

Case Report

Periosteal chondroma of rib at adulthood

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ABSTRACT

Periosteal chondroma is a rare, benign, slowly growing cartilaginous tumors. They are found in long and tubular bones, mostly. It occurs in second and third decades of life. Rib is an rare location to found periosteal chondroma. We demonstrates a case report of periosteal chondroma founded in the rib that belongs 44-year-old man. We also examine isocitrate dehydrogenase 1 (IDH-1) mutation of this unusually located periosteal chondroma immunohistochemically and IDH-1 mutation was determined as negative. This article is aimed to present 44 years old man's periosteal chondroma developed at rib that is an unusual location and age for periosteal chondromas.

Keywords: rib, periosteal chondroma, juxtacortical chondroma,

Introduction

Periosteal chondroma, also defined as juxtacortical chondroma, benign cartilaginous lesion which arises on cortex beneath periosteum. Periosteal chondromas are relatively uncommon tumors which form less than %2 of all chondromas. Although it can develop at any age, most of the patients in 2nd and 3rd decade of life [1]. Their common presentations include palpable, mostly painful masses and swelling [2,3]. The most common sites for periosteal chondroma are phalanges of the hands and feet and the proximal humerus [4]. This article is aimed to present 44 years old man's periosteal chondroma developed at rib that is an unusual location and age for periosteal chondromas [5].

Case Report

A 44-year-old man who presented with palpable and painful mass which appeared recently at his rib referred to our clinic. He stated that his pain at the location of mass had gradually increased and does not relieve by nonsteroidal anti-inflammatory drug (NSAIDs) which prescribed for his fibromyalgia previously. He does not have any hospital admission for this mass at his rib before. He does not have any chronic disease other than fibromyalgia that is diagnosed 2 years ago. His drug history was duloxetine and etodolac for his fibromyalgia. In his family history there is no malignancies, genetically inherited disease. The patient has a history of traffic accident in 2014 when he involved in a car accident and was brought to the emergency room. In his physical examination after trauma, pain at the right hemi-thorax was noted. Any other physical examination abnormalities were detected. He did not receive any surgical intervention; complete blood count follow-up was performed, and he was discharged from hospital after adequate period of observation. In his physical examination in our clinic, swollen and tender mass was found at his 6th rib of costal arch. His biochemical tests were in the normal range. Chest radiography was performed, and no significant change was noticed. Subsequent thorax tomography revealed a mass lesion at 6th costochondral junction arised on the surface of rib (Figure 1).

Partial rib resection surgery was successfully performed to the patient. The resection material of the right 6th costa-chondral region was sent to pathology. They

are bone tissue in the size of 3.5x1.5x2 cm and associated tissue in the size of 2.5x1.5x1 cm and tissues in the size of 3x3x0.5 cm, which were sent as a multiple fragment. Histopathological investigation of lesion was compatible with periosteal chondroma (Figure 2). Margins of resected tumor material which was send to the pathology laboratory was reported as tumor free. During his post-operative fallow up no complication was occurred, and the patient was discharged 3 days after his surgery. After partial rib resection surgery, patient's symptoms were regressed, and patient had reduced pain. By the time this article was written, 3 months later of patient's surgery, no recurrence of periosteal chondroma was encountered.

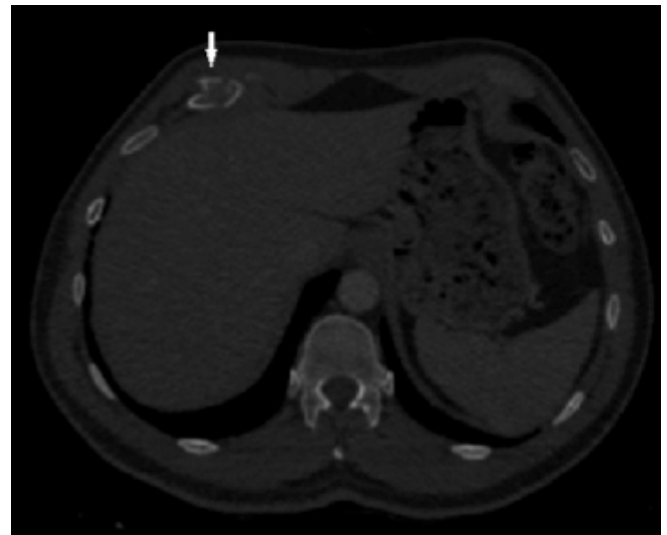


Figure 1. Axial reformation, mediastinal window of CT scan shows a lesion on the surface of rib. This lesion is radiolucent at partial places and in some regions calcification and lobules which have cartilaginous density can be seen. Note the asymmetry of bone and surrounding soft tissue relative to same rib level alignment of opposite sight. Calcification can be seen in the medullary cavities of lesion.

