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CASE REPORT : CAPILLARY HAEMANGIOMA OF THE RIB

General Surgery

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ABSTRACT

We present a case of a 47-year-old female, diagnosed as a rib haemangioma and underwent a surgical resection of the affected rib.

KEYWORDS

Hemangioma, Rib, Surgery.

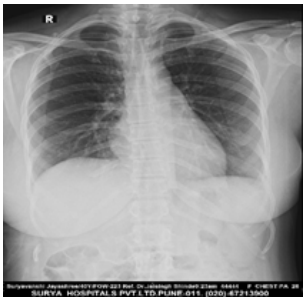
INTRODUCTION

Hemangiomas are benign vascular tumors. Bone hemangiomas have a low incidence, accounting for 1% of all bone tumors. They are predominantly found in the spine and skull while uncommonly observed in the ribs or long bones. Though congenital in origin, they present later in life as a hard swelling. Treatment is by excision with good results.

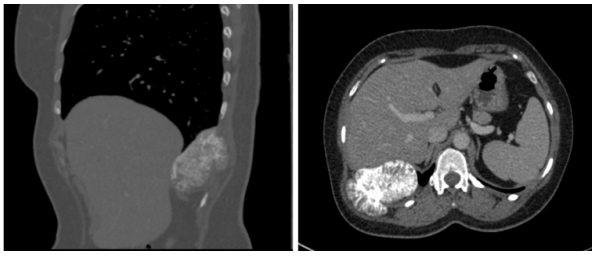
CASE REPORT

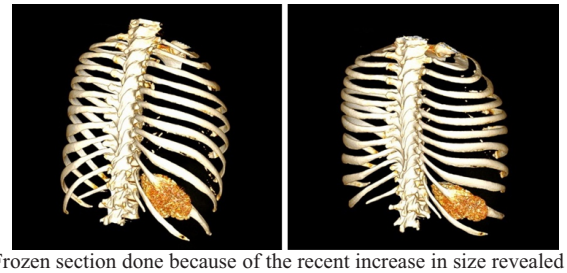
A 47 year old female presented with a painless swelling on the right posterior chest wall for the past 4 years, increasing in size since 6 months. Examination revealed a well-defined, ovoid, hard and rough surfaced swelling, of size 8 × 5 cm, along the long axis of the 10th rib. There was no significant past history of trauma, fever, or loss of weight, nor did any family member have a similar bone swelling.

Haematological laboratory investigation were within the normal range. Chest roentgenogram on anterior-posterior was inconclusive and the swelling not well appreciated.



CT scan of the chest revealed a mass in relation to the tenth rib. It was better appreciated on the reconstructed 3D CT image.

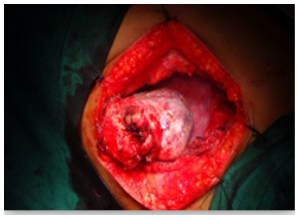




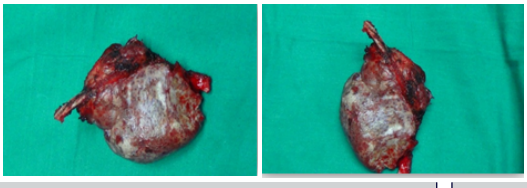
Frozen section done because of the recent increase in size revealed a vascular lesion, probably a cavernous haemangioma.

A diagnosis of haemangioma of the rib was made and patient was advised surgery.

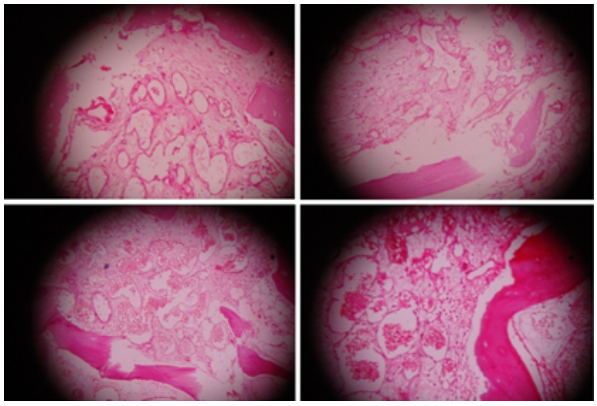
A postero- lateral thoracotomy incision was taken. The affected rib was exposed adequately with the anterior and posterior normal rib margin being dissected for 2cms on either side of the tumour and cut at that level. There were no soft tissue adhesions which enabled the tumour being removed en bloc. The lung was free, so also the diaphragm. The bleeding was average. Thoracotomy incision closed in the routine manner with an intercostal thoracic drain. Post-operative period was uneventful and patient discharged on 5th POD.



