Case Report

A case report of chronic intermedullary inflammation of bone in a child

ABSTRACT

Osteolytic lesions refer to the destruction of any part of bone due to a disease process (pertaining to dissolution of bone, especially loss of calcium). Osteomyelitis is the inflammation of the bone caused by an infecting organism. Although bone is normally resilient to bacterial colonization, events such as trauma, surgery, infections, the presence of foreign bodies, and anemia may disrupt bony integrity and lead to the onset of bone infections. Sometimes, osteomyelitis causes no signs and symptoms or the signs and symptoms are hard to distinguish from other problems. This may be true for children, older adults, and people whose immune systems are compromised. Here, we report a case of chronic intermedullary inflammation of bone in a child.

Keywords: Dental caries, inflammation, intermedullary, osteolysis, osteomyelitis, pathological fracture, suppuration

INTRODUCTION

Osteolysis means the destruction of bone due to disease process, pertaining to the dissolution of bone especially loss of calcium from bone. Osteolysis may be caused by bone pathologies like tumors, cysts, or chronic inflammation. Among these, osteomyelitis (OM) of jaw is a frequently occurring osteolytic infections in dental practice, especially increased in old age, rural, and developing countries because of inadequate access to oral healthcare professionals. Based on the clinical onset and presence of suppuration, osteomyelitis can be classified as acute or chronic and suppurative or non-suppurative. The predisposing factors for osteomyelitis include radiotherapy (ORN), uncontrolled diabetes, malnourishment and patients on immunosuppressive therapy, complication of extractions, etc.[1,2]

In children, osteomyelitis can be due to improper antibiotic use, poor oral hygiene, malnutrition, radiation therapy, traumatic injuries, virulence of microorganisms, etc.[2]

The diagnosis of osteomyelitis (OM) is based on the history of the patient, clinical examinations, radiographic, and surgical findings. Histopathological examination aids in the confirmatory diagnosis of osteomyelitis. Here we report a case of an extensive osteolytic lesion of the right mandible in a 7-year-old child, which was diagnosed as chronic suppurative osteomyelitis.

CASE REPORT

A 7-year-old female patient was reported with a chief complaint of pain and swelling over the right lower jaw

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region for the past 11/2–2 months and also pus discharge from the lower back tooth region. The patient had consulted a nearby dentist for the same and was prescribed medication. However, pain and pus discharge were not noted at the time of examination.

The extraoral examination revealed facial asymmetry due to a diffuse swelling over the right side lower half of the face extending superiorly 3 cm below the lower eyelid, inferiorly 2 cm below the body of the mandible, medially to the lateral border of the nose, and laterally to the angle of the mandible [Figure 1a]. Multiple cervical lymph nodes were palpable. On general examination, the patient was poorly built and malnourished. The patient had restricted mouth opening. The intraoral examination revealed, dental caries in relation to 75, 85 with tenderness on percussion irt 85. The right buccal vestibule was partly obliterated due to swelling, and there was no evidence of pus discharge noted [Figure 1b]. The patient was diagnosed as dentoalveolar abscess irt 85 and was also advised for radiographic examination.

Orthopantomogram (OPG) revealed an extensive hazy moth-eaten appearance in the body, ramus, right subcondylar, and sigmoid region. The lower border of the mandible was discontinuous irt 85, 46 regions, and evidence of root resorption was seen in relation to 85 [Figure 2].

Computed tomography (CT) revealed a large ill-defined osteolytic lesion with bony destruction in the body and ramus, body, condylar, and coronoid process. A floating tooth is seen in the body along with extensive destruction of buccal and lingual cortices. Based on clinical and radiographic findings, a provisional diagnosis of osteomyelitis of right hemi-mandible with bony destruction was given [Figure 3].

Routine blood investigations revealed an elevated ESR, elevated neutrophils percentage, reduced hemoglobin, decreased serum ferritin levels, marginally reduced serum calcium level, and negative for HbsAg and HIV. A Mantoux test was done to rule out tuberculosis, which was found to be negative. The routine urine investigation and renal function test were revealed within normal limits. Liver function test shows normal findings except for slightly elevated alkaline phosphatase level (214).

