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CASE REPORT

Periosteal Chondroma of Mandible: Report of a Case and Differential Diagnosis

[1]Pritam Kumar Mankapure, [2]Suresh R Barpande, [3]Jyoti D Bhavthankar

ABSTRACT

Cartilaginous tumors make up the second largest group of bone neoplasms. These are common in the long bones of the body. Chondromas among them are mostly painless, intraosseous lesions with presence at both endosteal and periosteal location. They are relatively rare in jaws owing to their membranous development and are reported to occur in cartilage bearing areas of jaws. This article is aimed at presenting a case of periosteal chondroma in mandibular premolar region with an emphasis on its histogenesis and differential diagnosis because of its similarity with some other exophytic bony lesions in the area.

Keywords: Cartilaginous tumors, Enchondroma, Mandible, Periosteal chondroma.

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INTRODUCTION

Chondroma is a benign tumor composed of mature hyaline cartilage.[1] Though it is a well recognized entity in certain areas of bony skeleton, it is uncommon in jaws. In more than 10,000 bone lesions at the Mayo Clinic, there were no cases of enchondroma in the jaw or facial bones. Among 1243 chondromas in four large series reported, only four (0.32%) were in the head and neck region.[2] According to their location, they can be classified as enchondroma for central, periosteal chondroma for cortical and soft tissue chondroma for extraskeletal location.[3] Among these, periosteal chondromas are rare and comprise 2.2% of all benign tumors.[4] Here, we report a case of periosteal chondroma in premolar region of mandible with its differential diagnosis.

CASE REPORT

A 60-year-old female patient visited our department with a complaint of gradually enlarging swelling in lower left region of face since 10 to 12 years which began as a peanut size. There was no accompanying pain or discharge and no history of trauma. The medical history was noncontributory. Extraorally, it manifested as slight bulge beneath left corner of mouth (Fig. 1). Intraorally, approximately 2.5 × 2.0 cm, bony hard, roughly oval swelling was seen over buccal alveolar region of 33 to 35 extending from marginal gingiva to obliterate buccal vestibule inferiorly. The surface of swelling was nodular with stretched overlying mucosa (Fig. 2). There was slight tenderness on palpation. Associated teeth were firm but appeared slightly displaced lingually. Periapical radiograph and orthopantogram showed no significant change in radiopacity at lesional site. However, occlusal radiograph showed a radiolucent lesion with radiating bony

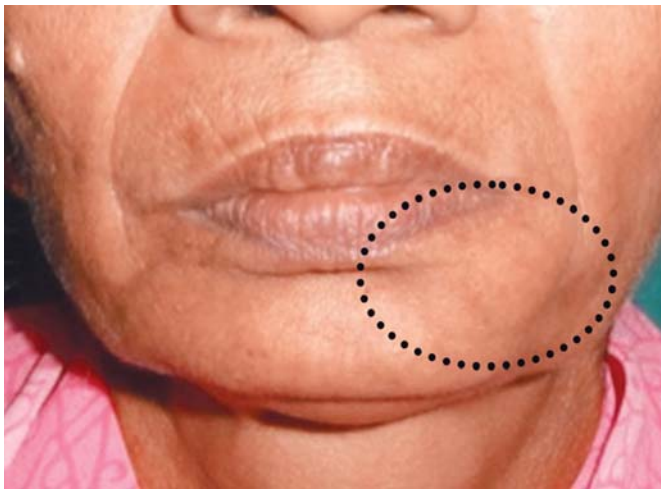

Fig. 1: Slight bulge beneath left corner of mouth

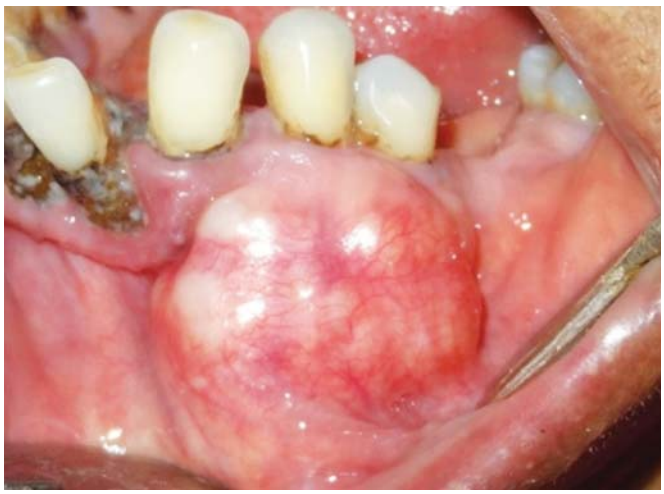

Fig. 2: Intraorally, hard, well-defined, nodular swelling with overlying stretched mucosa