Gross appearance of the tumor was a 84 X 65 X42 mms in dimension and weighed 42gms. It had a bosselated appearance encasing the shaft of the rib, extending externally but more internally. The internal surface of the tumor was covered with smooth parietal pleura.



Microscopic examination showed a capillary-cavernous haemangioma.



DISCUSSION

Hemangioma is a neoplastic entity which arise from the blood vessels. William Hunter first described this anomaly in the mid-18th century in the context of an iatrogenic creation of arteriovenous fistulas by phlebotomists.[1] Two theories have been postulated for the etiology of hemangiomas. The widely supported theory claims that they are congenital in origin, developing from abnormal embryonic sequestration. The other theory proposes that hemangiomas are traumatic in origin.[2] They can also be inherited in a family as an autosomal dominant trait. Relatives with mild expression of the gene are often discovered. Lack of a cohesive system led to hampering of the understanding of these entities.

Costal originated haemangiomas have been reported as isolated case reports in literature.[3] Skin is by far the most common primary site of hemangioma, but any organ including the bone can be affected. Bone haemangiomas account for approximately 1% of all osseous tumors.[4] while most of these cases occur in the skull or vertebral bodies some have been reported to originate from the rib.[5] Primary tumors of the rib are uncommon, accounting for 6–10% of all primary bone tumors. According to the Japanese Bone Tumor Registration Database, from 1972 to 1994 there were only eight cases of hemangioma of the rib.[6] They have a predilection for the metaphysical/diaphysial region, but can involve the epiphysis across the joint space.

Females are a little more commonly affected than males (55:45) with an increased frequency in the 4th and 5th decade. Although haemangiomas of the bone are from birth, why they present later in life is unknown and is mostly an incidental finding. The majority of the rib hemangiomas are solitary, one-third of cases can have multiple lesions involving other bones or other internal organ like the liver. Half of them were reported as asymptomatic. Symptoms are attributed to its size and location. Occasionally they may present with pain and swelling when associated with a pathological fracture. Few of them present with rapid accumulation of one-sided pleural effusion mimicking malignant pleurisy[7]

Radiographically, bony hemangioma usually present as a lucent, well-demarcated entity. It expands and thins the cortex in flat bones, but complete cortical disruption is not present. Most cases of bone hemangioma are diagnosed by characteristic radiographic findings which reflect the formation of reactive spicule produced by the lesion. These findings are called 'sunburst appearance' for skull hemangioma, 'corduroy-like appearance' for vertebra hemangioma and 'honeycomb appearance' for rib hemangioma. In the present case it was difficult to appreciate any of such signs.[8] CT is useful in delineating the size and location of the intraosseous and extraosseous components of the tumor. 3D reconstructed images is a valuable tool in making a diagnosis. MR images typically show a lobulated or septated mass with a thin, well-defined rim of low signal intensity. MRI features of hemangiomas depend on the proportion of fat and vascularity of the lesions. Fat content reveals high signal intensity on T1-weighted MR images, whereas vascular parts show high signal intensity on T2-weighted images [9] Hemangiomas of the rib usually expansively grow and disrupt the cortex thus can be misdiagnosed as aggressive tumors or infectious processes.[10]

18F-FDG PET can detect the elevated glucose metabolism of cells, and is widely used for differentiation between benign and malignant neoplasms. Malignant lesions tend to be 18F-FDG avid and benign lesions generally show lower 18F-FDG avidity.[11] Establishing a diagnosis before surgery is difficult and, given the non-specific nature of the radiographic features, it is easy to confuse these tumours with other malignant bone lesions. Though the reports in the literature are sporadic, rib hemangioma is part of the differential diagnosis of chest wall tumors.[12] There are two types of hemangiomas on histology: cavernous and capillary. The cavernous type consists of large dilated vessels lined by a single layer of endothelial cells surrounded by a fibrous stromal layer. The capillary hemangioma, which is less common, shows numerous tortuous small vascular channels lined with epithelium.[13]

A definite histological diagnosis is essential for treating chest wall tumors. There are three types of biopsy methods for chest wall tumors: needle aspiration biopsy, excisional biopsy and incisional biopsy. Ayala and Zornosa demonstrated that the diagnostic rate of needle biopsy for primary bone tumors was 83% in malignant tumors and 64% in benign tumors. Needle biopsy is strongly recommended when there is a strong suspicion of myeloma or metastatic disease [14] Excisional biopsy can be generally applied in lesions less than 2 cm in diameter. Open biopsy has some problems that may interfere with a definitive therapy. For example, infection of the biopsy wound may delay initiating the appropriate therapy and the initial resection may interfere with identification of the area which should be removed in a later radical resection.[15] We did an FNAC considering its recent increase in size.

