Sultan Qaboos University Med J, May 2022, Vol. 22, Iss. 2, pp. 300–303, Epub. 26 May 22
Submitted 22 Dec 20
Revision Req. 26 Jan 21; Revision Recd. 23 Feb 21
Accepted 17 Mar 21
https://doi.org/10.18295/squmj.4.2021.072

CASE REPORT

Mediastinal Hydatidosis with an Unusual Presentation

A rare case report

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ABSTRACT: Hydatidosis is a common zoonotic disease with a high prevalence in developing countries. While a solitary cyst with unilateral lung involvement is common, bilateral involvement and multiple cysts are rare, only seen in 20% and 30% of the cases, respectively. Likewise, extensive involvement of extrapulmonary tissues and mediastinum is rare. We report an unusual case of mediastinal hydatidosis mimicking an intrathoracic malignancy in a 24-year-old female patient. She presented in the year 2020 with a history of left-sided chest pain and heaviness in the left hemithorax for a period of two months. Diffuse, multiple fluid-filled cystic lesions with internal echoes throughout the mediastinum, lung, pericardium, diaphragm and chest wall were observed in contrast-enhanced computed tomography of the thorax. An incidental cystic lesion in the liver was also noted. Since serology for echinococcosis was negative, a differential diagnosis of intrathoracic malignancy was considered. However, intraoperative and histopathologic findings were suggestive of hydatidosis.

Keywords: Hydatid Cyst; Diseases Thoracic; Computed Tomography; Magnetic Resonance Imaging; Mediastinum; Case Report.

Hydatid disease caused by larval stage of *cestode echinococcus* is one of the major zoonotic diseases of public health significance with a reported mortality rate of 2–4%.[1] Although lungs are the second most commonly involved organ, intrathoracic extrapulmonary involvement is uncommon, seen in 5–7% of cases.[1,2] We hereby report a rare presentation of mediastinal hydatidosis with extensive lung involvement resembling malignancy.

Case Report

A haemodynamically stable, healthy 24-year-old female patient with no significant medical history presented to the Department of Cardiothoracic Surgery at Raipur, India in the year 2020 with heaviness and left-sided chest pain for two months. Respiratory symptoms—including fever, breathlessness, cough, haemoptysis, dysphagia, or dysphonia—were absent. In addition, the patient's weight or appetite were unaffected. A decreased left-sided chest movement, deviation of trachea to the right side, dullness over the left hemithorax and decreased vocal fremitus were observed during the chest examination. Furthermore, diminished breath sounds with decreased vocal resonance was noted on the left side. Routine blood investigations as well as liver and renal function tests were within standard limits. The pulmonary function test showed a restrictive pattern and sinus tachycardia was noted on the electrocardiogram; however, the findings on echocardiogram were normal. The chest X-ray posterior-anterior view showed opacity on the left hemithorax [Figure 1]. Contrast-enhanced computed tomography (CECT) of the thorax showed multiple fluid-filled cystic lesions throughout the mediastinum as well as within the lung parenchyma with atelectasis, with the largest measuring 10 × 7 cm [Figure 2]. An incidental hepatic cyst measuring 4 × 3 cm was also noted [Figure 2A]. A fibre optic bronchoscopy showed a deviated trachea with compressed left upper lobe bronchus and no endobronchial growth. Although the findings were suspicious of hydatid cyst, immunoglobulin G titres for hydatid serology was negative and needle aspiration was inconclusive. Therefore, differential diagnosis of various intrathoracic malignancy were considered.

The patient underwent left posterolateral thoracotomy under general anaesthesia with a double-lumen intubation. Intraoperatively, glistening white fluid filled cystic lesions resembling hydatid cysts were present throughout the lung parenchyma and mediastinum, with dense vascular adhesions between the cyst wall and thoracic cage. Parenchymal preserving cyst excision of the lung parenchyma with captionage and cystectomy for the cysts in the mediastinum, diaphragm, pericardium and the chest wall were performed [Figure 3A]. Histopathology was suggestive of multiple daughter cysts with a germinal layer and scolices suggestive of hydatid disease [Figure 3B]. The postoperative course was uneventful and the patient was prescribed albendazole 400 mg twice

