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Case Report

# Rib spur causing a hemothorax, pneumothorax, and diaphragmatic injury in a pediatric patient ☆

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ABSTRACT

Pleural effusion is a relatively common condition encountered in the pediatric emergency department. Evaluation of pleural effusion in the emergency department typically includes advanced imaging such as computer tomography or ultrasound, as well as diagnostic thoracocentesis. We report a case of a 10-year-old female with a rib spur at the anterolateral left sixth rib that caused a hemothorax, pneumothorax, and diaphragmatic injury. The patient underwent video-assisted thoracoscopic surgery and resection of the rib spur. The procedure was well-tolerated without any complications.



## Introduction

Pleural effusion is a relatively common condition encountered in the pediatric emergency department (ED). Studies have shown the most common etiology of these effusions is infection, followed by malignancy, renal disorders, trauma, and heart failure [1–3]. Evaluation of pleural effusion in the ED typically includes advanced imaging such as computer tomography (CT) or ultrasound (US), as well as diagnostic thoracocentesis. Here, we report an unusual case of spontaneous hemothorax, pneumothorax, and diaphrag-

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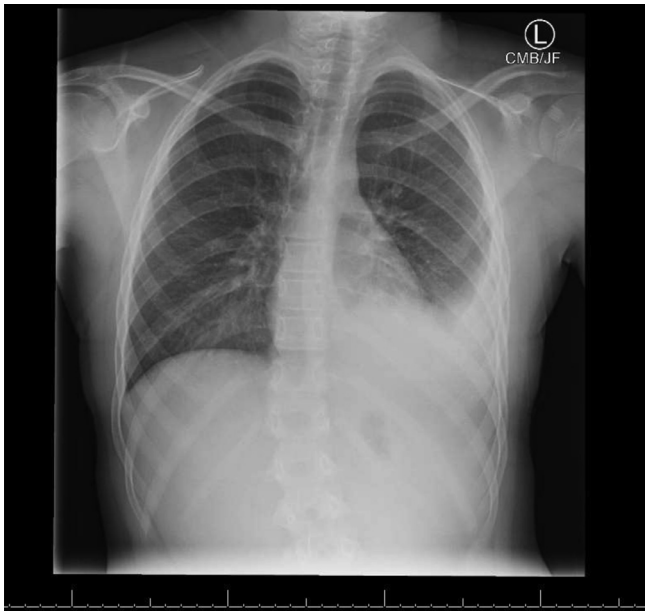


**Fig. 1 – Frontal chest radiograph demonstrating a moderate left pleural effusion.**

matic injury secondary to pleural injury caused by a rib spur.

## Case report

A 10-year-old girl with no past medical history and a family history of osteogenesis imperfecta presented as a transfer from outside urgent care after discovery of left-sided pleural effusion by chest radiograph to the pediatric emergency department. The patient reported left upper abdominal and lower chest pain intermittently over the last 2-3 days. The patient had no increased work of breathing but did report some dyspnea with more intense exertion. The patient denied any recent sick contacts, illnesses, fevers or chills, changes in intake, nausea, vomiting, diarrhea, night sweats, weight loss, and fatigue. There was no history of trauma, weight loss, or easy bleeding or bruising. There was no family history or personal history of pleural effusions. She had otherwise been in her usual state of health and was up to date on immunizations.

On arrival to the ED, patient's vital signs were SpO2 100%, BP 126/76, HR 103, T 36.8, and RR 24. Physical exam revealed a well appearing, well-nourished child in no acute respiratory distress. She had diminished breath sounds in the left lower lung field, and reported pain with deep inspiration. Work up in the ED included a complete blood count, comprehensive metabolic profile, and PT, PTT, INR, all of which were within normal limits.

Chest radiograph again demonstrated a small to moderate left pleural effusion (Fig. 1).

A bedside US was obtained which was positive for left sided pleural effusion (Fig. 2).