The patient was subjected for an incisional biopsy under general anesthesia as she was not co-operative. Multiple soft and hard tissue specimens along with carious 85 were submitted for histopathology. Smear from the subjective area was submitted to microbiology for microbiological investigations which was reported negative for acid fast

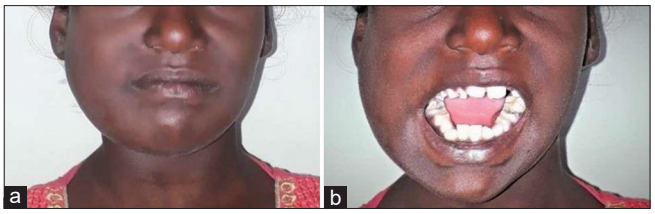

Figure 1: (a) Image showing extraoral swelling of right side of the face (b) Intraoral features showing obliterated buccal vestibule with restricted mouth opening and dental caries in relation to 75,85

Figure 2: Orthopantomogram showing hazy moth-eaten appearance in body, ramus right subcondylar region

Figure 3: Computed tomography showing ill-defined osteolytic lesion with bony destruction

bacilli and mixed microbial colonies in Grams stain along with degenerating blood cells.

The incisional biopsy revealed a fibrocellular connective tissue stroma infiltrated with chronic inflammatory cells like lymphocytes, plasma cells, neutrophils, eosinophils, macrophages, and foam cells. Also, numerous endothelium-lined capillaries of varying sizes along with extravasated RBC's and areas of hemorrhage were seen. Two eosinophilic areas, resembling osteoid lined by osteoblasts, are seen. Bits of fragmented ones are also seen with osteocytes present in it. Few areas of the section revealed

spindle cells with wavy nuclei resembling nerve fibers. So, a diagnosis of chronic inflammation with respect to the received biopsy specimen was given [Figure 4].

Hemi-mandibulectomy of the right mandible was performed under GA and submitted for histopathology. The hemi-mandibulectomy specimen with buccal cortical plate resorbed and sequestered revealed a necrotic and reddish brown inflammatory tissue on the surface was also submitted. The ramus seems to be resorbed extensively, whereas the condylar head and coronoid process were submitted as separated bits, as there has been a pathological fracture due to osteolysis [Figure 5].

Hematoxylin and eosin stained section reveals areas of delicate stroma with numerous capillaries and extravasated RBCs, mixed inflammatory cell infiltration comprising numerous plasma cells and also lymphocytes, neutrophils, eosinophils, macrophages, and few mast cells. Some areas of bone seem to be lamellar with osteocytes within lacunae, while many areas of bone show irregular margins due to resorption with multinucleated and mononucleated giant cells. Most of the marrow spaces show numerous blood vessels of varying sizes within a delicate connective tissue stroma along with inflammatory cells like lymphocytes, plasma cells, and acute inflammatory cells especially neutrophils and eosinophils [Figure 6].

As the soft tissue sections from certain parts of the specimen revealed chiefly the medullary areas predominantly infiltrated with plasma cells and eosinophilic leukocytes, the patient was referred for serum M protein and also sections were submitted for immunohistochemical (IHC) analysis to rule out plasmacytoma and eosinophilic granuloma.

IHC reports revealed negative for plasmacytoma and eosinophilic granuloma (negative for S-100, CD-1a, kappa and lambda chains) [Figure 7]; hence, on clinico-pathological correlation, a histopathological diagnosis of chronic suppurative osteomyelitis of right mandible has been considered.

DISCUSSION

The term osteomyelitis originated from the ancient Greek words, osteon (bone) and muelinos (marrow), which refers to an inflammatory condition of bone and bone marrow that develops usually after a chronic infection. In the oral and maxillofacial region, osteomyelitis is more common in the mandible than in the maxilla because of the dense, poorly vascularized cortical plate, and single blood supply from the neurovascular bundle.[2,3]

Based on the onset of infection, osteomyelitis of the jaw can be classified as acute or chronic and suppurative or non-suppurative based on the presence or absence of pus discharge. In its acute stage, suppurative OM of the mandible is distinguished by deep-seated intense pain, high intermittent fever, mental nerve paresthesia, and a clearly identifiable cause.[3,4]

The chronic osteomyelitis (OM) can be focal or diffuse sclerosing OM and Garres OM. Chronic diffuse sclerosing OM is mainly reported in the mandible, which is characterized by the long history of recurrent pain and inflammation without suppuration and usually arises as a complication of tooth extraction in adults. In the maxillofacial region, OM is more common in fifth and sixth decades of life and has been associated with a systemic disease.[3]

