

PERIOSTEAL CHONDROMA OF THE MANDIBLE: A RARE CASE REPORT.

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ABSTRACT

Chondromas are benign tumors of mature hyaline cartilage. They are common in the long bones of the body but were rarely reported in the head and the neck. Following their location, chondromas can be classified as: enchondromas, periosteal chondromas and extra-skeletal or soft tissue chondromas. In this paper, the authors present a rare case of recurrent periosteal chondroma occurring in the left premolar region of mandible of a 38-year-old woman. Review of the literature about clinical, radiological and histological features as well as treatment of this tumor is also made, with an emphasis on its differential diagnosis with chondrosarcoma because of their overlapping features.

Keywords: Cartilaginous tumors, head and neck, Mandible, Periosteal Chondroma, chondrosarcoma

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INTRODUCTION

Chondroma was first described by Muller in 1838.

[1] It is a benign tumor of mature hyaline cartilage.

[2] The tumor is more commonly found in long bones and rarely located in the head and neck region.[3] It has been reported in the tongue [4] ; the temporomandibular joint (TMJ) [5, 6] ; the cheeks [7] ; the hyoid bone [8] and the anterior maxilla in adult patients with a higher rate.[3] Given its rarity, it is necessary to differentiate it from true chondrosarcoma or other lesions of more frequent presentation [9].

Depending on their locations, chondromas can be classified as:

- a. Enchondromas which originate from the marrow in the medullary cavity of the bony skeleton
- b. Juxtacortical or periosteal chondromas which originate adjacent to the periosteum below the cortical surface. They are rare and account for 2.2% of all benign tumors [10], and
- c. Extra-skeletal or soft tissue chondromas.[2,10, 11]
- d. The authors report a rare case of a periosteal chondroma in the premolar region of the mandible, with an emphasis on its differential diagnosis with chondrosarcoma because of their similar features.

PATIENT & OBSERVATION

A 38-year-old female presented to the Center for Consultation and Dental Treatment of Rabat with a chief complaint of painful swelling on the left anterior mandible involving buccal mucosa. The patient reported that she underwent a traumatic avulsion of the tooth 36 two years before; the duration of healing delay exceeded 2months. She also reported that it took her a long time for the socket to heal and that the swelling started shortly after. Extraorally, a slight bulge beneath the left corner of the mouth was leading to gross facial asymmetry (**Fig. 1**).



Fig. 1: Slight bulge beneath the left corner of mouth, leading to gross facial asymmetry

Intraorally, in the left mandibular body, a roughly oval swelling of approximately 1.5×1.0 cm was seen in the buccal part of the 35 and 36. The tumor was obliterating the left inferior buccal vestibule. The overlying mucosa was stretched (**Fig. 2**). There was a slight tenderness on palpation.



Fig. 2: intraoral, roughly oval swelling obliterating the inferior buccal vestibule, with overlying stretched mucosa.

The orthopantomogram (**Fig. 3**) showed an ill-defined mixed (radio opaque-radiolucent) lesion which appeared to be superimposed to the cortical bone at the inferior left molar region.



Fig. 3: orthopantomogram: teeth 35 and 36 are absent. An ill-defined mixed lesion is seen in the left mandibular premolar region.

Decision was to proceed with the excision of the tumor under local anesthetics. It appeared to be a benign cartilaginous lesion which was well limited (**Fig. 4**). The lesion was excised, leaving a raw cortical surface of bone indicating it's periosteal.



Figure4: well limited lesion with a benign cartilaginous aspect

Histopathological findings showed a well limited lesion (fig 5). It consisted of cartilaginous nodules separated by a hyaline matrix. These nodules were moderately cellular, with presence of chondrocytes housed individually in lacunary spaces. The nuclei were rounded with dense chromatin. Some binucleated cells were found. Mitoses were undetectable and necrosis was absent. These histopathological features described above were in favor of a chondroma (**Fig. 6**).

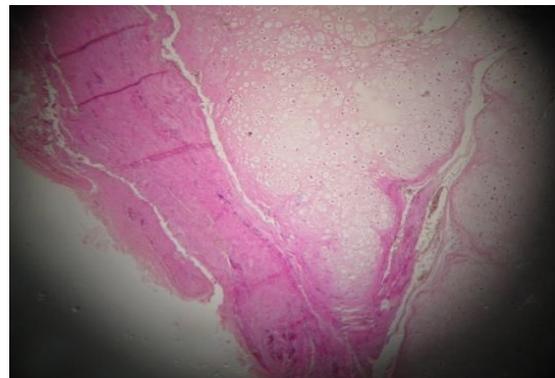


Fig 5: photo-micrograph showing well limited lesion with cartilaginous and moderately cellular lobules (hematoxylin and eosin stain, x4)