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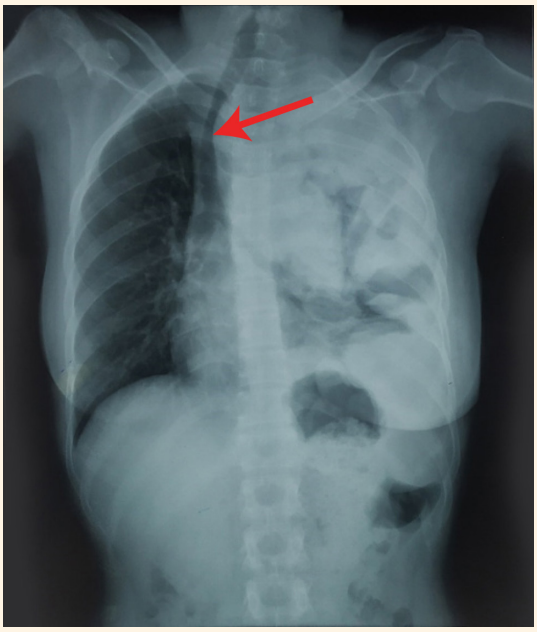

Figure 1: Chest X-ray posterior-anterior view showing left opacified hemithorax with contralateral tracheal deviation (arrow).

daily for a period of three months. Postoperative chest X-ray as well as a computed tomography scan of the chest after three months of follow-up did not show any recurrence [Figure 4]. The patient underwent laparotomy and cyst excision after six months for a hepatic cyst. Informed consent was obtained from the patient for publication of this report.

Discussion

Hydatidosis is a zoonotic disease transmitted via feco-oral route to humans from animal hosts including dogs and sheep. *Echinococcus granulosus* is the most common source of infection accounting for 95% of 2–3 million cases reported globally.[3] The infection prevalence varies widely by region, with higher prevalence in Mediterranean regions. Following the liver, the lungs are the second most common organ involved with a higher predilection for the right lower lobe.[4,5] However, in the current case, extensive involvement of left side was present. The cysts are often solitary and unilateral in distribution, while bilateral involvement and multiple cysts are not unusual, seen in 20% and 30% of cases, respectively.[5] Moreover, extensive mediastinal cystic involvement is rare, seen in less than 4% of cases.[6–8] The presenting symptom depends on the size, location of the cyst and degree of compression of the mediastinal structure. Vertebral destruction, superior vena cava syndrome and Bernard Horner's syndrome have been reported previously with mediastinal cysts.[9] Apart from the chest pain and heaviness of chest, no other associated symptoms were present in the current patient.

Considering the age, presence of diffuse involvement of lung parenchyma and opacified left hemithorax on CECT thorax and chest X-ray and negative serology for hydatid cyst, a wide variety of differential diagnoses were considered including mediastinal hydatidosis, germ cell tumour, pulmonary sarcoma and diffuse pulmonary metastases.[10] High attenuation wall and a low-density content are characteristic of hydatidosis. Germ cell tumours constitute 1–3% of intrathoracic malignancies affecting males with a mean age of 25–35 years.[11] Lobulated heterogeneous mass containing soft tissue elements with fluid and fat has been seen with immature teratoma with calcification in 20–40% of the cases.[10,11] On the other hand, pulmonary synovial sarcoma is

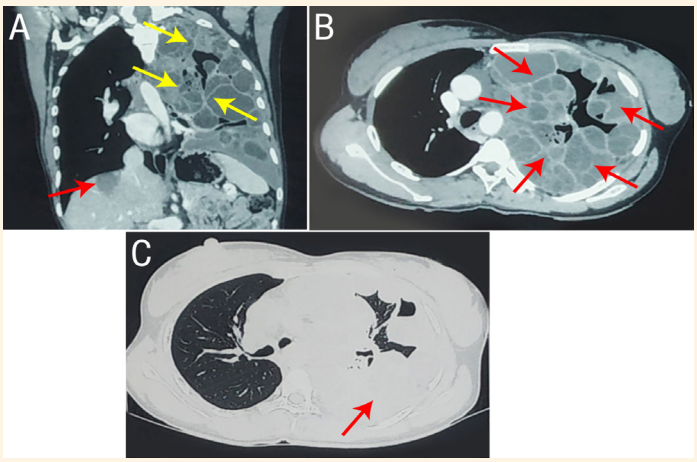

Figure 2: A: Contrast enhanced computed tomography (CECT) thorax coronal view showing multiple intrathoracic cystic lesions with thickened septa (yellow arrows) and hepatic cyst (red arrow). **B:** CECT thorax axial view showing interconnected cystic lesions (arrows) with mediastinal shift to the opposite side. **C:** Axial view of upper lobes with lung window showing consolidation of the left lung (arrow) with minimally visible parenchymal markings.