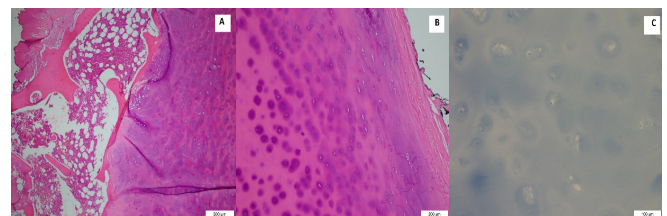


Figure 2. Low-power view showing well differentiated hyaline cartilaginous tumoral mass associated with the bone cortex (H-EX40) (A), low - power showing lobules of hyaline cartilage surrounded by a periost (H-EX100) (B), immunohistochemical IDH-1 showing negativity in chondroid tumor cells. (X200) (C).

Written informed consent was obtained from the patient for publication of his data.

Discussion

Periosteal chondromas are benign hyaline cartilage neoplasm of bone surface that arises beneath of periosteum. According to WHO they are forming less than two percent of all chondromas, and they occur with males outnumbering females by three to two. They are commonly seen in long bones. Typical location for periosteal chondroma is proximal humerus [6]. Since our case was found in unusual location and age, this case report is about to provide unique contribution to literature.

Clinically, periosteal chondromas present as palpable mass generally induce pain and swelling around [3]. Radiological findings of periosteal chondromas are scalloping of adjacent bony cortex, presence of cartilaginous matrix appearing as small dots of calcification and possible soft tissue mass component [7]. Mass caused by periosteal chondroma leads to erosion over the cortical bone seen with radiodensities. Medullary extension is not seen [8]. Gross features include encapsulated, solid masses of glistening white tissue. Periosteum overlay the tumor on its boundaries. The cortex underlies the tumor may thickened, eroded. Size of tumor is generally less than 3 cm in diameter. Histopathologically, periosteal chondroma has a well-defined border with cortex underlies. Matrix is composed of hyaline cartilage. Atypia can be seen in neoplastic cortex from mild to moderate. Although histological features resemble to other chondromas, periosteal chondromas are more cellular [2,3,8].

IDH-1 (isocitrate dehydrogenase 1) mutation has described recently in a wide variety of benign and malign cartilaginous tumors [9]. Periosteal chondromas have high frequency rate for IDH-1 gene mutation [6]. IDH-1 mutations were detected with a high percentage (6 out of 8) in the study of Amary et al. The overall frequency of IDH1/IDH2 mutations in conventional central and periosteal cartilaginous neoplasms was found to be 56%. Mutation-positive central and periosteal cartilaginous tumors occurred more frequently in the tubular bones of the hands and feet than at other anatomical 90% of the acral-based tumors revealed a mutation, compared to 53.2% of tumors in the long bones of the appendicular skeleton, and 35.1% of those in the flat bones [10]. Other study also support the finding of IDH-1 mutation in periosteal chondromas [11]. When we re-

viewed the literature it has been seen that this mutation status does not have any effect on prognosis. However, IDH-1 inhibitors may have a part in the treatment, since this mutation has a role during tumorigenesis, in the future. In this case immunohistochemical staining studies was performed towards IDH-1 mutation and result was negative.

During differential diagnosis procedure; periosteal chondrosarcoma, periosteal osteosarcoma should be considered by clinicians. Histopathological evaluation and radiography are important to distinguish and clarify final diagnosis. Periosteal osteosarcoma generally has periosteal neoplastic bone formation covering lesion and demonstrates more cytological atypia. Periosteal chondrosarcomas are larger and more cellular, also have more cytological atypia [3]. While evaluating radiology; there are obvious differences that can help us to differentiate periosteal chondroma from periosteal osteosarcomas and periosteal chondrosarcomas. Periosteal chondrosarcomas demonstrate popcorn calcification on radiography whereas periosteal osteosarcoma demonstrates perpendicular spicules of calcification [12]. Since our specimen had well defined border with hyaline matrix which has no myxoid changes within it and had no hyperchromasia, binucleation or double cells within lacune our final diagnosis was periosteal chondroma, though immunohistochemical staining towards IDH-1 mutation was negative. In addition to that, no invasion to adjacent structures and less cytological atypia supports our diagnosis of periosteal chondroma.

Treatment options for periosteal chondromas are partial resection, total resection and intralesional resection of the lesion. These treatment options seem to be effective and have low recurrence rates [13].

In conclusion, this case report reveals clinical symptoms, histopathological and radiological characteristics, as well as differential diagnosis of periosteal chondroma which was founded at unusual location. For clinicians, while evaluating mass or bone tumor on radiography, considering periosteal chondroma as one of the preliminary diagnosis leads to early and proper surgical treatment for patients. Even though more data are needed, as an etiological factor, blunt trauma to the thorax region may be associated with subsequent periosteal chondroma. Moreover, we believe that our study is about to have unique contribution to current literature by exam-

ining IDH-1 mutation in periosteal chondroma which is found in rare location and unusual age.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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Authors' contributions

CBK, TUK, HS, FECS, EA: conceived and designed the current case report, co-wrote the paper, collected the clinical data. The authors discussed the case under the literature data together and constituted the final manuscript.

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