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Case report

THORACIC OUTLET SYNDROME SECONDARY TO A CAVERNOUS HEMANGIOMA OF THE FIRST RIB

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Abstract

Bone hemangiomas are slow-growing benign tumors, with rib involvement being exceptionally rare. Thoracic outlet syndrome caused by a bone neoplasm of the first rib is even more uncommon, with only a few cases documented in the literature. We present the case of a patient with a clinical diagnosis of neurogenic thoracic outlet syndrome secondary to a bone tumor of the first rib, treated using a combined approach of video-assisted thoracoscopy and posterior access for resection. To our knowledge, this is the second reported case in the literature of a young man with thoracic outlet syndrome secondary to a cavernous hemangioma of the first rib.

Key words: bone neoplasms, cavernous hemangioma, thoracic outlet syndrome

Resumen

Síndrome del opérculo torácico secundario a un hemangioma cavernoso de la primera costilla

Los hemangiomas óseos son tumores benignos de crecimiento lento, y el compromiso de las costillas es extremadamente raro. El síndrome del opérculo torácico secundario a una neoplasia ósea de la primera costilla es aún más inusual, con muy pocos casos descritos en la literatura. Presentamos el caso de un paciente con diagnóstico clínico de síndrome del opérculo torácico neurogénico secundario a un tumor óseo de la primera costilla, tratado mediante un abordaje combinado de videotoracoscopia y acceso posterior para su resección. En nuestro conocimiento, este es el segundo caso reportado en la literatura de un hombre joven con síndrome del opérculo torácico secundario a un hemangioma cavernoso de la primera costilla.

Palabras clave: neoplasias óseas, hemangioma cavernoso, síndrome del operáculo torácico

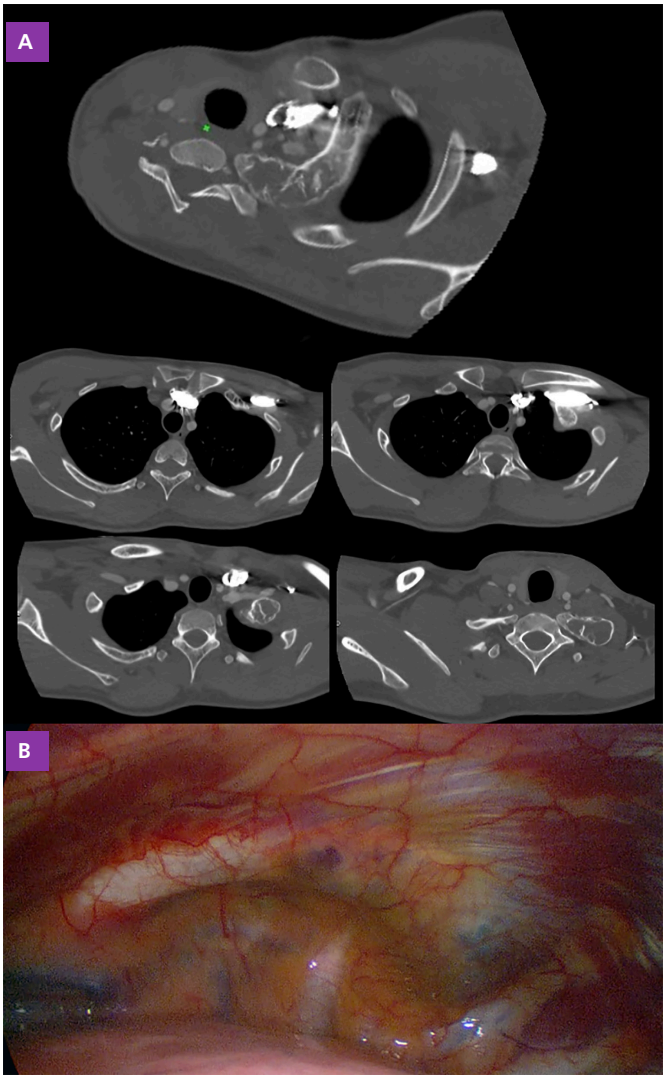
Bone hemangiomas are slow-growing benign tumors. They represent approximately 1% of all bone tumors and generally occur in women between the fourth and fifth decades of life. The most frequently affected bones are the vertebrae and the skull[1]. Rib involvement is exceptional and usually asymptomatic, often discovered incidentally during imaging studies performed for other reasons. Thoracic outlet syndrome (TOS) due to a bone neoplasm of the first rib is even more uncommon, with very few cases reported in the literature. To our knowledge, this is the second reported case in the literature of a young man with thoracic outlet syndrome secondary to a cavernous hemangioma of the first rib.

Case report

We present an 18-year-old male with no past medical history who had a one-year history of left pectoral edema associated with paresthesias and pain in the left shoulder and arm. The symptoms were exacerbated by physical activity. After several visits to the emergency department, and with a normal physical examination and soft tissue ultrasound results, the Department of Orthopedics and Traumatology requested an MRI of the shoulder to better evaluate the soft tissues and the glenohumeral joint. The MRI revealed an osseous abnormality at the thoracic outlet, suggesting the need for a chest CT and a consultation with a thoracic outlet specialist.