[1]Postgraduate Student, [2]Professor and Head
[3]Associate Professor

[1-3]Department of Oral Pathology and Microbiology, Government Dental College and Hospital, Aurangabad, Maharashtra, India

Corresponding Author: Pritam Kumar Mankapure, Postgraduate Student, Department of Oral Pathology and Microbiology, Government Dental College and Hospital, Aurangabad-431001, Maharashtra, India, Phone: 9011179100, e-mail: pritammankapure@gmail.com

trabeculae from cortex outward as overhanging radiopaque edges in swelling (Fig. 3). Computed tomography scan showed exophytic bony lesion confluent with buccal cortex in premolar region of mandible (Fig. 4).

After histopathologic confirmation of chondroma on incisional biopsy, the lesion was excised into leaving a raw area on cortical surface of bone indicating its periosteal origin. Histopathological findings of excised specimen were consistent with those on incisional biopsy. Beneath the fibrous capsule, lobules of cartilage were seen separated from overlying epithelium. At deeper part, these lobules were surrounded by mineralization in form of C's and O's (Fig. 5). Cartilage cells were small, uniform in size with slightly hyperchromatic nuclei and scanty cytoplasm with indistinct cell outline, embedded in hyaline basophilic stroma. These cells were residing within varying sized lacunae. Few binucleated cells were seen. Mitoses were absent (Figs 6 and 7). Based on the clinicopathologic features, final diagnosis of periosteal chondroma was made. Postoperative course was uneventful. Patient was followed up for 1 year and was free of recurrence.

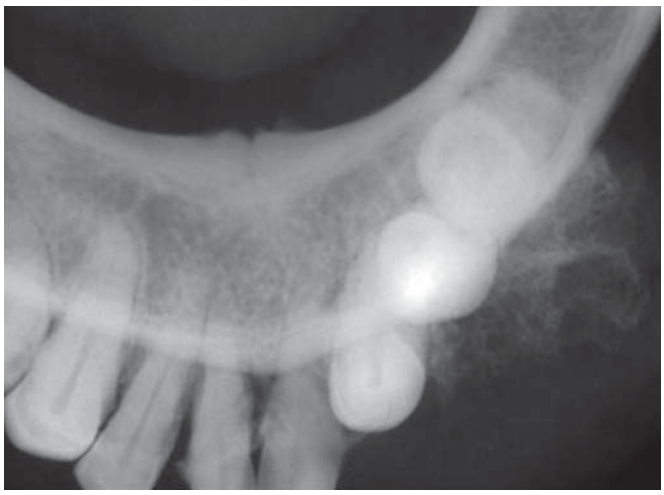

Fig. 3: Protruding edges of bony trabeculae (occlusal radiograph)

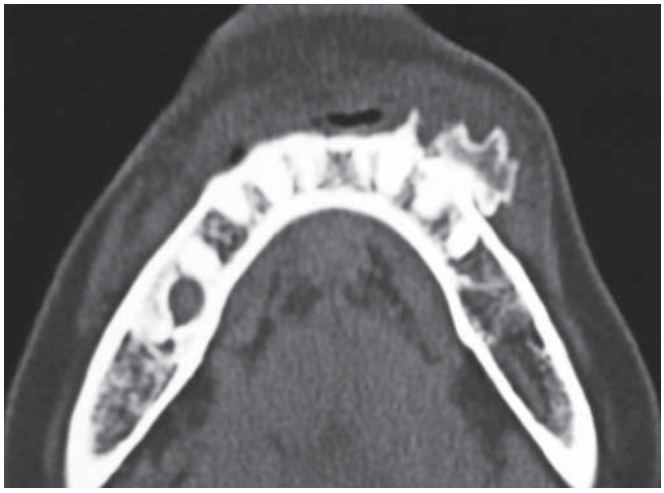

Fig. 4: Exophytic bony lesion on left body of mandible in continuity with cortical bone (CT scan—sagittal view)