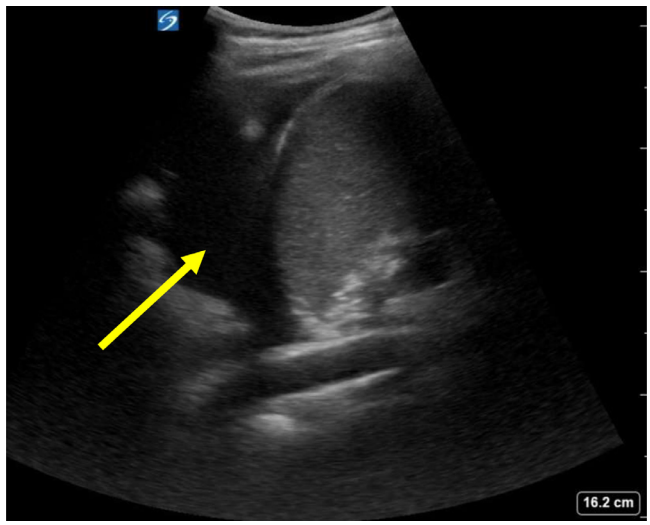


**Fig. 2 – Ultrasound demonstrating a pleural effusion. The effusion appeared anechoic and simple.**

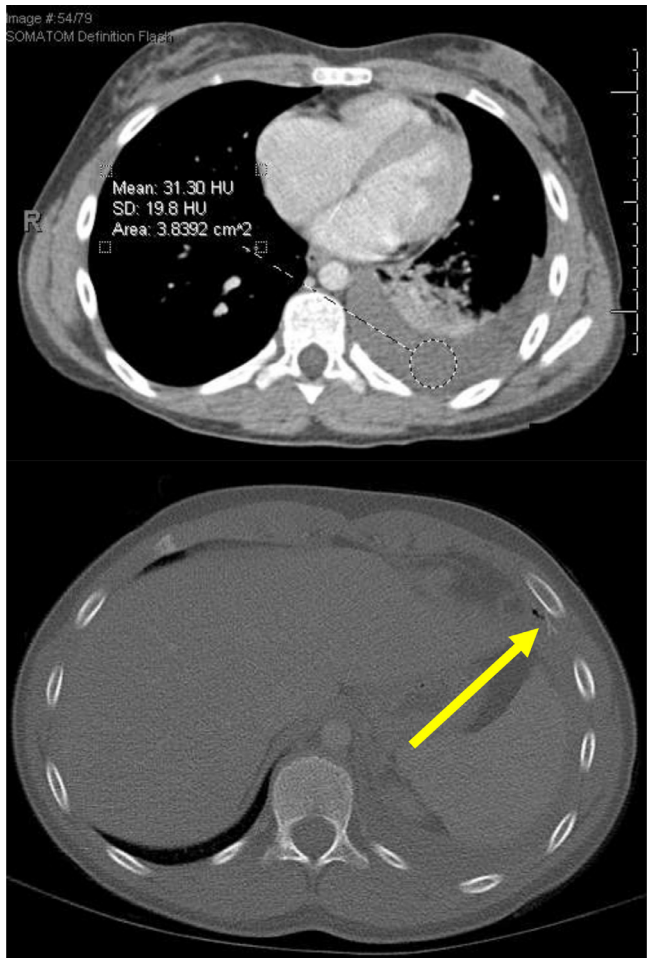


**Fig. 3 – CT Chest demonstrating a left hemothorax as well as a bone spur.**

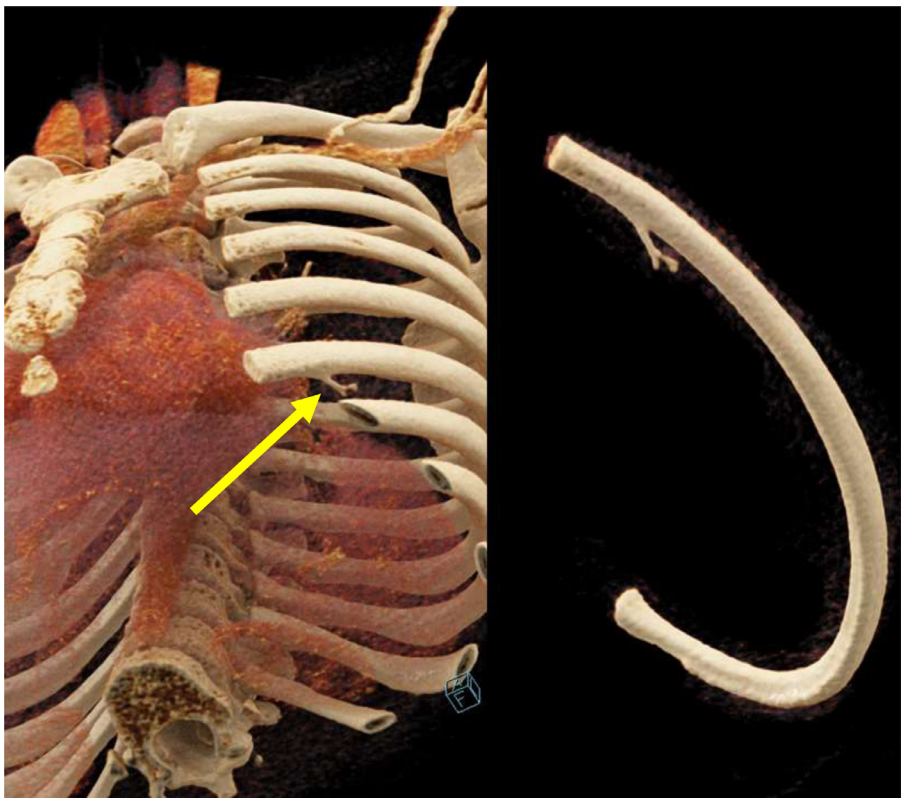

**Fig. 4 – 3D reconstructions demonstrating a bifid rib spur on the left 6th rib. The involved rib has been segmented out on the second image.**

A contrast-enhanced chest CT was obtained to further evaluate the etiology of the pleural effusion, specifically to assess for a malignant or infectious etiology given the lack of preceding trauma. The study demonstrated a moderate left pleural effusion that was higher density than simple fluid (approximately 31 HU), indicating a component of blood product. There was also a trace left pneumothorax. Best viewed on the sharp lung/bone reconstructions there was what appeared to be a bifid spur originating off of the left sixth rib, that measured approximately 1.0 centimeter in length, and which projected into the chest cavity in the region of the lung base and hemidiaphragm (Fig. 3). This anomalous structure was particularly well appreciated on 3D reconstructions processed after the study (Fig. 4).