Rib lesions entail a variety of differential diagnoses. Because hemangiomas rarely occur in the ribs, they are often misdiagnosed. The differential diagnosis of a rib lesion includes both benign and malignant lesions.

Benign primary rib lesions include osteochondroma, fibrous dysplasia, eosinophilic granuloma and aneurysmal bone cysts. Rib lesions, such as fibrous dysplasia, eosinophilic granuloma aneurysmal bone cysts, giant cell granuloma and osteochondroma, may have characteristic imaging findings that allow a specific diagnosis. Fibrous dysplasia presents as a painless, expanding, lytic area in ribs. Solitary plasmacytoma is a rare tumor that is associated with latent systemic disease in the majority of affected patients. Plasmacytoma and multiple myeloma are typically seen as well-defined, punched-out lytic lesions with associated extrapleural soft-tissue masses, similar in appearance to most metastatic lesions. Aneurysmal bone cysts occur as expansile lesions with well-defined inner margins on roentgenograms. Fluid levels are commonly seen in aneurysmal bone cysts. Thoracic giant cell tumors often arise in subchondral regions of the flat and tubular bones of the chest wall.[16]

Grossly the lesion is characteristically hard. In few cases there is a central softening which pulsates, confusing it for a bone aneurysm. Though the tumour breaks through the cortex, the periosteum is never breached. However if you incise the lesion during surgery, bleeding can be uncontrollable. Ossification is common to nearly all haemangiomas, though rarely they undergo cystic changes.

The histopathological diagnosis of a haemangioma is not difficult in an en bloc specimen. However, diagnosis from biopsy or tissue curettage is challenging to the pathologist. This is due to the destructive nature of these procedures, disrupting thin-walled blood vessels and resulting in histological sections showing non-diagnostic empty spaces with scattered bone trabecular. This causes hindrance in the pre-operative diagnosis of haemangioma by any invasive procedure. Histologically, haemangiomas can be classified as cavernous, capillary, venous or mixed, depending on the type of vascular involvement. Cavernous haemangioma is the most common type in the peripheral bones and accounts for up to 50% of all cases reported. They are predominant in the medullary and the intracortical portion of the bone and are not yet reported in the subarticular epiphyseal bone. Pure capillary haemangioma accounts for 10% of all these types as reported in the literature. The other varieties are rare. Haemangiomas are slow growing and malignant degeneration is virtually unknown.

The treatment of solitary hemangiomas is surgical excision, but large lesions pose a threat of bleeding profusely during surgery. Vertebral hemangioma embolization is done by trans-arterial, percutaneous or direct intraoperative by onyx (ethylene vinyl alcohol polymer) and alcohol respectively. Temporary embolization using gel foam in rib

hemangioma causes shrinkage in size and vascularity of the lesion easing the excision of the lesion. Recurrences after resection have been reported after treatment of very extensive lesions with deep involvement of chest wall tissues.[17] Other management options for symptomatic hemangiomas include radiotherapy. Medications used to treat hemangioma include glucocorticosteroids have been used to slow the growth of a hemangioma. Other medical therapies have included alpha2a-interferon, vincristine and the beta blocker propranolol. Interferon has been associated with serious side effect. Vincristine has generally been reserved for use in patients with endangering hemangioma. Propranolol is new in the treatment of hemangioma. Laser therapy is used to treat superficial hemangioma.[18]

CONCLUSION

The aetiology of haemangiom of the rib is yet to be delineated, although the majority of opinions suggest its origin to be congenital Rib hemangioma is an uncommon primary benign tumor. Atypical lesion and its rarity cause uncertainty in its diagnosis, diagnosed radiologically. Haemangiomas should be included in the differential diagnoses of an osteolytic and expansile lesion of the ribs on radiology, especially in asymptomatic or mildly symptomatic patients. Surgical excision of the haemangioma is the treatment of choice and is considered definitive. Alternative treatments, with drugs, arterial embolization etc are promising but remain unproven. Our case highlights this rare entity treated by surgical excision.

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