The term chronic OM is used for primary or secondary cases with a duration of more than 4 weeks from the onset of symptoms. The primary chronic OM is a non-bacterial chronic inflammatory disease of unknown etiology and associated with other systemic conditions such as SAPHO syndrome and Majeed syndrome. Secondary chronic OM (SCO) of the jaw is usually caused by bacterial infection of dental origin (pulpal disease, post-tooth extraction, or foreign bodies).[3,5]

The main cause of chronic suppurative OM of the jaw is odontogenic infections which might occur as a complication of dental infections. The various organisms causing OM are *Staphylococcus aureus*, alpha and beta hemolytic

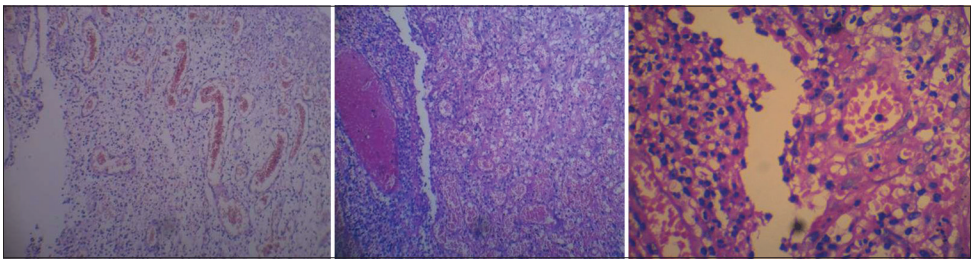

Figure 4: Histopathology of incisional biopsy specimen—hematoxylin and eosin stained tissue section showing fibrocellular connective tissue stroma with numerous blood capillaries and inflammatory cells, osteoid lined by osteoblasts

Figure 5: Excisional biopsy showing hemi-mandibulectomy specimen of right mandible

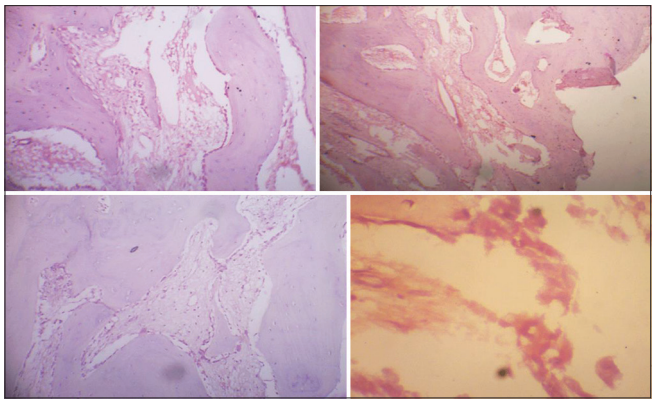

Figure 6: Histopathology of excisional biopsy specimen, showing decalcified bone with osteocytes within lacunae, bone resorption with scalloped margins, multinucleated giant cells adjacent to resorption areas

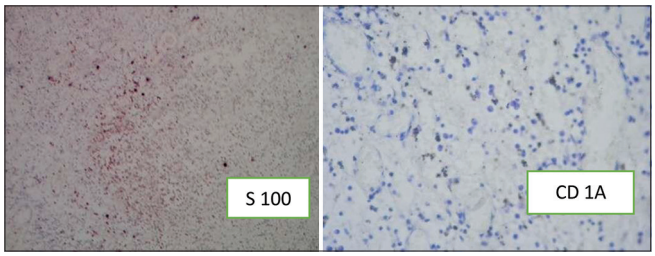

Figure 7: Immunohistochemistry analysis for S 100 and CD1a (negative)

streptococci, *Peptostreptococci,* and *fusiform* bacteria. Initially, the infection involves the medullary tissue that promotes suppurative infection, hyperplasia, and resorption.[6] The disease becomes recognized in the calcified portion of the bone leading to resorption, and the pus in the medullary cavity or beneath the periosteum leads to an impediment of blood supply. The predisposing factors are exposure to radiation, immune-compromised status, uncontrolled diabetes, and poor socioeconomic status which includes anemia.[7,8]

Iron is the most biologically essential element for living. Body requires iron for oxygen transport by hemoglobin and myoglobin for the proper functioning of many cells and tissues. Low iron status can lead to decreased physical activity and increased susceptibility to infection.[9,10] Iron deficiency anemia is most commonly seen in pre-school age children, pregnancy mainly in low socioeconomic status. Iron plays a vital role in bone mechanism, through Vitamin D activation and deactivation, also acts as a co-factor for the hydroxylation of prolyl and lysil residues of procollagen.[10,11]