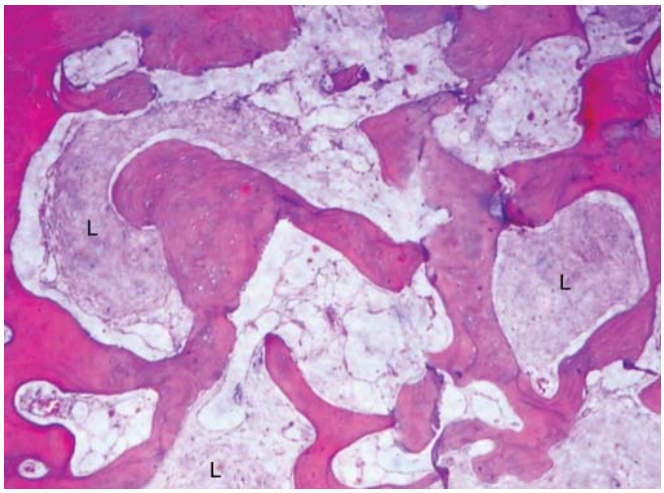

Fig. 5: Lobules of cartilage (L) surrounded by mineralization in form of Cs and Os (H & E, ×4).

DISCUSSION

Cartilaginous tumors are one of the commonly encountered tumors in the long bones, bones of hands and feet but are relatively uncommon in head and neck region.[1,5-7] Reported sites for cartilaginous tumors in head and neck are ethmoid sinuses, nasal septum, and larynx correlating with their intrinsic cartilaginous nature.[8] In jaw bones, these lesions are more common in condyle and coronoid process with cartilaginous components. This had tempted some authors to refer to them as hamartomatous growth.[9] However, these tumors are also reported in other parts of jaws like body, ramus and symphysis of mandible.[2] In such cases, aberrant embryonic cell rests and multidirectional differentiation of mesenchymal cells has been implicated in origin of tumors at these sites.[10]

Chondromas demonstrate a wide age range, being most common in 3rd to 4th decades of life and having approximately equal sex distribution. In most cases, chondromas are solitary lesions. However, in Ollier disease, multiple and widespread chondromas are seen with a tendency to be unilateral. In another presentation termed Maffucci syndrome, skeletal chondromatosis is seen in association with soft tissue angiomas.[1] Oral soft tissue chondromas have been found in the tongue, cheek, hyperplastic tissue of denture-bearing areas, and on the nasal surface of the soft palate.[11]

Chondroma seldom develops in membrane bones without any vestigial cartilaginous remnants. In mandible, such remnants are present in mental region, coronoid and condylar process and in maxilla, in anterior premaxillary region, atleast during embryonic development.[7,9,12-14] Enchondromas of the maxilla are located in the anterior region adjacent to the nasal spine and nasal septum, while mandibular enchondromas have been observed mostly in the

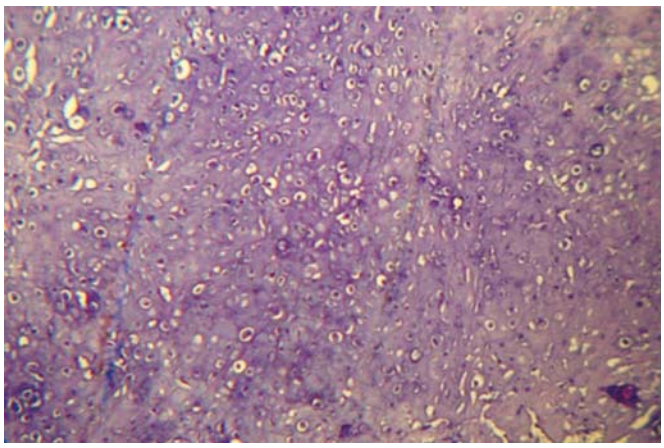

Fig. 6: Numerous round to oval cells residing in varying sized lacunae; embedded in basophilic stroma (H & E, ×10)

