

CASE REPORT

Osteochondroma of the palate: An interesting and an unusual case presentation

Kranti Kiran Reddy Ealla, Surekha V Reddy, Srinivas Gadipelly¹, CR Charan

Department of Oral and Maxillofacial Pathology and ¹Oral and Maxillofacial Surgery, MNR Dental College, Sangareddy, Hyderabad, Andhra Pradesh, India

Address for correspondence:

Dr. Kranti Kiran Reddy Ealla,
Department of Oral and Maxillofacial Pathology,
MNR Dental College and Hospital,
Fasalwadi, Narsapur Road, Sanga Reddy,
Andhra Pradesh - 502 294, India.
E-mail: drekrkr@yahoo.co.in

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ABSTRACT

A 40-year-old Indian male patient was referred to the Department of Oral and Maxillofacial Surgery with a slowly enlarging intra-oral, right-sided palatal swelling of one-year duration, with a previous diagnosis of osteochondroma. Extraorally, patient presented with a mild right-sided facial swelling. On intraoral examination, the palatal swelling was extending from the distal aspect of canine to the distal aspect of second molar with involvement of the maxillary tuberosity. The swelling was non-tender, bony-hard in consistency and covered by normal mucosa. The medical history was non-contributory with no relevant family history of any skeletal disease. Despite the attempt for complete removal of the tumor previously, it recurred within six months. The present article reports an extremely rare clinical case of endosteal (central) osteochondroma, manifesting itself as a radiopaque mass in the right posterior aspect of the palate.

Key words: Cartilage, osteochondroma, palate

INTRODUCTION

Osteochondroma (OSCH), also called as osteocartilaginous exostosis, is a common benign tumor of endochondral axial skeletal bones, while its occurrence in the mesenchymal craniofacial bones is extremely rare.^[1] They usually occur in the long bones of the lower extremity. Incidence of these tumors in the craniofacial geography is extremely uncommon. Nevertheless, few cases have been reported in the base of the skull, zygomatic arch and maxillary sinus with high frequency of tumors occurring in coronoid process or the mandibular condyle.^[2,3]

The OSCHs are categorized as cartilaginous tumors because of their characteristic progressive endochondral ossification of growing cartilaginous cap. The fact that the jawbones develop by intramembranous ossification elucidates the rarity of these tumors in the jaws. Condyle being primarily cartilaginous in origin is the most affected site in the jaws by OSCH. The original reports described lesions exclusively in the condyle and coronoid processes.^[4] Osteochondromas are very rarely encountered in

the palate. According to English medical literature from 1966 to April 2013, via the Medline database, it has been revealed that there are almost no cases of palatal OSCH reported till date.

The tumor occurs predominantly in males rather than females (M/F ratio 1.6:1) with an age range of 20 to 30 years.^[5] Osteochondromas can develop as a single tumor (75%) or as multiple tumors (25%).^[5,6] The multiple variant also called as osteochondromatosis is usually inherited as an autosomal dominant trait. The increased propensity of sarcomatous transformation in multiple osteochondromas signifies the importance of delineating single from the multiple tumors.^[7]

In the current article, we describe a unique case of osteochondroma of the posterior palate with recurrence, never reported before in the literature.

CASE REPORT

A 40-year-old male was referred to the Department of Oral and Maxillofacial Surgery with a chief complaint of swelling in the right posterior aspect of the palate [Figure 1]. The past medical history revealed that he had a similar swelling 3 years back, which was removed under general anesthesia. A histopathological diagnosis of osteochondroma was then rendered. Six months following the initial surgery, the patient noted a recurrence in the same area. Despite the progressive increase in size of the recurrent lesion, patient did not undergo further investigations because of its asymptomatic nature.

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However, the patient was convinced to undergo the treatment after 1 year. Orthopantomogram (OPG), magnetic resonance imaging (MRI) and computerized tomography (CT) scan were performed as pre-operative investigations. The coronal, axial and 3-dimensional CT images revealed a hyper dense mass developing from the posterior palate and extending into the infratemporal space [Figure 2].

