Case report

An unusual cause of hiccup: costal exostosis. Treatment by video-assisted thoracic surgery

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Abstract

The described case is of a 15-year-old boy who presented with a persistent hiccup and repeated episodes of left-sided chest pain. At computed tomography scan an exostosis originating from the costo-chondral junction of the left 4th rib was seen. The tip of the exostosis reached the external surface of the pericardium. Removal of a 2 cm rib segment including the implantation basis of the exostosis was achieved by video-assisted thoracic surgery. Symptoms disappeared after surgery. This report shows an exceptional symptom of costal exostosis.

Keywords: Exostosis; Osteochondroma; Symptoms; Hiccup; Video-assisted thoracic surgery

1. Introduction

Exostosis of ribs represent an unusual clinical condition [1–6]. It is generally asymptomatic and few cases of intrathoracic complications have been reported [1–6]. They include hemothorax, diaphragm or pericardial laceration as well as visceral pleura injury. We report a case of a costal exostosis responsible for repeated episodes of hiccup and describe its surgical treatment by video-assisted thoracic surgery.

2. Case report

A 15-year-old boy was seen in the out patient clinic for left sided chest pain of 3 week duration. Frequent episodes of hiccup (several times a week) were also present for the last 4 months. There was no event apparently initiating these episodes and they resolved spontaneously few minutes after the onset. His past medical history was unremarkable. On the other hand a history of multiple familial exostosis was known. Plain chest X-ray revealed a left parahilar opacity. Thoracic computed tomography (CT) scan showed an exostosis originating from the intrathoracic aspect of the anterior arc of the left 4th rib. The exostosis frankly penetrated into the homolateral hemithorax toward the mediastinum for 4 cm approximately and apparently an adhesion relied its tip to the pericardium. No other anomaly was evident. Lung function tests were strictly normal. The symptomatic character of this exostosis necessitated surgery.

Intrathoracic exploration was carried out by video-assisted thoracoscopy. A 10.5 mm thoracoport was inserted in the 7th intercostal space is the posterior axillary line. The exostosis originated from the costo-chondral junction of the 4th rib and its tip was close to the pericardium. Surprisingly the phrenic pedicle had been hooked by the tip of exostosis thus loosing its habitual contact with the pericardium over approximately 3 cm. Several other millimetric exostosis originating from the costo-chondral junctions of other ribs were also present. All were smooth surfaced.

Video-assisted thoracoscopy allowed the precise individualization of skin projection of implantation basis of the exostosis. So an elective 2 cm incision was carried out at this level. The 4th rib was freed from upper and lower intercostal spaces over a length of 4 cm approximately under thoracoscopy control. It was subsequently cut some millimeters medially and laterally to implantation basis of the exostosis, thus resulting in an approximately 3 cm rib
resection. After bone section, mobilization of rib fragment allowed easy separation of exostosis from phrenic pedicle under direct visual control; section of some adhesions was necessary to this purpose. Parietal reconstruction was achieved by approximation of pectoralis major fibers and the thoracoport incision was employed for chest drainage positioning.

Postoperative course was uneventful and the patient was discharged on the 4th post-operative day after radiological control of properly functioning of left diaphragm. Symptoms had completely disappeared and cosmetic results were excellent.

Pathologic examination of the partially resected rib specimen revealed a smooth surfaced spiny bony projection from its middle part of 2 cm long and 0.5 cm width (Fig. 1). Histologically, the lesion corresponded to a bony outgrowth, capped by cartilage, on the surface of the bone. This cartilage cap, covered itself by periosteum, showed columns or clusters of evenly distributed chondrocytes, sometimes being binucleated, but mainly with no cytonuclear atypia (Fig. 2). Medullary bone showed fatty or hematopoietic marrow, with many calcified debris. Periosteum showed secondary synovial chondrometaplasia and calcified debris. These aspects were consistent with costal osteochondroma.

3. Comment

Primary tumors of the thoracic cage account for 7–8% of all primary skeleton tumors, thus being rarely encountered in clinical practice [7]. Osteochondromas represent a small percentage of thoracic cage tumors: in the series by Teitelbaum [8] dealing with 90 primary tumors of thoracic skeleton, there were four osteochondromas, only one arising from a rib.

Osteochondromas originates from a separation of a portion of epiphyseal growth plate cartilage from the main epiphysis. This results in the laying down of an abnormal bony spur (exostosis). Ribs are involved at the costochondral junction or at the vertebral end. Exostosis may be solitary or multiple: in this last case, hereditary is the rule. Hereditary multiple exostosis is an autosomal dominant condition characterized by the presence of numerous osteochondromata which usually appear in childhood and enlarge until puberty.

Rib exostosis is completely asymptomatic in most cases: however they may present as a swelling in the chest wall or give rise to pain (as observed in our case). Few cases of hemothorax complicating a rib exostosis have been reported: injury of pleura [1], diaphragm [2], heart [3] and possibly lung [4] were at the origin of this complication.
Exceptional complications of rib exostosis include bronchiectasis [5] and spinal cord compression [6].

Rib exostosis may be difficult to recognize on standard chest X-ray. Thoracic CT scan is usually useful in establishing the diagnosis and evidencing possible associated intrathoracic abnormalities.

In the absence of symptoms, no treatment is generally proposed: otherwise surgical removal of the exostosis is necessary. Standard thoracotomy has been employed in almost all the cases described so far. Simansky and associates have recently reported the resection by video-assisted thoracoscopy of a rib exostosis complicated by diaphragmatic injury [2]. They employed a laminectomy rougeur passed through a thoracoscopy incision to remove the exostosis. In our patient a different approach was used: the camera was used to recognize the rapport of exostosis with the phrenic pedicle and to identify the most appropriate site of the 2 cm skin incision employed for segmental rib resection. As the risk of malignant transformation is reported to be 1–2% [9], we performed a segmental rib resection rather than mere excision of exostosis in order to obtain wide margins thus preventing recurrence and possibly malignant transformation. Once rib resection carried out, the tip of the exostosis could be freed from the phrenic pedicle through the same incision under direct visual control. Functional and cosmetic results were also excellent.

Hiccup may represent an unusual complication of rib exostosis. Video-assisted thoracic surgery allows a satisfactory management of this condition.

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References