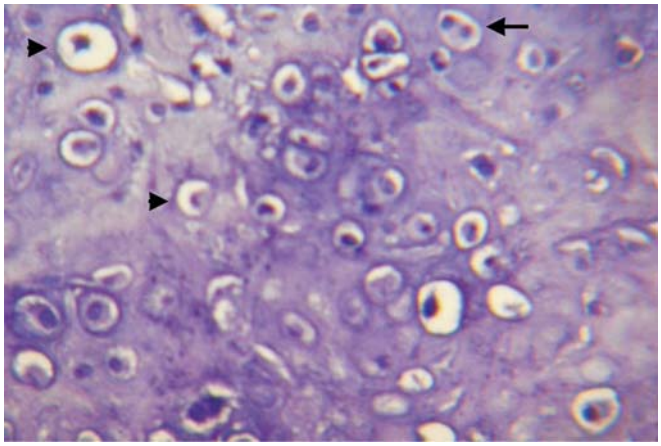

Fig. 7: Slightly hyperchromatic nuclei with scant cytoplasm and indistinct cell outline residing in varying sized lacunae (arrowheads). Few binucleated cells were seen (arrow) (H & E, ×40)

condyle and coronoid process.[5,9,11] The development of lesion in other areas of lower jaw is certainly in accordance with the vestigial cartilaginous remnant theory because of presence of Meckel's cartilage in vicinity of developing mandible and cartilaginous origin of mental ossicles.[8,12,13] Schaffer in 1888, described a hard tissue type, different from bone and cartilage and termed it as chondroid bone.[15] Several reports of its sighting along sutural lines in craniofacial region appeared since then, even in mandibular symphysis.[16] Also, the tissue formed during the rapid growth period of alveolar process at the crest, resembles chondroid bone.[17] However, its contribution in neoplastic histogenesis is questionable. One of the report of chondroma in ribs suggested surgical trauma to induce chondroma formation.[4] Trauma may induce metaplastic cartilaginous transformation.[10] In present case, no such history of trauma was evident.

Lichtenstein and Hall (1952) were first to describe periosteal chondroma as a separate entity and it was termed as juxtacortical chondroma by Jaffe (1956).[18] Lichtenstein suggested pluripotency of periosteal cells as source for origin of these tumors. However, Kermer C et al[8] (1996) had preferred vestigial cartilaginous theory of origin. They are more frequent in appendicular skeleton. Most cases occur before 50 years of age, commonly in 2nd to 3rd decades and male predilection has been reported in a ratio of 2:1.[1,8] Clinically, they manifest as slow growing painless with ovoid or variable convexity.[18] In present case, painless, slow growing nature of lesion was unalarming for patient until it attained a noticeable size. Overlying skin or mucosa is seldom involved due to gradual expansion of the lesion as seen in present case.[5,6]

Radiologically, tumor may show irregular, variably radiopaque, and radiolucent mottled mass protruding from cortex.[6] It shows so called overhanging edges of bone and soft tissue masses with variable pattern of calcification.[5,19] deSantos and Spjut (1981) defined three diagnostic radiographic criteria for periosteal chondroma[18] which are as follows:

1. Scalloping of peripheral cortex.
2. Overhanging edges of bone.
3. Variable amount of calcification or ossification of cartilaginous matrix.

Radiologic work-up, in present case, confirmed the origin of lesion from cortical surface with presence of over-hanging edges and calcified matrix. In present case, calcification was more pronounced to a degree that scalloping was unassessable. Areas of calcification or ossification should be considered as evidence of development of lesion over a long period.[5]

Clinical differential diagnosis includes periosteal osteoma, osteoblastoma, osteoid osteoma, osteochondroma, low-grade chondrosarcoma and peripheral osteosarcoma. Exostoses are localized bony protuberances that arise from cortical surfaces, are usually bilateral, lobulated or smooth surfaced and rarely attain such a large size. Osteoma, usually periosteal type, presents as radiologically dense well-circumscribed radiopacity. Osteoid osteoma and osteoblastoma can be differentiated by the rapid growth in most cases and deep dull pain. Most cases of osteoblastoma are central but periosteal osteoblastomas have been described. The distinction from these osteogenous tumors can be difficult clinically and radiologically in presence of ossified cartilage and warrant histopathologic examination.[1,2,7] Osteochondroma is a benign hamartomatous process which manifests as pedunculated or sessile mass protruding from the parent bone. It usually develops in children and ceases growing with cessation of skeletal growth.[2,20]