The patient was admitted, and ultimately underwent video assisted thoracoscopic surgery (VATS). A total of 1000 ml of blood was evacuated from the thorax. A diaphragm laceration was also identified with pulsatile blood flow. This laceration was repaired, and hemostasis was achieved. The bone spur was identified on the sixth rib and was removed successfully (Fig. 5). Pathology report of the bone spur showed fragments of reactive bone with woven bone formation and hyaline cartilaginous areas with endochondral ossification, consistent with an osteochondromatous cartilaginous cap. Genetic testing for osteogenesis imperfecta and bone fragility panel were ordered. This identified 1 heterozygous pathogenic variant in the CTNS gene (partial deletion exons 1-10), consistent with carrier status only for autosomal recessive cystinosis. This finding was not thought to be the etiology of the bone spur. The patient's postoperative course was uneventful. The chest tube was removed on the second postoperative day, and she was discharged later that day.

In her first follow-up appointment 2 weeks post-discharge, she continued to do well and only reported having 2 episodes of recurrent chest pain that was located in the area her chest tube was placed. A chest radiograph was obtained subsequently, which did not reveal any abnormalities. The patient was also seen 1 month later, at which she had no more episodes of recurrent chest pains and was without complaints.

## Discussion

In the pediatric population, rib spurs are quite rare and difficult to diagnosis as the majority of them tend to be asymptomatic. Typically, these bony spurs manifest as osteophytes, osteochondromas, or exostoses.

