

A unique case of multiple osteochondroma: Mandibular symphysis and femur

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Anuradha Navaneetham, K. Arati Rao¹, Satish Kumaran², Hitesh H. Baweja²

Hosmat Dental Implantology and Facial Aesthetic Center, 45, Magrath Road, ¹M R Ambedkar Dental College, Bangalore, Karnataka, ²Institute of Dental Sciences, Bareilly, Uttar Pradesh, India

Address for Correspondence:

Dr. Anuradha Navaneetham, # 7, Cornwell Road, Langford Garden, Bangalore - 560025, India.

E-mail: anufaxmax@gmail.com

ABSTRACT

Osteochondroma is a common benign tumor of the axial skeleton rarely seen in the facial bones. When encountered in the facial skeleton, it is commonly found in the mandible, usually in the condyle or coronoid processes. There are only two earlier reported cases of the tumor in the mandibular symphysis. We report an unusual case of osteochondroma of the mandibular symphysis found in conjunction with osteochondroma of the distal femur and, stress the need for long-term follow-up in patients with multiple lesions.

Key words: Mandibular symphysis, osteochondroma, osteochondromatosis

INTRODUCTION

Osteochondroma or osteocartilaginous exostosis is a common benign tumor of the axial skeleton not commonly seen in the facial skeleton because the facial bones exhibit intramembranous ossification whereas the tumor usually appears in endochondral bones. It consists of a bony outgrowth covered by a cartilage cap. It is a slow growing tumor, which parallels the skeletal growth of the individual. In the facial skeleton, it is most frequently seen in the condyle and coronoid processes. There have been only 2 previous reports of osteochondroma arising from the mandibular symphysis region.^[1,2] When multiple osteochondromas are present, they are usually seen in long bones. We report a unique case of osteochondroma of the mandibular symphysis found in conjunction with osteochondroma of the distal femur.

CASE REPORT

A 13-year-old Nigerian boy was referred to our unit by the orthopedic department for evaluation of a hard swelling on the right chin region. The patient was found to have a bony hard swelling on his left thigh region. A plain radiograph showed a bony exostosis near the distal femur (Figures 1a and b). On examination of the facial region, a painless bony hard swelling

was found on the right submental region. The patient had noticed these swellings one year back. A three-dimensional computed tomography (CT) showed a sessile bony protuberance of the right symphysis region [Figure 2]. Detailed case history revealed no other family members to have similar bony outgrowths. Due to the age of the patient and site of the swellings, a clinical diagnosis of osteochondroma was made. Simultaneous excision of both the tumors was done under general anesthesia by orthopedic and maxillofacial surgeons. The tumor at the symphysis region was exposed through an intraoral approach [Figure 3]. The tumor was excised using surgical burs and chisels [Figures 4 and 5]. The tumor measured 15 mm in length and 10 mm in diameter [Figure 6]. The tumor from the femur region measured 35 mm in length and 30 mm in diameter. Histopathological examination [Figure 7] of both tumors confirmed the clinical diagnosis of osteochondroma. Patient was free of recurrence till 6 months after surgery.

DISCUSSION

Osteochondroma is a slow growing benign tumor arising from the cortex of the bone and contains a prominent cap of cartilaginous tissue. The tumor may be sessile or pedunculated and often have an elongated shape.^[3]

