Chest wall resection for multifocal osseous haemangioma

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Abstract

Intraosseous haemangioma is a rare and benign primary tumour of the bone. We report the case of a 76-year old woman who presented the exceptional condition of multifocal cavernous haemangiomas involving the spine and the ribs, requiring spinal and chest wall resections to confirm the diagnosis and treat the symptoms.

Keywords: Chest wall • Multiple haemangioma • Surgical resection

CASE REPORT

A 76-year old woman consulted our thoracic surgery clinic for chronic thoracic pain. She declared no past medical history. She had undergone the surgical resection of a symptomatic cavernous haemangioma of the fourth lumbar vertebrae 3 years before. Over the last year, she complained of worsening intercostal paraesthesia and increasing shortness of breath. Over the last weeks, she reported some episodes of tachycardia. Physical examination found a left chest wall mass. Chest X-rays showed the presence of an osteolytic tumour on the fifth rib on the left side, and a smaller tumour on the fourth rib on the right side (Fig. 1A). Chest computed tomography (CT) confirmed the presence of two osteolytic tumours of the ribs. The first tumour was 10 cm in diameter, spread over the fourth to sixth ribs and reached the pulmonary artery into the fissure on the left side. The second tumour was 3 cm in diameter, and was limited to the fourth rib on the right side (Fig. 1C and D). Positron emission tomography (PET)-CT showed a non-significant hypermetabolism of both tumours. Measured respiratory function was normal [Forced Expiratory Volume in 1 second (FEV1) 1.59 l; 96%].

The case was discussed during a thoracic tumour board conference. As the patient was symptomatic, the mass was bulky, the diagnosis was unknown and the surgical resection was expected to be complete, a surgical management of the left-sided tumour was decided. Surgical exploration revealed that the lesion was abutting the lung in the fissure without invading it. The patient underwent a left en bloc parietectomy of the fourth, fifth and sixth ribs, and a chest wall reconstruction using two titanium bars (Stratos®, MedXpert, Strasbourg, France) reinforced with an endothoracic non-resorbable prosthetic mesh (Mersuture®, Ethicon, Issy-Les-Moulineaux, France). The blood losses were minimal, and the postoperative course was uneventful. The patient was discharged home on postoperative day 8 with a complete recovery of the left lung volume (Fig. 1C). Pathological examination of the surgical specimen showed that the bone was blown, but not destroyed, by a well-circumscribed lesion composed of multiple thin-walled blood vessels (Fig. 2A and B). The lesion was lined by the costal periosteum, and the resection was complete. The pathological slides were compared with those of the spinal resection performed 3 years before, and the findings were similar. The final diagnosis was therefore multifocal intraosseous cavernous haemangiomas involving the spine and ribs.

The chronic thoracic pain completely resolved postoperatively. The patient benefited from a close follow-up of the right chest wall tumour. After 30 months of follow-up, no additional treatment was performed, and the patient is still asymptomatic.

DISCUSSION

Intraosseous haemangioma accounts for 1% of primary bone tumours, and occurs more frequently in the spine and skull. Inversely, rib tumours represent 6–10% of the primary bone tumours [1]. Aetiologies can be benign (fibrous dysplasia, osteochondroma, aneurysmal bone cyst, eosinophilic granuloma and haemangioma) or malignant (chondrosarcoma, osteogenic sarcoma, myeloma, Ewing sarcoma family tumours). Between 60 and 80% of the primary rib tumours are malignant [1]. Among benign tumours, intraosseous haemangioma may be unifocal or more rarely multifocal, involving the same or different bones, therefore mimicking metastatic disease [2]. Multiple rib haemangiomas have not been reported to date.

The clinical presentation of intraosseous haemangioma is highly variable according to the size, number and location of the tumour. A rib haemangioma is usually asymptomatic. However, large tumours may account for neuropathic pain and dyspnoea. Pleural effusion and thoracic outlet syndrome have also been described. A malignant origin should be suspected in case of rapid growth or invasive radiological features [3].

On CT imaging, the classical corduroy and sunburst appearance is common for axial lesion but extremity lesions may have a coarse
trabecular bone pattern or soap bubble appearance, often with osteolysis. On pathology, haemangiomas are thin-walled vascular tumours expressing the endothelial marker CD-31, but not the lymphatic marker podoplanin [4]. Atypical radiological and histopathological presentations may hinder diagnostic determination. Furthermore, as clinical and radiological signs lack specificity, some authors advocated for a preoperative percutaneous biopsy before complete surgical resection of the tumour [5].

In our case, the slow growth over 3 years and the radiological aspect suggested a benign primary rib tumour. Percutaneous biopsy can be associated with bleeding in case of benign tumour, or seeding in case of malignant tumour. We, therefore, decided to perform a complete first-line surgical resection. Despite the lack of prospective evidence, we feel that direct reconstruction of the ribs using titanium bars and non-resorbable prosthetic mesh allowed early mobilization with physiotherapy [5]. The prognosis of patients with haemangioma of the rib is excellent, with no recurrence after complete surgical resection [1–5]. In our case, a prolonged follow-up has been organized, to allow early diagnosis and prompt surgical resection of the right-sided lesion if it becomes symptomatic, or if the speed of growth is worrisome.

Conflict of interest: none declared.

REFERENCES