The panoramic radiographic features of osteomyelitis include thickened lamina dura, lytic destruction breaching buccal or lingual cortices, radiolucent areas, bony destruction, and sequestrum formation (onion skin or moth-eaten appearance). Computed tomography shows increased sclerosis of trabecular bone, widening of the mandibular foramen, defective thickening of cortical plate, periosteal reaction, sequestrum, and narrowing of bone width. Bone scintigraphy used when multifocal systemic disease is suspected. In the present case, extensive destruction of cortices, floating tooth appearance, and hazy moth-eaten appearance was noticed, so a diagnosis of osteomyelitis of the right mandible was given.[12]

In cases of osteomyelitis, necrosis occurs once ischemia sets in. The proteolytic enzymes are liberated with the destruction of bacteria along with vascular thrombosis and ischemia leading to necrosis and suppuration. When pus accumulates, the intramedullary pressure is increased, which results in vascular collapse, venous stasis, and further ischemia. When the pus accumulates underneath the periosteum, it is elevated from the cortex and further reduces the vascular supply to the bone. As this continues to accumulate, the periosteum is breached and followed by the development of mucosal or cutaneous abscesses and fistula.[13-15]

In our present case, dental caries in relation to 85 led to the development of periapical infection that further progressed to the development of chronic osteomyelitis with extensive resorption of buccal cortex, ramus of mandible, and separation of coronoid and condyle causing pathological fracture with an iron deficiency status. Though our case was clinically and provisionally diagnosed as osteomyelitis with a differential diagnosis of eosinophilic granuloma, a careful multifactorial approach had to be followed to arrive at a diagnosis of chronic suppurative osteomyelitis.

Eosinophilic granuloma (EG) is a destructive bone lesion arising from the clonal proliferation of Langerhans cells and difficult to make a correct diagnosis without a pathological evaluation. In the head and neck region, EG is frequently found to affect the mandible, soft tissue adjacent to the involved bone. In EG of jaws, destruction of alveolar bone is thought to be one of the characteristic signs, and in this location, the disease may stimulate severe localized periodontitis or

periapical infection. These lead to malocclusion of teeth, purulent discharge from lesions, pathological fractures, etc. The area of the destroyed bone is replaced by soft tissue (reddish brown in color), and over time, lesions become fibrous and greyish. The other symptoms include localized swelling, pain, and low-grade fever along with elevated ESR and leukocytosis.[16,17]

The clinical and radiographical examinations of EG are not specific and can be easily misdiagnosed as osteomyelitis, odontogenic cyst, bone cyst/tumors, or lymphoma. So, histopathology involving immunohistochemistry using various antibodies is used for confirmatory diagnosis. EG shows positivity for S-100, CD-1a and CD-207, and negative for CD-45. In our case, the sections were negative for both S-100 and CD-1a; hence, eosinophilic granuloma and plasmacytoma were ruled out.

Histopathologically, decalcified bone revealed lamellar bony areas with osteocytes within lacunae. Some areas of bone margin seem to be scalloped showing resorption and few mononucleated and multinucleated giant cells seen adjacent to the resorption areas. Most of the marrow spaces show numerous blood vessels of varying sizes within a delicate stroma along with inflammatory cells like lymphocytes and more of plasma cells. In few areas, more of eosinophils are also seen. On correlating with clinical and radiographic findings, a histopathological diagnosis of chronic suppurative osteomyelitis was given.

However, the disease is completely curable if treated early with a judicious use of antibiotics and surgical intervention, thus emphasizing the fact that well-excised and timely treatment plans will have a good prognosis and healing rate. Also proper treatment regimen for anemia and prevention will improve the quality of life and bone health.

CONCLUSION

As our case was provisionally diagnosed as osteomyelitis with a differential diagnosis of eosinophilic granuloma, a very careful method of approach histopathologically had to be followed to arrive at a diagnosis of chronic suppurative osteomyelitis because of extensive osteolysis of the mandible in a child without presenting typical features of an extra/intraoral sinus discharging frank pus. Additionally, iron-bone relationship has to be emphasized as the bone metabolism in anemia is not fully addressed, as prolonged or untreated iron deficiency could result in severe bone loss and increased risk of osteolysis.

Though diagnosing osteomyelitis is a difficult challenge due to the vast variation in clinical presentation, early diagnosis is a key to successful treatment and histopathology plays a critical supportive role in final diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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