Low-grade variants of osseous malignancies like low-grade chondrosarcoma and peripheral osteosarcomas may lack any pathognomonic features and can manifest as slow growing, painless tumor. Periosteal or juxtacortical chondrosarcoma arises from surface of bone and may erode the cortex.[6] Size can be a clue as chondrosarcomas are larger

than 3 to 4 cm.[2] Periosteal osteosarcoma is a surface lesion that contains cartilage, but has more active biologic behavior. Also, parosteal osteosarcoma, a very uncommon form of osteosarcoma, because of its slow growth and lower tendency for metastasis; should be considered in differential diagnosis. Indolent behavior of present lesion with absence of permeation into underlying cortex on radiographic investigation ruled out these osseous malignancies.[2,6]

Histologically, periosteal chondromas show a lobulated configuration of hyaline cartilage with chondrocytes within well-formed lacunae covered by periosteum. The chondrocytes are small, with indistinct cytoplasmic borders; nuclei are typically small, round and densely hyperchromatic. Occasional binucleate cells may be found but are not numerous. The cartilage lobules are found in nests between the cancellous bone trabeculae and separated from them by a clear zone (Fig. 7). Calcification and ossification were seen mostly in form of semicircles and circles described as Cs or Os.[1,5,7]

Enchondroma and periosteal chondroma share histopathological features. However, enchondromas are comparatively hypercellular than periosteal variety.[6,21] Chondromas in Ollier's disease and Maffucci syndrome are hypercellular, contain atypical nuclei and binucleated chondrocytes in a predominantly myxoid stroma, all features suggestive of low-grade chondrosarcoma.[1,2]

Histopathological differential diagnosis includes chondroid tumors like osteochondroma, low-grade chondrosarcoma and chondroblastic osteosarcoma. Osteochondroma is differentiated by presence of a cartilaginous cap, composed of hyaline cartilage. At deep aspect of cartilaginous cap, where it interfaces with bone, endochondral ossification is seen.[2,22,23] The distinction between chondroma and low-grade chondrosarcoma can be challenging. Torbaghan SS et al suggested that lobulation pattern and fibrous tissue formation around the tumor can be an effective and helpful indicator for histopathologic differentiation. There is regular lobulation pattern in chondroma with a fibrous capsule which is almost inactive with little blood vessels and low cellularity. On the other hand, because of fast growth, chondrosarcoma produces an irregular, asymmetrical lobulation pattern. The blood vessels and active blast cells are more numerous in the chondrosarcoma capsule.[24] In the present case, tumor tissue seemed to exhibit regular lobulation pattern with calcification around lobules in focal areas. However, assessment of fibrous capsule for differentiation was not possible as the ossification of neoplastic cartilage was very extensive (Fig. 7). Occasional cases of chondroma may have features like chondrocytes having large, open-faced nuclei with a visible chromatin pattern and numerous binucleate cells. In such cases, a radiologic pattern showing an absence of cortical destruction is an important point that favors a benign diagnosis. Mitoses are extremely rare to nonexistent in chondroma, and the finding of more than a rare mitotic figure indicates a high probability that the tumor is malignant.[1,2,7]

Chondroblastic osteosarcomas, in some cases, may be composed almost entirely of malignant cartilage growing in lobules with only small foci of direct osteoid production by tumor cells. It can be distinguished from chondroma by presence of malignant features of cartilage cells and presence of osteoid tissue.[2]

Malignant transformation in long standing chondroma is much debated, with some authors claiming that all chondrosarcomas arise from pre-existing chondromas, whereas others claim to find no evidence of a pre-existing chondroma. However, it is well accepted that patients with Ollier's disease and Maffucci's syndrome have a high risk of malignant change, with a reported incidence that varies from 12 to 50%.[2] In present case, there were no signs of sarcomatous differentiation and macroscopic and microscopic characteristics were indicative of chondroma.

Treatment is usually directed toward total surgical removal of the tumor. Chondromas should be excised as low-grade chondrosarcomas with 1 cm peripheral margin considering the extreme rarity of this lesion above the clavicles and very thin line of differentiation from the benign histologic appearance of low-grade chondrosarcoma.[24] Although, recurrence is rare, patient is followed up for 9 months and follow-up examinations will be continued due to recurrence with malignant transformation as a possible complication.

CONCLUSION

To the best of our knowledge, a case of periosteal chondroma in noncartilage bearing area of lower jaw has not been previously reported in english language literature. However, diagnosis of any type of chondroma in jaws and facial bones should be viewed with great skepticism because of propensity to recur and act in a malignant manner.

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