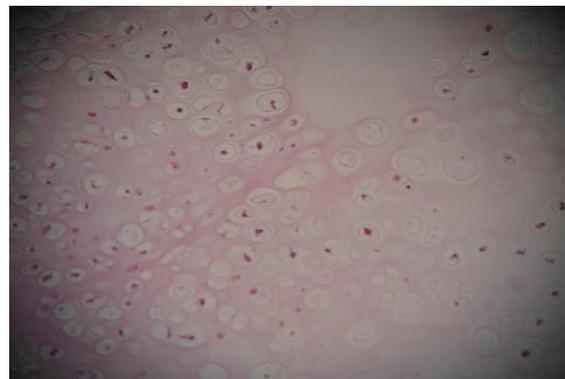


Fig. 6: Chondrocytes, without major nuclear atypia (haematoxylin and eosin stain, x10)

On the basis of all clinical, radiographic, and histopathology features, final diagnosis of periosteal chondroma was retained. The postoperative course was uneventful. The patient was notified of the benefit of a bi-annual follow-up, since this tumor has a high recurrence rate, can be miss diagnosed or can potentially transform to a chondrosarcoma.

Unfortunately, the patient missed the first appointment 6 months later, and came back one year later with a recurrent painful swelling in the same region with an extension to the lingual side of the mandible. The patient underwent a mandibular CT-scan, which revealed an exophytic bony lesion infiltrating the medullar bone of the left mandibular body, with a heterogeneous aspect (fig 7). A malignant transformation was suspected given the exacerbation of symptoms and the fast growth of the recurrent lesion. She was then referred to a department of Maxillofacial Surgery.

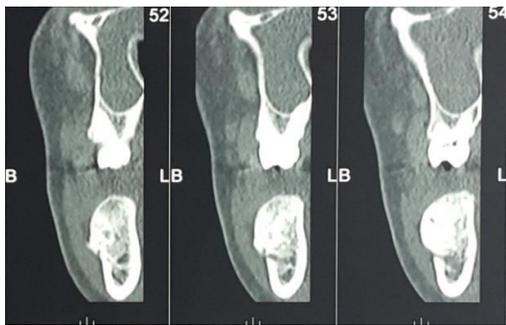


Fig 7: coronal reconstruction of CT-scan showing exophytic heterogeneous hyperdense lesion infiltrating medullar bone of the mandibular body.

DISCUSSION

Chondromas are benign tumors of uncertain etiology [9]. They are composed of mature cartilage that may arise from residual cartilage within bone as a hamartomas [11] (enchondromas), or from mesenchymal cells, in the periosteum or in the soft tissue, that undergo neoplastic cartilage cell differentiation (juxtacortical chondromas or soft tissue chondroma). [12, 13] Chondromas are ones of the commonly encountered tumors in the long bones and bones of hands and feet. [10]

In a review of 8542 bone tumors by Dahlin and Unni, chondromas accounted for 12% of benign tumors and 2.8% of all tumors. [14] However, the occurrence in the maxillomandibular area is exceptional. [1] K. Matsuzaka *et al.* in 2002 [15], in a statistical review of 559 cases of osseous, cemento-osseous and cartilaginous lesions of the jaws lesions, only one case of chondroma was reported). The common location of the chondroma depends on its nature. Oral soft tissue chondromas have been found in the tongue, the cheek, and arising in hyperplastic tissue in denture-bearing areas. [9]

For the enchondroma, lesions of the maxilla are mostly located in the anterior region adjacent to the nasal spine and nasal septum, and those of the mandible, in the symphysis, the body, the coronoid process and the mandibular condyle. [16, 17]

Chondromas have approximately equal sex distribution. [8, 9] They are usually discovered in the third or fourth decade of life, although they have been found from the first to the eighth decade of life. [14, 17]

Periosteal chondroma was first described as a separate entity by Lichtenstein and Hall (1952) and it was termed as juxtacortical chondroma by Jaffe (1956). [18] It presents with some specific features. They are more frequent in the appendicular skeleton and are extremely uncommon in the head and neck region. Most cases occur before 50 years of age, commonly in the second to the third decades. Male predilection has been reported with a ratio of 2:1. [19, 20] The specific aspect of this case report is that the chondroma presents in a 38 year old female patient, in the mandibular region.

The chondroma of the jaw appears usually as a slow growing, painless and well limited tumor with an ovoid or a variable convexity. As in our case, the patient may experience an expanding mass or moderate symptoms for several years before the diagnosis is made. However, chondromas could easily be fortuitously discovered as firm, smooth nodules which may not expand and preserve a small size that can be fortuitously discovered in a routine examination. Since the lesion expands gradually, the overlying skin or mucosa is rarely involved, adjacent structures can be repressed and tooth mobility and root resorption are possible [1, 9, 10].

