Osteomyelitis as a Complication to Dental Extraction in Osteopetrotic Bone: A Case Report

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Abstract
Osteopetrosis of bone is a genetic disorder which is characterized by the generalized sclerosis of bone due to a defect in bone remodeling. The ensuing dense bone, with its minimal bone marrow spaces and limited blood supply, often predisposes to the development of osteomyelitis following trauma (like a tooth extraction) or infection. We have documented a case report in which a 39 years old male patient with undiagnosed osteopetrosis had undergone a dental extraction resulting in osteomyelitis of mandible.

Keywords: Osteomyelitis, Osteopetrosis.

Introduction:
Osteopetrosis (OP) (osteo means bone & petros means stone) or commonly called as Marble bone disease (dense rock like appearance) is also known as Fragalis Generalisata (affecting all bones of the body) or Albers-Schonberg disease, after the name of scientist who reported it.² It is a rare genetically inherited metabolic bone disorder in which the cancellous bone is replaced by dense cortical bone due to defective osteoclastic activity & remodeling.³ The prevalence rate of OP is 1 in 1,00,000-5,00,000.⁴ It can occur both in benign & malignant forms and can express as autosomal dominant or recessive genetic disorder.⁵ Osteomyelitis is the most serious complication of osteopetrotic bone.⁶

This article presents a case report in which a routine dental extraction has resulted in osteomyelitis of jaw in osteopetrotic bone.

Case Report:
A 39 years old male patient had reported to Department of Oral & Maxillofacial Surgery, Himachal Institute of Dental Sciences, Paonta Sahib, Himachal Pradesh (HP), with a chief complaint of draining sinus tract and grossly decayed teeth in the left lower back tooth region since four years. Patient revealed a history of dental extraction in the same region about four years back, which was followed by recurrent drainage of pus. Patient was advised antibiotics for the same but the draining pus kept on draining occasionally. Radiographic examination including Intraoral Periapical Radiograph (IOPAR) & Orthopantomograph (OPG) were advised which revealed an altered bony architecture in which all the cancellous bone was replaced by cortical bone. Computerized Tomography (CT scan) was advised for further evaluation. The CT scan reports revealed a generalized & homogenous increase in bone density with a hypodense area in the left first molar region of mandible suggestive of bone sequestrum.

Patient also had an undiagnosed generalized bone deformity with bowing of extremities and crippling during walking. Based on the clinical & radiographic interpretations, the patient was diagnosed with osteopetrosis in which the previous dentist had performed a dental extraction without establishing a
diagnosis of Osteopetrosis thereby resulting in osteomyelitis, a well-known complication of Osteopetrosis.

As the facility of hyperbaric oxygen therapy was not available in our setup and the patient was reluctant to go for aggressive surgical intervention, a conservative treatment plan was established. Systemic antibiotics were advised after obtaining the culture & sensitivity and root canal treatment was advised for the decayed tooth lying adjacently.

**Discussion:**

Osteopetrosis, first reported by Albers-Schonberg in 1904, is an extremely rare genetic metabolic bone disease characterized by a generalized increase in skeletal mass. This inherited disorder results from the congenital defect in the development or function of osteoclasts.

Osteopetrosis is classified into 4 types as: 1) **Severe infantile** - malignant type: associated with mutation of TCIRG & C1CN7 genes. Usually diagnosed in 1st year after birth and the patient do not survive for more than 20 years.

2) **Osteopetrosis with renal tubular acidosis & cerebral calcification**: associated with deficiency of carbonic anhydrase (CA) II and mutation in the gene encoding for CA II protein. This type of osteopetrosis is usually seen in early childhood but spontaneous regression may be seen.

3) **Benign autosomal dominant type**: the causative gene is still not identified. The patient usually does not present with any symptoms and is diagnosed radiographically.

4) **Intermediate mild autosomal recessive type**: may be asymptomatic or may present features of infantile type.

The patient who reported to us belonged to the third category, benign autosomal dominant type as he was diagnosed accidently on routine radiographs. However, the patient had bowing of extremities and crippling during walking. The disease represents a spectrum of clinical variants because of the heterogeneity of genetic defects resulting in osteoclastic dysfunction.

Patient is liable to have multiple orthopaedic problems like bone pain, long bones bowing, degenerative arthritis & coxa vora. Other problems include anaemia, bilateral cranial nerve palsies. The failure of bone resorption and remodeling may lead to progressive entrapment and compression of the various cranial nerves. This may lead to facial pain and paraesthesia. The pain characteristics are similar to those of paroxysmal trigeminal neuralgia (PTN). Our patient had bowing of long bones and crippling during walking.

Dental abnormalities include increased susceptibility to caries, constriction of neurovascular canals along with obliteration of marrow cavities and the pulp chambers & bone necrosis leading to osteomyelitis. There may be delayed eruption, early loss of teeth, enamel hypoplasia, malformed crowns and roots & thickening of lamina dura. Our patient had constriction of neurovascular canals & obliteration of pulp chamber of almost all teeth, grossly carious maxillary left second molar and proximal caries in maxillary left lateral incisor and mandibular left first molar.

Radiographically, it is seen as uniform increase in bone density without corticomедullary demarcation. The long bones have a white chalky appearance. They have an “Erlenmeyer Flask” deformity at their ends due to failure of metaphyseal remodeling. The vertebral column has a sandwich or rugger jersey appearance. Spondylosis of lumbar spine has been reported. A bone within bone or endobone phenomenon has been seen in small bones of hands. There is also an increase in the density of skull base particularly the anterior cranial fossa. Other complications include stunted growth, osteosarcoma, and pathological fractures. The most common site for fracture is the femoral shaft. Other location includes the inferior neck of femur and posterior tibia. The fracture healing occurs at normal rate. Our patient did not give any history of fractures.
Osteomyelitis in Osteopetrotic Bone

Case Report

Figure No. 1: Patient Clinical Picture

Figure No. 2: Draining Sinus Tract with Respect to Previous Extraction Site Present

Figure No. 3: OPG Showing an Altered Bony Architecture
Management of osteopetrosis patient requires a comprehensive approach to the characteristics clinical problems. Medical management includes efforts to induce the host mesenchymal cells to differentiate into normal osteoclast cells. Host osteoclasts stimulation has been tried using calcium restriction, calcitriol, steroids, parathyroid harmone & interferon.4

Dental treatment for patients with osteopetrosis, especially extraction of mandibular teeth, must be undertaken with great caution. As the patient is more susceptible to caries a prophylactic treatment should be considered.5

Osteopetrosis complicated by osteomyelitis can be treated by hyperbaric oxygen therapy, debridement, sequestrectomy or segmental resection followed by reconstruction. Bone marrow transplantation seems to be the only permanent cure for Osteopetrosis with limited access (45% success rate).4 Further long term studies are required to understand & manage the Osteopetrosis.

Our patient was treated with systemic antibiotics and root canal treatment was done of mandibular left first molar. All other carious tooth were restored with fluoride releasing Glass ionomer cement. On follow up there was no draining pus & the sinus tract started to heal. As the patient is susceptible to caries patient was encouraged to use fluoride containing mouth washes and tooth paste and he was advised for a routine six month dental checkup.

Conclusion:
Thus, we conclude the importance of correct diagnosis while tooth extraction especially in osteopetrotic bone which can be easily diagnosed via its peculiar radiographic features which if neglected can lead to severe complications like osteomyelitis.

References:


