Case Report

A Rare Case of Pediatric Osteochondroma Presenting as Hemothorax

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ABSTRACT

Isolated osteochondroma presenting as hemothorax is a rare entity. A 7-year-old boy presented with respiratory distress and diagnosed with hemothorax, and computed tomography showed osteochondroma and removal with resection of the rib was curative.

KEYWORDS: Curative resection, hemothorax, osteochondroma

INTRODUCTION

Osteochondroma is the most common benign lesion of the bones, commonly arising from the growing end of long bones. It is characterized as bony outgrowths from the surface with a cartilaginous cap. It can be solitary or multiple, with the former most commonly seen in clinical practice. Osteochondromas are benign developmental anomalies, characterized by the separation of epiphysis growth-plate cartilage from the main epiphysis. It is commonly seen on the long bones. However, the index child had an osteochondroma of rib presenting as hemothorax which is exceedingly rare, and hence, presented as a case report.

CASE REPORT

The index case was a 7-year-old boy admitted with complaints of difficulty in breathing along with left-sided chest pain of 2-day duration. There was no history of trauma or infections or any other respiratory symptomatology. The child had no swelling anywhere else in the body, and the parents were normal.

The clinical examination revealed evidence of left pleural effusion. Complete blood count within normal parameters and Mantoux test was negative, and the patient reported no history of recent contact with tuberculosis patients. Coagulation profile was also normal. Chest X-ray and ultrasound showed collection of moderate amounts of pleural fluid associated with collapse of the left lower lobe.

Diagnostic aspiration under ultrasound guidance revealed uniformly bloody fluid from the pleural cavity. Computed tomography (CT) of the chest (Figure 2) was done which revealed a speculated tumor mass over the left seventh rib near the anterior end, protruding toward the left lower lobe of the lung which is clearly evident upon three dimensional (3-D) reconstruction. Thus, a diagnosis of bony tumor causing perforative injury to the left lower lobe of the lung causing hemothorax was made.

The child was taken up for the surgery, and thoracoscopy was done initially to identify and localize the lesion and to rule out any complications. The bony spicule over the left seventh rib was resected, which was a 2 cm × 2 cm hard spicule with a smooth surface from the inner surface of the rib injuring the adjacent pleura and lung parenchyma.

The histopathological examination revealed nodular lesion comprising external fibrous tissue, middle cartilaginous tissue, and inner bony trabeculae enclosing the marrow cavity (Figure 3). There was endochondral ossification with proliferating capillaries, hematopoietic and fatty marrow elements within the lesion. The outer fibrous layer is seen continuous with the periostium of underlying bony tissue.

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These confirmed the diagnosis of osteochondroma of rib, in benign status.

**Discussion**

Being relatively common tumor of long bones, the incidence of osteochondroma of ribs is exceedingly rare, with an incidence of 1 in 50,000 only. Even rarer is the osteochondroma causing hemothorax in the pediatric age group with only <5 cases reported in previous literature. The rarity of the case was the main reason for reporting it.

Kadu et al., have previously reported a similar case of 9-year-old boy with osteochondroma of the right 6th rib. During his follow-up of 3-year period, the tumor did not increase in size or developed any complications and hence was managed conservatively.

Most of the patients are asymptomatic which makes it hard to diagnose. Only very few cases are observed with symptoms such as chest pain.

Osteochondroma starts early in childhood and grows till skeletal maturity is reached. It is asymptomatic when benign and small. The main complications include fracture of rib, vascular injury, osseous deformities, neural compression, and malignant transformation.

Our patient had a rare complication of hemothorax caused by the lesion, possibly due to chronic irritation of the chondroma spicule growing inward into the pleura, causing bleeding from pleural vessels.

The tumor peaks around the age group of 10–30 years with a 3% thoracic origin, which rises to 10% in hereditary multiple exostosis, as evident by the case report submitted by Maeda et al., where the children suffering from hereditary multiple exostosis and strong family history, eventually developed osteochondroma in the ribs at latter part of life. Hence, it is considered worthwhile to closely follow-up such patients with hereditary multiple exostosis.

Bess et al. emphasized the importance of preoperative radiological evaluation to provide optimal data about the lesions such as erosion, internal lytic areas, and destruction of adjacent structures, calcifications, and soft-tissue masses. The size of the cartilaginous cap serves as the best indicator of malignant transformation in osteochondroma.

However, according to Naidu et al., like in our index case, CT scan when coupled with reconstruction technique can help to localize the lesion, but the size...
is often underestimated because of cartilaginous cap, which can be better visualized by magnetic resonance imaging.\[8]\n
Malignant transformation of osteochondroma is very rare and has been seen only in $<1\%$ of solitary and $2\%$ of hereditary multiple exostosis cases.\[9]\n
Few other adult cases of osteochondroma of rib have been reported by Phatak et al., but all of them were asymptomatic and were incidentally found, unlike our index child who presented with hemothorax due to osteochondroma.\[4]\n
The presence of complications favored surgical excision as the mainstay of treatment unlike previously reported cases, which were asymptomatic and hence managed conservatively.

This emphasized the importance of osteochondroma as an etiological factor in hemothorax and should be given due consideration while considering a diagnosis in the pediatric age group.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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