CASE REPORT

Thorn-like costal osteochondroma presenting as hemothorax in an adult

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Summary Exostosis, also known as osteochondroma, results from a disorder of the growth-plate in which bone grows from the growth axis to form an irregular projection. This abnormality most commonly occurs around the femur, scapula, humerus, and ribs. Hemothorax and diaphragmatic rupture are known complications of exostotic growth, but they are rarely reported. Here we present a case, possibly the first one in Taiwan, with pin-like inward-facing exostosis in a 20-year-old man with hereditary multiple exostosis that caused hemothorax and required surgical intervention. Exostosis is asymptomatic in most cases and requires no treatment. However, when it causes bursitis or local entrapment of vessels, or when tendons or nerves are involved, surgical resection becomes necessary.

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1. Introduction

Hereditary multiple exostoses (HME) is an autosomal-dominant condition characterized by exostoses that appears in different parts of the skeleton. Lesions mainly occur in infants and children, but they usually cease to enlarge once the child reaches puberty. Rarely, the exostosis may cause injury to either the diaphragm or to the pleura, resulting in hemothorax. We will describe here an adult patient with HME who presented with thoracic hemorrhage caused by an inward-facing thorn-like exostosis without coverage by a cap of cartilage from its surface.

2. Case report

A man who was 20 years of age was admitted to the hospital due to sudden onset of left chest pain and dyspnea for...
several hours. A chest radiograph demonstrated a large pleural effusion or hemothorax over the left lobe (Fig. 1), and a chest tube was inserted that drained approximately 1000 ml of blood. The patient was sent to the intensive care unit for close observation, and then a chest computed tomography (CT) scan was taken once the vital signs were stable 2 days later; the scan revealed a broad-based bony excrescence over the right scapular body and right 7th rib and another pin-like bony excrescence over the left 10th rib with indentation of the left renal capsule (Figs. 2 and 3). Furthermore, a renal echogram showed multiple exophytic bony excrescences over the right anterior rib with indentation of the anterior liver surface. There was another long bony spur, approximately 3–4 cm in length, with indentation of the left renal capsule and no penetration into the left renal parenchyma.

During a left 9th to 10th intercostal space posterolateral thoracotomy, an osteochondromatous spur was found on the anterior arc of the left 10th rib near the costochondral junction. It measured 2 cm in length and presented with

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**Figure 1** Chest radiograph demonstrating a large pleural effusion or hemothorax over the left lobe.

**Figure 2** Chest computed tomography scan reconstruction that revealed a pin-like bony excrescence over the left 10th rib inward into the thoracic cage.

**Figure 3** Chest computed tomography scan (two slices) that revealed a pin-like bony excrescence over the left 10th rib with indentation of left renal capsule.

**Figure 4** Osteochondromatous spur on anterior arc of left 10th rib near the costochondral junction without coverage by a cap of cartilage.
a thorn-shaped tip, which was not covered by the cap of cartilage from its surface and eroded the diaphragm. The affected diaphragm was inflamed by a laceration. There was no active bleeding during the operation. The exostosis and the abnormal segment of the rib were excised (Fig. 4). The postoperative course was uneventful; there were no complications such as diaphragmatic rupture or recurrent bleeding, and the patient was discharged 7 days after the operation. Pathologic examination of the resected specimen (Fig. 5) revealed an outgrowth of mature bone covered by a well-differentiated cartilaginous cap that surrounded a subperiosteal bony projection. The cells in the cartilaginous cap resembled those of normal hyaline cartilage. The pathologic diagnosis was costal osteochondroma.

3. Discussion

HME represents a small percentage of thoracic cage tumors. Except for an inward-facing exostosis in the thoracic cage, most solitary HMEs are discovered before adulthood and usually are nontender, painless, slow-growing masses. However, when an exostosis is either large or occurs at certain critical anatomic sites, it can lead to symptoms with potential complications. An exostosis becomes symptomatic on account of mechanical irritation to soft tissues, and involvement of a rib can rarely lead to pleural irritation and hemothorax. Few cases of hemothorax complicating a rib exostosis, however, have been reported. The complication may be associated with injury to the pleura, diaphragm, heart or lung. Shearing of the pleura or the diaphragm by the sharp margins of the intrathoracic exostosis is potentially lethal. Diaphragmatic rupture, bowel obstruction, pericardial effusion, repetitive chest infections and loculated empyema have also been reported. In our case, shearing of the diaphragm by the sharp margins of the intrathoracic exostosis causing hemothorax should be considered.

To the best of our knowledge, with regard to thorn-like exostoses without coverage by a cap of cartilage, only four cases have been reported in past and the only known adult case as we reported. The diagnosis was suggested by CT scan and confirmed by surgery with pathologic finding.

Thorn-like exostosis not covered by a cap of cartilage is very rare. For symptomatic lesions, or with impending symptoms, such as an inward growing rib exostosis as reported here, surgical resection is the most appropriate treatment.

References