Extraorally, a mild right-sided facial asymmetry was noted. On intraoral examination, the palatal swelling was extending from the distal aspect of canine to the distal aspect of second molar involving the maxillary tuberosity. The swelling was lobulated, non-tender, bony hard in consistency and was covered by normal mucosa.

In accordance with the previous medical history and the present investigatory findings, the patient was operated under general anesthesia.

A standard Weber Ferguson incision extending from the right philtral ridge of the upper lip along the ala, following the contour of the nose up to the medial canthus and then turning perpendicularly, parallel to the lower eyelid below the outer canthus was made. Intra-orally, incision was made between canine and first premolar 5 mm lateral to midpalatine suture extending around the maxillary tuberosity to join the skin incision. Partial maxillectomy and removal of palatal bone on right side was done to accomplish 1 cm tumor-free margins. In the present case, the resulting defect was lined with split skin graft taken from left thigh and an obturator was provided to cover the defect and facilitate mastication.

The surgical specimen consisted of a single lobulated mass of bony hard tissue that measured approximately $5 \times 3 \times 2$ cm. A thin layer of fibrous tissue incompletely covered the tumor mass [Figure 3].

Histopathologically, the encapsulated lesion comprised of proliferation of bony trabeculae in the center and hyaline cartilage-like tissue in the periphery. The cartilaginous area was hypercellular with moderately enlarged nuclei in a myxoid background [Figures 4-10].

The bony trabeculae were conspicuously formed by a process of endochondral ossification from the peripheral cartilage along with different degrees of calcification. These features corresponded to those of osteochondroma. This recurrent lesion had histopathological features identical to that of the primary lesion.

DISCUSSION

Osteochondroma is recognized by the World Health Organization as an osteocartilaginous lesion protruding from the outer cortex of the affected bone.^[8] Many descriptive

terms have been proposed based on the OSCHs tissue activity, one being osteocartilaginous exostosis, implying reactive exostosis and on the other hand it is termed as osteochondroma, considering its benign nature.^[9]



Figure 1: Intra-oral picture showing swelling in the right posterior aspect of the palate

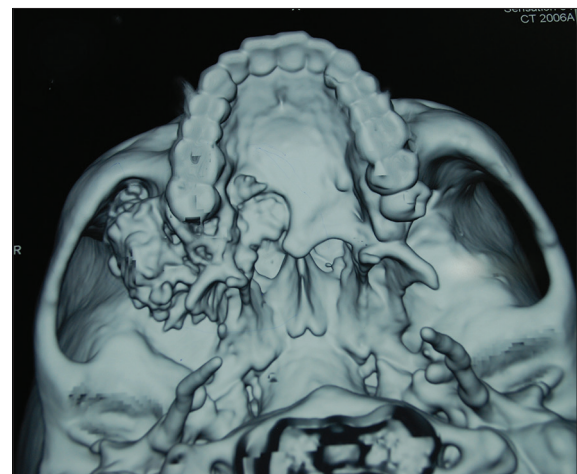


Figure 2: 3D CT revealed a hyper dense mass arising from the posterior palate and extending into the infratemporal space