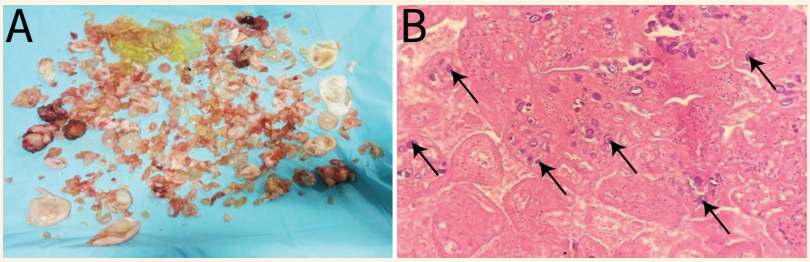

Figure 3: A: Innumerable hydatid cysts and daughter cysts with typical glistening white appearance. **B:** Hematoxylin and Eosin staining at ×50 magnification showing laminated and nucleated germinal layer giving rise to brood capsule. Protoscolices are seen within brood capsule (arrows).

characterised by well-circumscribed heterogeneous mass occurring between 16 to 77 years of life with equal sex predilection. Although well circumscribed, rounded soft-tissue attenuation noted with diffuse pulmonary metastases was reported in young patients and was considered as a differential diagnosis;[12] however, it is unlikely in the current case as the patient did not have any symptoms of metastases or malignant disease. Considering the above-mentioned differential diagnoses, posterolateral thoracotomy of the left side was planned. Intraoperatively, glistening white fluid filled cystic lesions were noted suggestive of hydatid disease, which was later confirmed by histopathology.

Although, magnetic resonance imaging (MRI) is superior in differentiating between solid and cystic lesions, its role in hydatidosis is very minimal. They appear as low signal intensity on T1-weighted and high signal intensity on T2-weighted images.[13] Similarly, cystic components of teratoma result in high signal intensity on T2-weighted images and low signal intensity on T1-weighted images with magnetic susceptibility artifacts in the presence of calcification.[14] Seminomatous germ cell tumours have a hypointense mass on T2-weighted images with relatively homogeneous enhancement.[14] In pulmonary synovial sarcoma, there is a heterogeneous signal intensity on T1-weighted images while in pulmonary metastases, diffusion-weighted images show high signal intensity.[12] Considering the feasibility, MRI was not suggestive in this case.

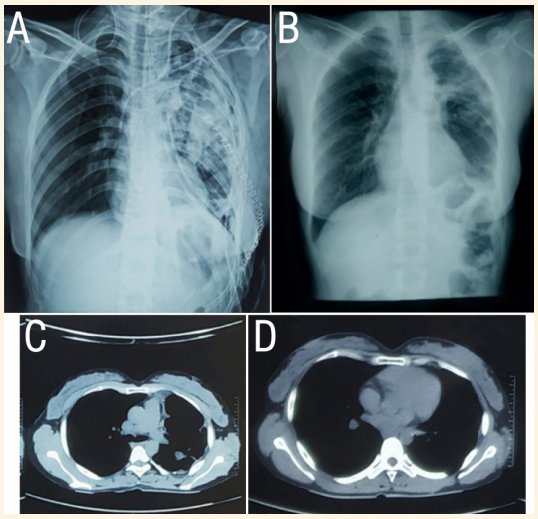

Figure 4: A: Immediate postoperative chest X-ray. **B:** Chest X-ray posterior-anterior view at follow-up after three months showing completely resolved cystic lesion. **C:** Computed tomography (CT) of the thorax at the three months follow-up at the level of carina showing no recurrent cystic lesions. **D:** CT of the thorax at the three months follow-up below the level of carina showing no recurrent cystic lesions.

Surgery is the most accepted treatment of choice for hydatid disease; however, in patients with recurrent cysts, multiorgan disease, poor general conditions and those who refuse to undergo surgery, medical management is the next best option.[13] Albendazole (10–15 mg/kg/day) is the drug of choice in hydatid disease. It inhibits microtubular assembly within the parasite resulting in glycogen depletion and finally autolysis of the cell.[3] Preoperatively, albendazole intake helps to soften the cyst and reduce the intracystic pressure while it prevents recurrence of the disease postoperatively. In the current case, albendazole was prescribed postoperatively for three months and no recurrence was observed during follow-up.

Conclusion

Due to its variable presentation, diagnosis based only on clinical and radiographic findings of mediastinal hydatidosis can be challenging. Although surgical excision remains the treatment of choice without extensive resection, a thorough understanding of all the radiographic differential diagnoses is vital while encountering such an extensive disease of the mediastinum to devise a treatment plan.

ACKNOWLEDGEMENT

I would like to thank Dr. Melisha R Pinto, MDS (email id: melisha.pinto@gmail.com) for proof reading and editing the manuscript.

AUTHORS' CONTRIBUTION

KD prepared and drafted the manuscript. NG edited the manuscript. Both KD and NG reviewed and approved the final version of the manuscript.

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