Spontaneous Hemothorax: A Rare Presentation of Costal Exostosis

Spontan Hemotoraks: Nadir Bir Kostal Ekzostoz Sunumu

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ABSTRACT

Costal exostosis is a typically benign tumor growth of the rib that usually remains asymptomatic but sometimes can develop a severe complication such as hemothorax and required emergency surgery. At this moment, we presented a case of 20 years old gentleman with no previous history of trauma presented to our hospital with progressively worsening shortness of breath and computed tomography of chest show features of hemothorax. He subsequently underwent video-assisted thoracoscopic that found costal exostosis perforating the lung. The costal exostosis subsequently resected, and post-operatively he was discharged well. We, with this, presented a rare case of costal exostosis, along with the discussion on clinical presentation and management.

Key Words: Spontaneous pneumothorax, Costal Exostosis, Osteochondroma

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ÖZET

Kostal ekzostoz, genellikle asemptomatik kalan ancak bazen hemotoraks ve gerekli acil ameliyat gibi ciddi bir komplikasyon geliştirebilen kaburga kemiğinin tipik olarak iyi huylu bir tümör büyümesidir. Bu anda hastanemize giderek kötüleşen nefes darlığı ve hemotoraksın göğüs görüntüleme özelliklerinin bilgisayarlı tomografisi ile başvuran, daha önce travma öyküsü olmayan 20 yaşında bir beyefendi olgusunu sunduk. Daha sonra, akciğeri delen kostal ekzostoz bulunan video yardımlı torakoskopik ameliyata alındı. Kostal ekzostoz daha sonra rezeke edildi ve ameliyat sonrası iyi taburcu edildi. Bununla birlikte, klinik prezentasyon ve yönetim üzerine tartışmanın yanı sıra nadir bir kostal ekzostoz vakası sunduk.

Anahtar Sözcükler: Spontan pnömotoraks, Kostal Ekzostoz, Osteokondrom

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INTRODUCTION

Non-traumatic or spontaneous hemothorax is a rare occurrence, which usually associated with the underlying medical condition most commonly in a patient with anticoagulant therapy or with concomitant pneumothorax (1) and the other causes of spontaneous hemothorax including a bleeding disorder, pleural malignancy, and rupture of the thoracic aortic aneurysm. Costal exostosis also is one of the causes of spontaneous hemothorax but occurs scarcely ever. It may be due to direct trauma to the pleura or diaphragm. We presented a case of spontaneous hemothorax secondary to costal exostosis along with a discussion on the management of the disease.

CASE REPORT

A 20-year- old Egyptian gentleman, presented to us with progressively worsening shortness of breath for a one-week duration. It is associated with productive cough and reduces effort tolerance. Otherwise, he does not complain of fever, chest pain, loss of appetite, or loss of weight. He does not have any medical illness or history of trauma before the onset of symptoms.

A chest x-ray was performed and showed opacification of the lower half of left hemithorax with a meniscus consistent with the presence of pleural effusion in the left hemithorax and computed tomography of the chest revealed attenuation of the pleural fluid mixed with some hyperattenuating areas inferiorly which may be due to presence of blood. A diagnostic pleural tap was performed that show blood in the left thoracic cavity. Subsequently, a video-assisted thoracoscopic exploration was performed to identify the causes of bleeding and the treatment of spontaneous hemothorax.

A left seventh rib exostosis noted perforating the lung and adhesion between parietal and visceral pleura over the left lower lobe noted. The costal exostosis resected and adhesiolysis performed. The patients' postoperative stay was uneventful, and the chest tube removed without complication. The patient was followed up for six months, and no recurrent hemothorax noted.

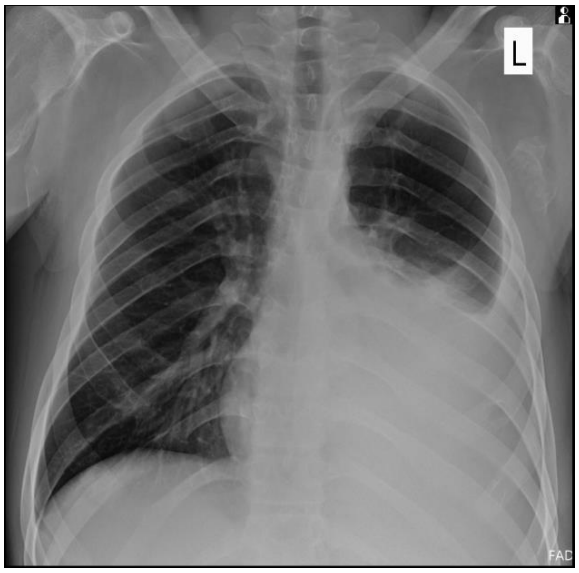

Figure 1: Chest radiograph showing left pleural effusion.

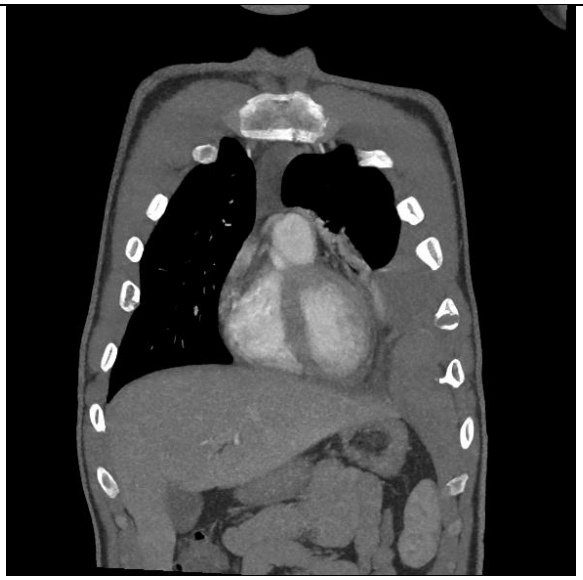

Figure 2: Computed tomography of chest showing left hemothorax with costal exostosis can be seen at 7th rib.