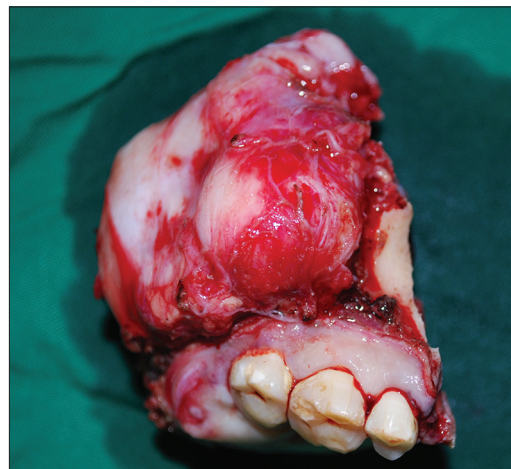


Figure 3: Gross specimen showing lobulated mass of bony hard tissue

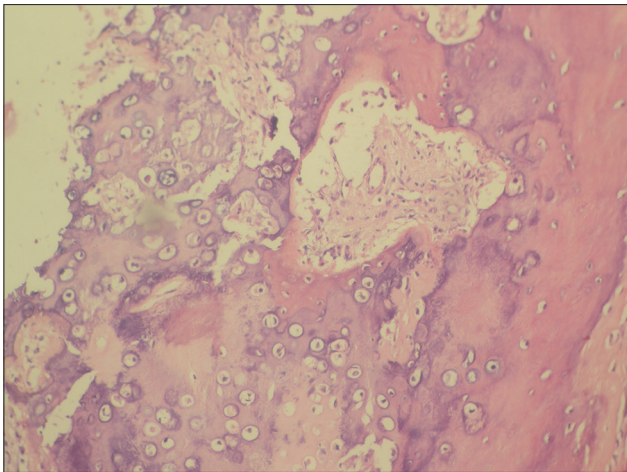


Figure 4: Photomicrograph showing hyaline cartilage with hypercellular chondrocytes (H&E stain, x100)

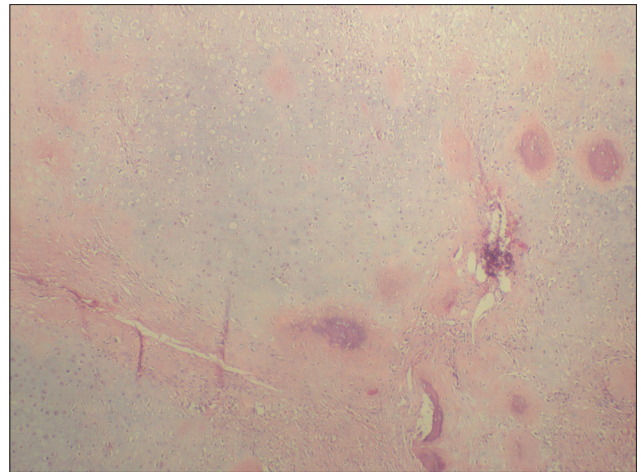


Figure 5: Photomicrograph showing a mixture of hyaline cartilage and trabeculae of bone (H&E stain, x40)

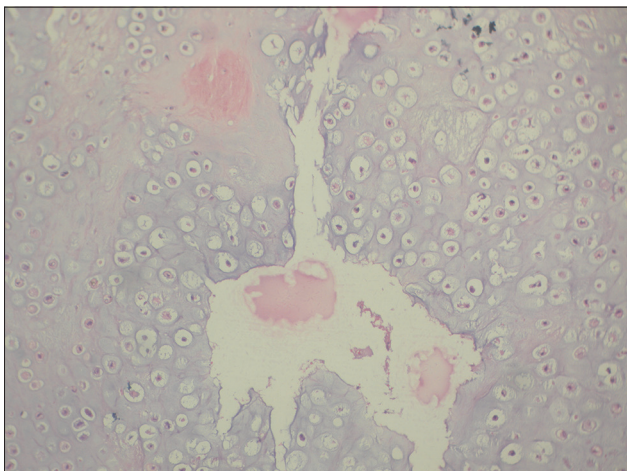


Figure 6: Photomicrograph showing a matrix of mature hyaline cartilage (H&E stain, x100)