The CT described an heterogeneous expansion of the left first rib with mild cortical involvement and a soft tissue component compressing elements of the thoracic outlet (inferior primary trunk of the brachial plexus and subclavian artery) suggestive of indeterminate origin but appearing benign (Fig. 1A). The main diagnostic consideration was fibrous dysplasia; however, other pathologies could not be ruled out. With a clinical diagnosis of neurogenic thoracic outlet syndrome secondary to a bone tumor of the first rib, it was decided to perform resection of the rib. A combined approach (video-assisted thoracoscopy and posterior access) was used for the resection. The first stage involved thoracoscopy (Fig. 1B), during

Figure 1. | A: Preoperative thoracic CT scan. Heterogeneous expansion of the left first rib with mild cortical involvement and a soft tissue component compressing elements of the thoracic outlet (inferior primary trunk of the brachial plexus and artery). B: Thoracoscopic view of the left first rib



which, despite the size of the rib, the sternal end of the rib, the anterior scalene muscle, and the middle scalene muscle were successfully sectioned, leaving the rib fixed only by the joint with the vertebra. The inferior primary trunk of the brachial plexus was significantly adhered to the rib, making it difficult to separate. As it was not possible to detach the rib via thoracoscopy due to its large size, a posterior approach was then performed to complete the separation from the vertebral joint. Despite having performed the posterior approach, we were unable to achieve a complete resection of the rib because with each maneuver exerted on the joint, the patient experienced episodes of asystole. Nevertheless, the rib was almost completely resected. The patient went uneventfully and was discharged on the 6th postoperative day. Pathology reported a proliferation consisting of medium and large caliber vascular structures with abundant intraluminal red blood cells compatible with cavernous hemangioma (Fig. 2A, B, C). A month after the surgery, a CT scan of the chest was performed to evaluate the amount of rib that remained unresected. The study described a minimal residual rib (Fig. 2D). The patient improved the symptoms secondary to nerve compression. The patient gave consent for the anonymous publication of his case.

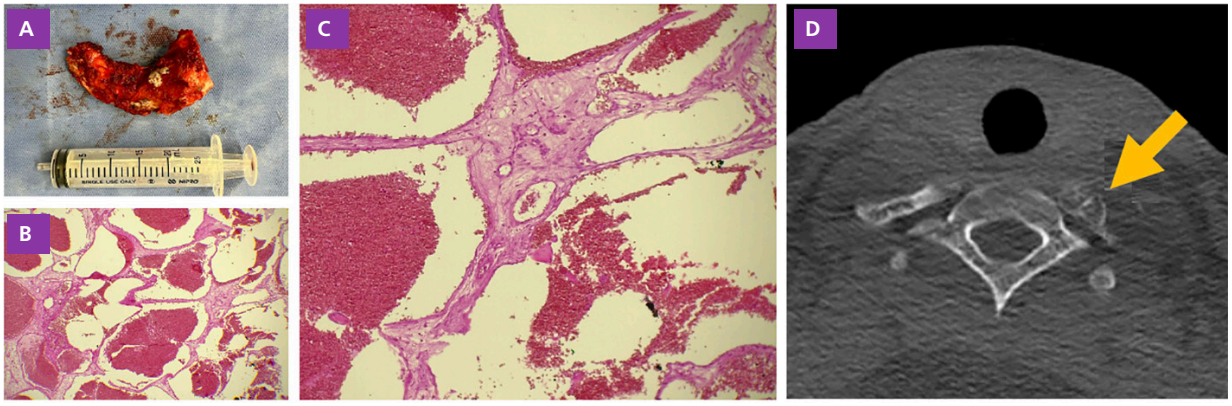
Discussion

Thoracic outlet syndrome secondary to a bone tumor is extremely rare. In 1991 Melliere et al. described that the histological type of bone tumor most frequently affecting the first rib and causing thoracic outlet syndrome was osteochondroma[2]. Among the two published cases of hemangioma of the first rib, only one resulted in venous TOS due to compression of the subclavian vein[3,4].

Imaging diagnosis is challenging because the radiographic characteristics are not pathognomonic and are often shared with other bone tumors. Tomographically, it typically presents as an expansible lesion with hyperlucent areas, without cortical involvement, and with fine trabeculations inside[5].

The resection of the first rib secondary to bone tumors, as described in all case series, has traditionally been performed through open approaches (transaxillary or supra/infraclavicular). Since we have been resecting the first rib by videothoracoscopy in thoracic outlets due to compression of neurovascular structures at the costoclavicular space since 2017[6,7], we decided to attempt an initial thoracoscopic approach because the anterior end of the rib was relatively healthy and less thickened. The initial goal was to advance as much as possible through this approach and then if needed, through a posterior approach, complete the disarticulation of the rib. Besides surgery, other therapeutic alternatives for this entity include radiotherapy, transarterial embolization, and direct intralesional ethanol injection[8,9]. However, for tumors affecting the first rib, preoperative diagnosis (biopsy) is exceedingly difficult due to limited acces due anatomical factors and, moreover the challenging nature of bone tissue processing for patholo-

Figure 2. | A: Macroscopic specimen of the resected left first rib. B, C: H&E photos (40x and 100x) showing proliferation of medium and large caliber vascular structures with abundant intraluminal red blood cells. D: Postoperative thoracic CT scan. Residual rib in contact with the vertebral joint that could not be resected



gists. In addition, imaging methods do not provide pathognomonic characteristics, making it impossible to rule out malignancy. Consequently, we believe that surgical resection is essential both for establishing a definitive diagnosis and for alleviating or resolving the symptoms caused by the bone tumor.

In conclusion, TOS secondary to bone neoplasms, particularly hemangiomas of the first rib, remains a rare and challenging clinical entity. Given the difficulty in achieving a definitive preoperative diagnosis through imaging or biopsy, surgical resection not only serves a therapeutic purpose but is also crucial for histopathological evaluation to exclude malignancy. Minimally invasive approaches such as videothoracoscopy, when feasible, provide a safe and effective alternative to traditional open techniques, allowing for reduced morbidity and faster recovery. However, a multidisciplinary approach involving careful preoperative planning is essential to optimize outcomes in these complex cases.

Conflict of interest: None to declare

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