Figure 3: Computed tomography of chest showing left hemothorax in lung window.

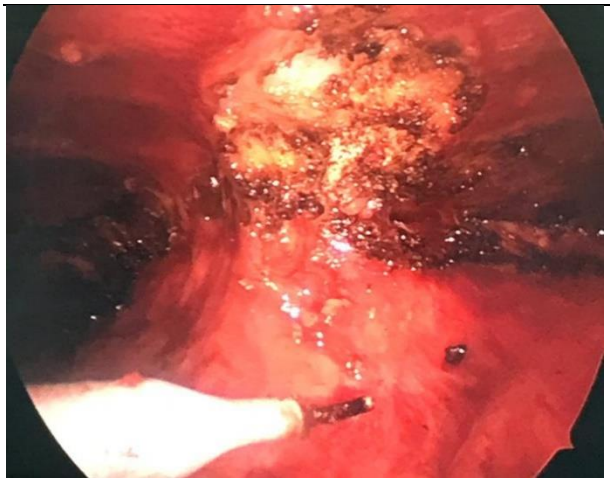

Figure 4: Laparoscopic view showing costal exostosis piercing the lung parenchyma.

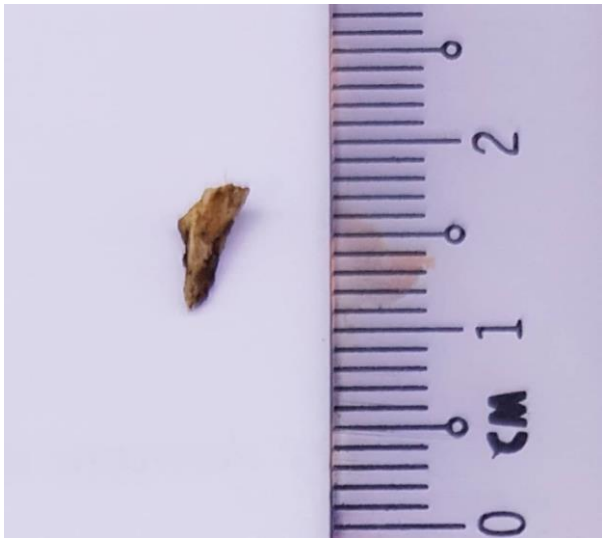

Figure 5: Bone extracted from surgery

DISCUSSION

Hemothorax defined as the presence of pleural fluid with a hematocrit higher than 50% of the patient's blood. It commonly presents secondary to penetrating or blunt chest trauma such as pleural biopsy, central lines insertion, or thoracocentesis (1). In the absence of any chest trauma, presence of blood in the pleural cavity, it is called spontaneous pneumothorax. Spontaneous pneumothorax is a rare clinical condition and maybe the first manifestation of an underlying coagulopathy, vascular malformation, connective tissue disease, thoracic neoplasm, and costal osteochondroma, similar to our case (1). A costal osteochondroma or exostosis is a benign outgrowth of bone cover with cartilage protruding from a rib. It may require emergent surgery due to its associated complications.

Osteochondroma is a second common benign neoplasm of the rib, and it represents 8% of rib tumors (2) and approximately 50% of benign rib tumors. They usually found in the anterior aspect at the costochondral junction (3). It usually occurs in children and young adults, with a mean age of 20-year-old (2). Costal osteochondroma can be presented as solitary exostosis or may be associated with hereditary disease, Hereditary multiple exostoses (HME) (4).

Hereditary multiple exostoses is an autosomal dominant benign clinical condition characterized by the formation of exostosis that usually occurs in the long bone. The patient will also have other skeletal deformities such as short stature, limb-length discrepancies due to abnormal bone growth (4).

For the patient with solitary costal exostosis, they usually do not have any positive family history and asymptomatic (5). However, they also can present with swelling, hiccup, chest pain, pneumothorax, and hemothorax (2-6). Symptoms may arrive as a result of mechanical irritation to nearby structures or organs. Several reports have suggested a theory of hemothorax; as a result of spontaneous rupture of a dilated blood vessel due to chronic inflammation that arises from contact friction during respiration between exostosis and pleura (5).

Diagnosis of costal exostosis usually comes with the first onset of its complication that arises. It is challenging to identified costal exostosis by plain chest radiograph alone as it may not be apparent. Computed tomography is more sensitive imaging to localize exact location costal exostosis by using the multiplane image (5). It also can help to differentiated osteochondroma with chondrosarcoma that may show cartilaginous calcification with nearby bone destruction.

Surgical removal of costal exostosis indicated when exostosis penetrating the intrathoracic structure such as hemothorax like our patient as it could be a life-threatening situation (7). Video-assisted thoracoscopic surgery (VATS) or thoracotomy is needed to resect the tumor and prevent recurrent hemothorax. VATS is a more preferred option as shown to reduce hospital stay, less blood transfusion, a shorter period of chest tube drainage, and less postoperative complications compared to open thoracotomy (8).

CONCLUSION

Costal exostosis is a rare clinical disease but should be in mind when a patient that presented with hemothorax without a history of trauma or surgery. Surgical removal of exostosis is indicated even in an asymptomatic patient that found during accidental finding as it is known to cause complication later in life. Thoracoscopy is an excellent procedure in managing this situation as it will provide good visualization for the evacuation of the hematoma and treating the exostosis while avoiding thoracotomy.

Conflict of interest

No conflict of interest was declared by the authors.

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