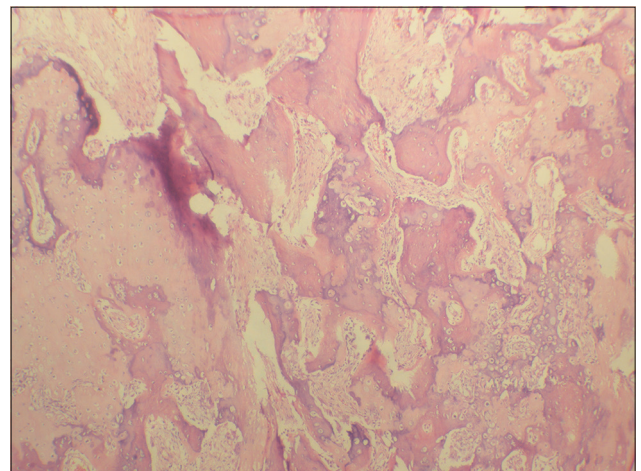


Figure 7: Photomicrograph showing a mixture of cartilagenous and osseous differentiation (H&E stain, x40)

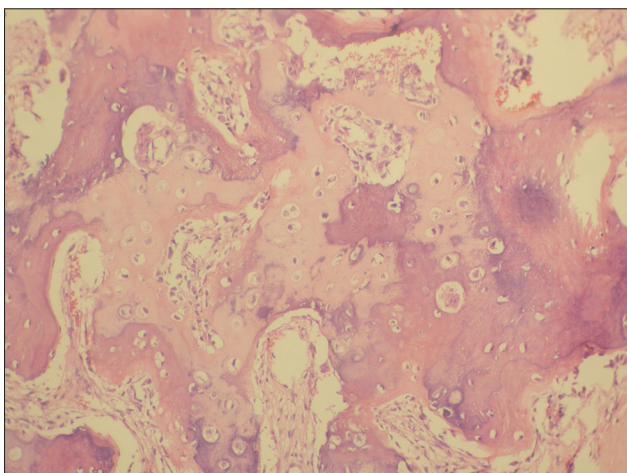


Figure 8: Photomicrograph showing a mixture of cartilagenous and osseous differentiation (H&E, x100)

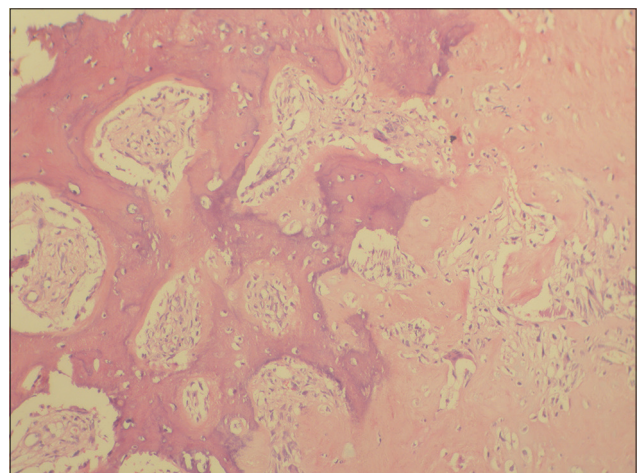


Figure 9: Photomicrograph showing a mixture of bony trabeculae and cellular connective tissue stroma (H&E, x100)

In the craniofacial region, OSCHs are common in the mandible because they develop by intramembranous ossification; however, their occurrence in the palate is extremely rare.

Our case belongs to the minority of osteochondromas that occur in the palate. The possible explanation for the present case arising in the palate could be a biologic reaction of the

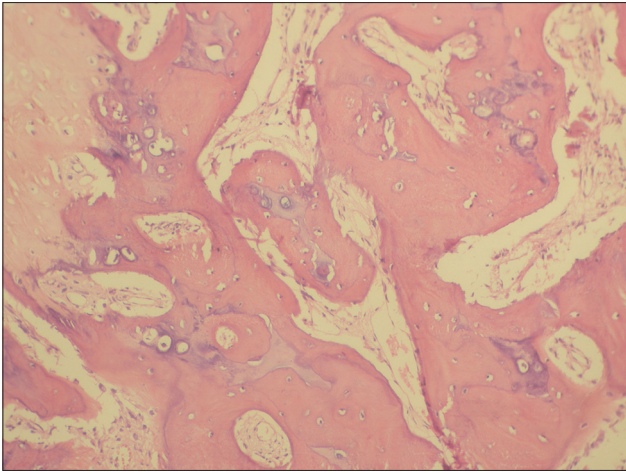


Figure 10: Photomicrograph showing an admixture of cartilagenous and osseous differentiation (H&E, x200)

