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Intraosseous hibernoma in the rib

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Abstract

A 64-year-old man was admitted with paroxysmal left-side thoracic pain. CT scan showed an irregular appearance, expansile, osteolytic lesion of the left seventh rib. Wide *en bloc* excision of the tumour was performed. Macroscopic examination showed that a 3.5 cm \times 3.0 cm \times 3.0 cm solid lesion with destruction of bone. Histological examination showed that the tumour cells were arranged in plate shaped and interspersed between the bone trabeculae. Mature adipocytes were noted in the tumour tissues. The immunohistochemical stainings showed that the vacuolated cells were positive for S-100 protein and negative for CD68 and CD34. These clinicopathological features were consistent with intraosseous hibernoma.

Keywords: Hibernoma • Rib • Surgical resection

INTRODUCTION

Hibernomas are rare benign soft tissue tumours, which originate from the brown adipose tissue residue in the foetus. They occur predominantly in thigh, buttocks and scapular area. They are rarely found in an intraosseous. It has been reported that hibernomas occur in vertebral body, ischiopubic ramus and iliac crest [1]. We report an exceptionally rare case of intraosseous hibernoma within the rib.

CASE REPORT

A 64-year-old man presented with paroxysmal left-side thoracic pain for 2 months. She had no history of chest trauma and oncology. No abnormal findings were found in physical examination.

Chest CT scan revealed an irregular appearance, expansile, osteolytic lesion, involving the left seventh rib, it located on the medial side of the rib and next to the lung tissue. Calcification was not observed inside the mass. The CT value was about -51 to 26 U. *En bloc* resection was performed. The incision was located in the posterior axillary line of the left seventh rib. Wide *en bloc* excision was performed, including the tumour with 3-cm margins on both sides along with the seventh rib, sixth and seventh intercostal muscles and parietal pleura. The thoracic wall was reconstructed without patch. There was no postoperative complication. The patient was discharged on the fourth postoperative day.

Macroscopic examination showed that the lesion was solid with a grey-yellow colour and bone destruction, measuring $3.5 \text{ cm} \times 3.0 \text{ cm} \times 3.0 \text{ cm}$. Histological examination showed that the tumour cells were arranged in plate shaped and interspersed

between the bone trabeculae (Fig. 1A). The cytoplasm was characterized by eosinophilic and granules. The nucleus was centrally placed, small, round and hyperchromatic. Mature adipocytes were noted in the tumour tissues (Fig. 1B). The immunohistochemistry (IHC) showed that the vacuolated cells were positive for S-100 protein and negative for CD68 and CD34. It conforms to the pathological characteristics of hibernoma. The patient was followed up by CT every 6 months to 1 year, and no recurrent tumour was found after 2 years. The written informed consent was obtained from the patient.

DISCUSSION

Hibernomas are an uncommon benign tumour originating from remnants of brown adipose tissue [2]. They often occur in the thigh, upper trunk and neck. To our knowledge, intraosseous hibernoma of rib is vanishingly rare in the literature. The pathogenic mechanism of hibernoma has not been elucidated. Hibernomas occur most commonly in adults at 40-70 years of age and more common in men [3]. Intraosseous hibernomas are mostly asymptomatic and detected incidentally on image examination. Hibernomas present clinically as an asymptomatic soft tissue mass, and symptoms are present when they compresses adjacent structures. This tumour was located at the lower margin of the rib and near the parietal pleura. The preoperative pain of this patient may be due to compression of the local periosteum or pleura by the intraosseous hinernoma of rib.

CT scan is value to demonstrate the characterization of intraosseous hibernomas in ribs. Such as heterogeneous sclerotic lesions with trabecular thickening and filled with tissue as dense

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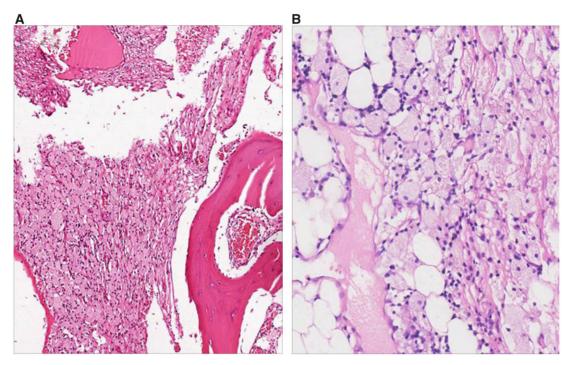


Figure 1: (A) Histologic of the intraosseous hibernoma of the rib: the tumour cells interspersed between the bone trabeculae (\times 10). (B) The tumour consisted of both polygonal cells with eosinophilic granular cytoplasm, and multivacuolated adipocytes in the tumour tissues (\times 20).

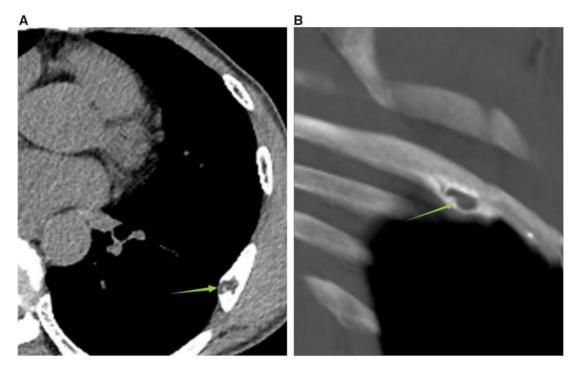


Figure 2: (A) CT showing lesion with a expansile bone and focal cortical disruption within the rib. Fat-dense areas in the lesion. (B) The location of the intraosseous hibernoma was visualized by three-dimensional reconstruction of the rib.

as fat. CT scan also provides information about the size and location, the extent of cortical destruction and the presence of infiltration to pleura or soft tissue. CT is generally useful for percutaneous biopsy and making operative plan and follow-up evaluation after surgery [4]. In our case, CT three-dimensional reconstruction was performed by us, which clearly show the site and help to make operative plan (Fig. 2B). Because if brown adipose tissue in a hibernoma is activated, it could affect the body's energy balance [2], Significant weight loss was also described in rare cases and was attributed to excessive thermogenesis by the hibernoma [5]. The mainstay of treatment is surgical excision. Surgery must consist of wide resection with tumour-free margins. In our case, The hibernoma was underwent surgical resection with 3-cm free margins, which was in agreement with the standards reported in literatures that 2- to 3-cm free margins for rib tumours to be considered sufficient [4].

IHC is specific for the pathological diagnosis of intraosseous hibernoma. IHC features of hibernoma highlights that the vesicles in the cytoplasm of the cells were positive for S-100 and negative for CD68 and CD34 [1].

The differential diagnosis of intraosseous hibernoma of rib may include benign tumours such as fibrous dysplasia, osteochondroma, aneurysmal bone cyst and haemangioma or metastatic and primary malignant tumours such as chondrosarcoma, osteogenic sarcoma, myeloma and Ewing sarcoma [1].

Intraosseous hibernoma of rib is a rare and benign tumour. The imaging features of intraosseous hibernoma are usually non-specific, but the immunohistochemical characteristics are specific. Hibernoma should be included in the differential diagnosis of rib tumours. The recurrence rate is very low. Complete surgical excision for definite diagnosis is required.

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Data availability

The data underlying this article are available in the article and in its online supplementary material.

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