In most cases, chondromas are solitary lesions. However, special clinical forms exist [1, 10]. In Ollier disease, multiple and widespread chondromas are seen, with the maxillomandibular area exceptionally involved. It affects the young patients, with a tendency towards unilateral lesions and an increased rate of malignant transformation. [1, 10] In another presentation termed Maffucci syndrome, skeletal chondromatosis is seen in association with soft tissue angiomas. [19] No maxillomandibular involvement has been reported. [1]

The radiographic findings of a chondroma are not specific. As in our case, an irregular, radiopaque and radiolucent mottled mass may be seen. [9] However, the lesion most often presents as an irregular homogeneous radiolucent area, sometimes with fine intralaseous calcifications, generally well limited, displacing adjacent bone structures without peripheral osteocondensation. [1, 9]

In most cases, the lesion is approached with an incisional biopsy [4, 9, 10] or an exploration with an excisional. A CT scan may be useful for planning and guiding the exploration as well as for

determining the extent of the lesion. [12] Histology is characterized by a lobular arrangement of hyaline cartilage, with well-formed lacunae containing small and regular chondrocytes, [10, 12] Chondrocytes are most often mononuclear. Their nuclei are typically small, round and densely hyperchromatic. [1, 9, 21] Occasional binucleate cells may be found but are not numerous. [10] Calcifications are sometimes observed in this vacuolar chondroid substance. [18]. Enchondroma and periosteal chondroma share similar histopathological features. However, as in our case, periosteal chondromas are comparatively hypocellular than enchondromas. [22]

The differential diagnosis with chondrosarcoma is the most challenging [10]: 20% of chondrosarcomas in the cephalic region are initially considered to be chondromas. [21] Clinical and radiographic features are often of little use in distinguishing between chondroma and well-differentiated chondrosarcoma. [10] The size of the lesion is a useful characteristic. Most of described chondromas measure from 1 to 3 cm when most of chondrosarcomas are greater than 5.5 cm. [17] Additional signs of persistent, relentless pain and radiographic signs of cortical erosion and soft tissue extension are warning signs of malignancy. [16] All the clinical and radiological features of our case were in favor of a benign lesion with the exception of pain, but this was not sufficient to conclude that there was malignancy. As described below the histopathological examination can't rule out a malignancy, some clinical and radiological feature may be helpful, the most important criteria that were in favor of a chondroma are: the well limited lesion and the raw bony surface left after excision but of course checkups would've been more helpful to determine whether or not the excisional biopsy took out the entire lesion.

Even histological analysis cannot formally rule out the diagnosis of chondrosarcoma [23] because of overlapping histological features, [17] especially between chondroma and low-grade chondrosarcoma. The distinction between these lesions is based on evidence of invasion and the discovery of more than one rare mitotic figure. The presence of one of those signs indicates a high probability of malignancy. [9] Conversely, mitotic figures are extremely rare to nonexistent in the benign chondroma. [16] Finally, some authors suggest that the pathologist should examine multiple blocks of the cartilaginous tumors because chondrosarcoma may be only focal in a larger lesion. [9]

Occasional chondroma cases may show histological features such as chondrocytes with large open-face nuclei with a visible chromatin pattern and many binucleate cells. In such cases, a radiological pattern

showing a lack of cortical destruction is an important feature that favors a benign diagnosis. [10]

The malignant transformation in chondroma is controversial. Some authors claim that all chondrosarcomas arise from pre-existing chondromas, whereas others found no evidence of a pre-existing chondroma. [10] In a significant number of cases, curettage was responsible for local recurrence. The resection with a margin of normal soft tissue and bone is preferred. [16, 17] In our case the lesion was excised. A raw cortical surface of bone was left after excision. Some authors believe that lesions diagnosed as chondromas should be excised as low-grade chondrosarcomas with 1cm peripheral margins. They consider that this approach is not overmuch aggressive given the potential for recurrence of chondrosarcoma in this area and the extreme rarity of chondromas. [12] Radiotherapy is contraindicated because the tumor is not radiosensitive on one hand, and on the other hand, because this could enhance the potential of malignant transformation [16] Follow – up is essential due to the increased rate of recurrence with malignant transformation as a possible complication. It's recommended to have checkups every 6 months for an indefinite period of time. Suspicious areas must be explored and biopsied. [9, 10, 12]

CONCLUSION

Chondroma is a challenging tumor regarding the diagnosis and the treatment. The diagnosis is based on the histopathological findings. When suspecting this diagnosis, an excisional biopsy must be performed. Chondrosarcoma has to be seriously discussed as a differential diagnosis. Clinical and radiological findings as well as histopathological features help distinguish between chondroma and chondrosarcoma. Long term clinical and radiological follow-up is mandatory since it is known for its great ability to recur with controversial possibility of malignant transformation.

COMPETING INTERESTS

The authors declare no competing interest.

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