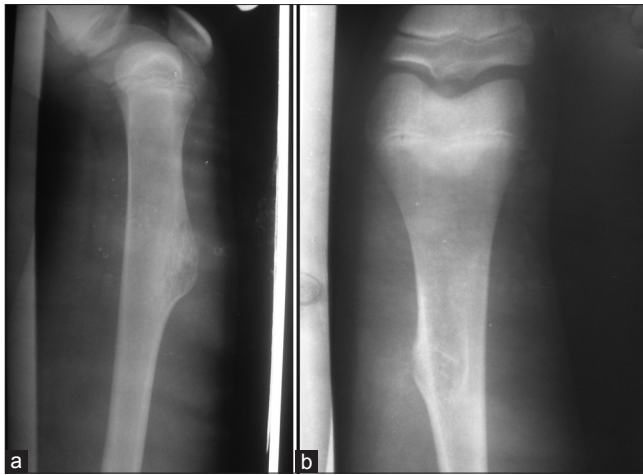


Figure 1: (a and b) Plain radiographs showing tumor on Lt. femur

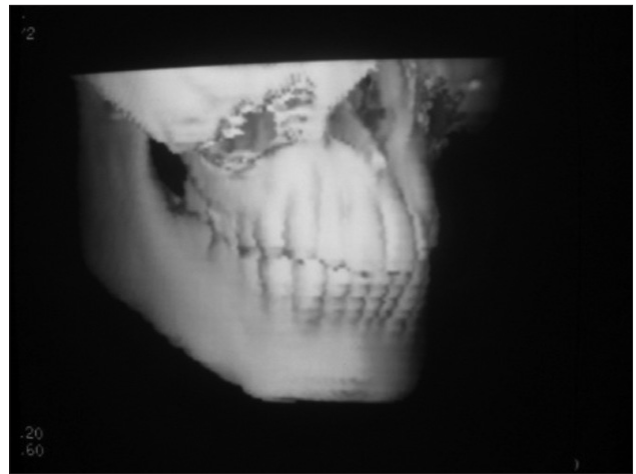


Figure 2: Three-dimensional CT showing tumor on Rt. symphysis

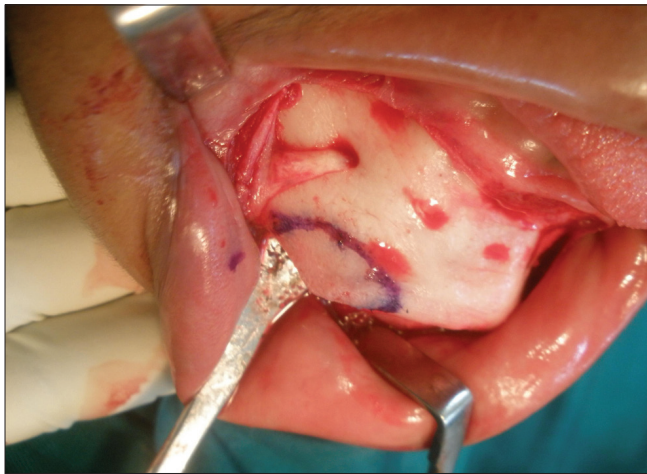


Figure 3: Intraoperative view of the tumor on Rt. symphysis region

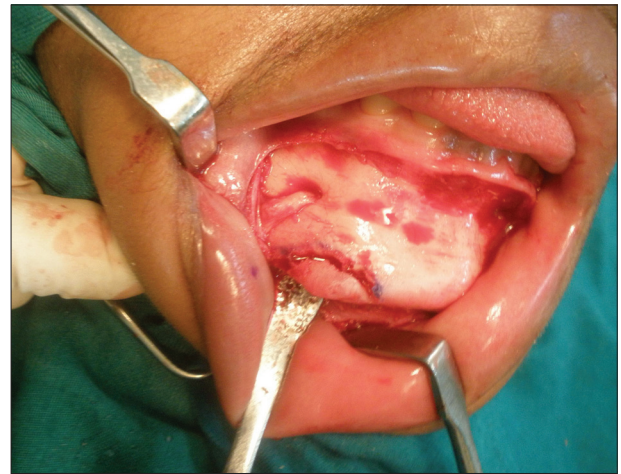


Figure 4: Osteotomy of tumor



Figure 5: Excised specimen

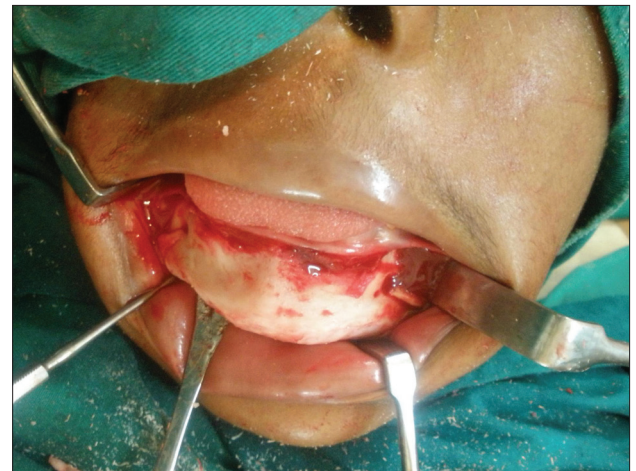


Figure 6: View of Symphysis after excision of tumor

It is most frequently encountered in the mandibular condyle and coronoid processes in the facial skeleton. It has been reported in maxilla, nasal septum, mandibular body, angle and symphysis regions. In the long bones, it is usually found at or near the

epiphyseal-metaphyseal junction. The distal metaphysis of femur and proximal metaphysis of tibia are more commonly involved.^[4]

