consideration in patients presenting with recurrent abscesses. A multidisciplinary and holistic approach to management is recommended.

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None

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