Osteophytes are composed of fibrocartilage and bone, typically found in peripheral margins of joints that originate from periosteal or synovial tissue, and are particularly common in the setting of degenerative changes [4]. Although the exact mechanism behind osteophyte formation is unknown, it is thought that they are the result of a cellular response

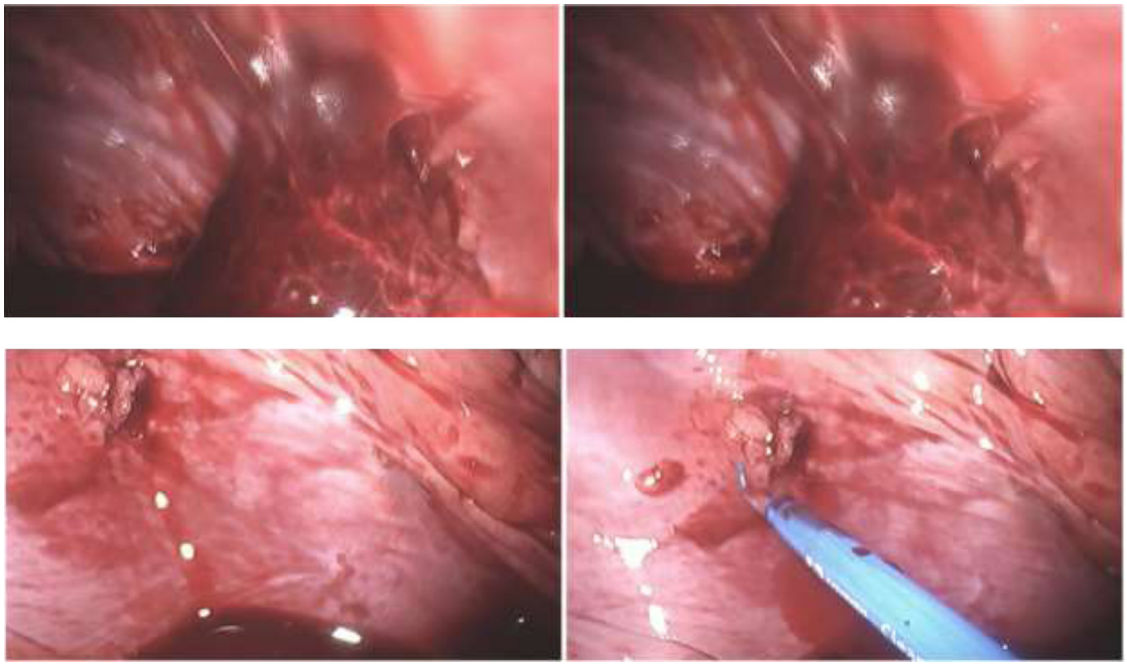

**Fig. 5 – Video-assisted thoracoscopic surgery showing clotted blood on the anterior and inferior aspect of the lung, which was removed revealing the rib spur.**

to the environment, in particular injury, growth factors, or cytokines [5].

On the other hand, osteochondromas are the most common bone tumor in children. They account for 20-50% of all benign osseous tumors [6]. Approximately 40% of osteochondromas are diagnosed by the time patients are 10 years old [7]. Multiple osteochondromas are present in the setting of multiple hereditary exostoses [8]. Osteochondromas can also develop as a result of prior injury/insult, with 1 example being prior radiation therapy.

Few cases of costal exostosis causing a spontaneous hemothorax have been reported in the literature [1,9–13]. A recent systematic review of hereditary familial exostosis with rib involvement identified 11 cases of spontaneous hemothorax in children [14], all of whom underwent initial X-ray followed by a diagnostic CT. The majority underwent VATS with exostosis removal; the one who did not undergo surgical intervention developed recurrent hemothorax 4 days later [15]. A separate case series noted two additional cases of hereditary multiple exostoses, which ultimately required surgical intervention, and cited complications of leaving in the bony protusions, such as spinal cord compression, brachial plexus palsy, hemothorax, diaphragm laceration, pneumothorax, and hemopericardium [16]. The pathogenesis has been postulated by Lin et al, that sharp edges of osteochondromas cause repetitive trauma to the diaphragm and pleura during the respiratory cycle leading to the development of hemothorax [13]. Of note, our patient had no family history of exostosis.

In the setting of a spontaneous pleural effusion/hemothorax in the setting of an otherwise healthy pediatric patient, a chest CT with 3D reconstruction can be useful to scrutinize the chest wall for potential causative osseous irregularities. This can help guide the clinical team to the correct cause of the effusion/hemothorax, as well as lead to definitive therapy.

## Patient consent

Signed consent was obtained from the patient's parents for publication.

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