The origin of this tumor is controversial; developmental,

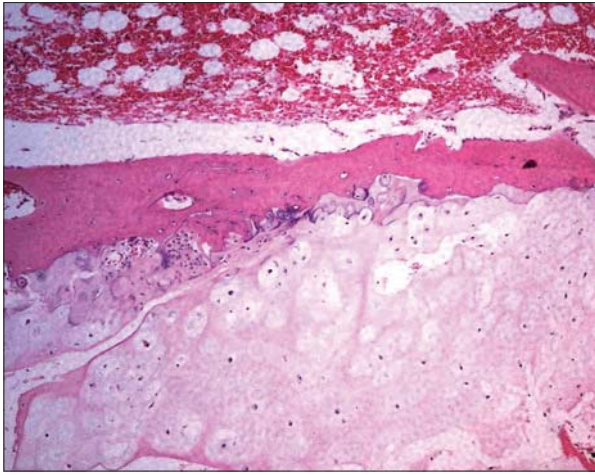


Figure 7: Hematoxylin and eosin-stained slide of histopathologic appearance of tumor showing bony components with a cartilaginous cap

neoplastic, reparative and traumatic etiologies have all been suggested. In the mandible the condyle, coronoid and symphysis regions have cartilage precursors. In the mental region, on either sides of the symphysis one or two cartilages appear and ossify in the seventh month post-conception and form mental ossicles, which become incorporated into the intramembranous bone. Residues of these cartilage precursors in the symphysis appear to give rise to osteochondromas of this region.^[5] Lichenstein proposed that all periosteum has the potential to give rise to osseous and cartilaginous cells; thus, an osteochondroma may arise due to induced or spontaneous metaplasia of the periosteum.^[6] The above two theories may best explain the presence of osteochondroma in the symphysis region.

Osteochondromas are slow growing tumors that parallel the skeletal growth of the individual and usually cease growth after puberty.

The accepted modality of treatment is surgical excision. Recurrence rates are low (2%).^[6] These tumors can occur single or as a part of a syndrome known as osteochondromatosis or Hereditary Multiple Exostosis (HME), an autosomal dominant disorder.^[4] This distinction is important because solitary tumors have 1% risk of sarcomatous changes as compared to 11% risk for patients with osteochondromatosis.^[7] Patients with osteochondromatosis exhibit multiple tumors and may have associated vascular and neurological symptoms, pain and deformity of affected bones (there may be limb shortening of affected bone up to 4 cm). HME is associated with mutations in at least three different genes termed EXT genes, two of which are known to be tumor suppressor genes. HME usually appears by the age of 2-10 years with mean age of diagnosis being 4 years.^[8] Presence of pain is usually an indicator of malignant transformation. Continued growth after puberty raises the possibility of chondrosarcomatous changes. Also greater the size of the cartilage cap (> 2 cm) greater are the risks for malignant change.^[9] In most cases, plain radiographs suffice in diagnosis.

However, for lesions in the spine, pelvis or shoulder, the spiral and cross-sectional computed tomographs allow reconstruction of slices to get an accurate picture. Increased exposure to radiation in children is a drawback with CT scans. T2-weighted magnetic resonance imaging (MRI) is helpful in identifying the size and nature of the cartilage cap and other associated soft tissues but cannot be used in claustrophobic patients and those with pacemakers.

Ultrasonography can be used to assess cartilage cap and associated complications such as arterial or venous thrombosis, aneurysm and bursitis but, cannot be used to assess underlying bone.

Angiography is standard for assessing the vascular occlusion and malignant nature of lesion but, is invasive and may lead to anaphylaxis or renal toxicity due to the contrast medium used. Radionuclide scanning has a high sensitivity but low specificity and also does not help to distinguish between osteochondroma and osteosarcoma. It is not easily available. Experience with 2[fluorine 18]-fluoro-2-deoxy-D-glucose (FDG) positron emission tomography (PET) is limited. It is not easily available and is expensive.^[9]

Given the young age of our patient, we continue to monitor him on a regular basis to check for recurrence and growth of any new tumors. To summarise, Osteochondroma is a slow growing benign tumor that shows low risk of recurrence or sarcomatous changes. Simple surgical excision is usually curative. However, in patients with multiple tumors, it is important to screen for the presence of osteochondromatosis. Regular clinical and radiological follow-up after surgery is essential to detect the growth of new tumors.

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