periosteum in response to induced or spontaneous metaplasia of the periosteum. In turn periosteum being pluripotential, can give rise to cartilaginous cells with subsequent endochondral ossification resulting in osteochondroma.^[10]

Osteochondromas are seen more often in young patients, second to fourth decade with no sex predilection.^[11] More than 50% of osteochondroma patients display jaw expansion that can lead to marked deformity of the face. However, pain is considered to be an uncommon feature.

On radiographic analysis, OSCHs appear as radiopaque mass on the conventional peri-apical and panoramic radiography or on CT.^[12] However, radiographic interpretations often lead to confusing and uncertain conclusions of the exact bone pathology. The unusual location of the present case and its radiographic feature emphasize the importance of a preoperative bone biopsy for determining an exact diagnosis.

The histopathology of osteochondromas is distinctive, although there are significant variations. The lesion features chondrocytes as the main cellular component arranged in clusters in parallel, oblong lacunar spaces. The mineralized tissue comprises of mature hyaline cartilage, regular bony trabeculae produced by endochondral ossification. They are uncommon, can grow rapidly and may recur after attempted excision.

Histologically, osteochondroma needs to be distinguished from osteoma, benign osteoblastoma, chondroma, chondroblastoma and bizarre parosteal osteochondromatous proliferation (BPOP).^[13]

Treatment of choice for osteochondroma depends on its clinical course, on the presence of complications and cosmetic reasons. Preoperative evaluation of the patient requires physical examination, CT, MRI and a biopsy of the lesion. The tumor

has to be completely excised including the surrounding bone and periosteum in order to avoid recurrences. The recurrence rate reported amongst the osteochondromas is less than 2%.^[14] However, inspite of conventional treatment, our case recurred within six months emphasizing the importance on close and regular follow-up.

The recurrence rate of osteochondroma is attributed to the treatment procedure. It can range from 0% to 15% when treated with wide resections while those treated with marginal or intra-lesional resection show 57% to 78% recurrence.^[15] So it is advised to completely resect the overlying perichondrium, as improper excision results in recurrence. Although soft tissue recurrence is very rare, it can occur as a result of leakage of myxomatous cartilage into the surgery bed. Mortan in 1964 stated that a greater discrimination must be made in using ominous description recurrence because rare true recurrence must strongly favor malignancy.^[16]

Malignant transformation in these tumors is rare, which accounts to less than 2%.^[17] Low-grade chondrosarcoma and osteosarcoma are the common malignant counterparts that arise in the cartilage cap and at the base of the OSCH respectively.^[18,19]

Considering the rates of recurrence and malignant transformation, a regular clinical check-up and radiographic evaluation should be performed aiming to exclude malignancy. On postoperative radiographs, any progressive erosion or destruction of the bone adjacent to the previously treated osteochondroma or the presence of a soft tissue mass with irregular calcification suggest malignancy.^[20]

In view of the recurrence in the present case, a more radical operation involving subtotal maxillectomy was performed to remove the recurrent osteochondroma. The resulting defect was reconstructed using split skin graft taken from the left thigh and an obturator to cover the defect and facilitate mastication.

In conclusion, osteochondromas are uncommon, which need to be recognized and managed appropriately in view of their distinctive clinical behavior. Despite the complete excision of the lesion in the present case, our patient needs to be followed up carefully, considering its aggressive nature, since it recurred after a previous attempt at excision.

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