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Left Hemi-Diaphragmatic Hydatid Cyst - A Rare Presentation and Diagnostic Challenge

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Abstract

Echinococcosis or hydatid cyst disease is among the oldest diseases known to man. Common presentations include involvement of liver and lungs. Hydatid cyst in diaphragm without hepatic involvement is rare and less than 100 cases are reported in literature. We are describing a case of a young female who presented with cough anddyspnea secondary to large cysts that appeared to involve the right lung and spleen on initial imaging studies. Peroperatively, however, the presumed spleenic cystwas surprisingly found to be lying within left hemidiaphragm. Complete surgical resection of the cysts was performed with uneventful post-operative recovery.

Keywords

Hydatid cyst, Diaphragm, Echinococcus

Introduction

Hydatid Cyst (HCs) is a zoonosis caused by cestode Echinococcus granulosus and is endemic in many parts of the world. The most common organs that are affected by E. granulosus are liver and lungs (78%) [1]. Diaphragmatic localization is very rare with the incidence of around 1%, and most of these are generally associated with hepatic hydatid cysts [2]. According to Kjosseve all, less than 100 cases of diaphragmatic HCs have been reported in the international literature so far and most of them had concomitant involvement of right hemidiaphragm [1]. Herein, the authors report a case of a young female who presented with cough and dyspnea. Initial imaging studies showed large hydatid cysts involving the right lung and spleen. Peroperatively , however, the presumed spleenic cyst was surprisingly found to be lying within left hemidiaphragm.

Case History

A 16 years old girl presented with cough, dyspnea and pain in left hypochondrium for 15 days. She denied history of fever, weight loss and hemoptysis. Her past and family history was negative for congenital or hereditary diseases. General physical examination was unremarkable. Her respiratory system examination revealed dull percussion note and absent breath sounds in right mid and lower chest. A mass was palpable in the left hypochondrium which was interpreted as splenomegaly. Her ultrasound abdomen revealed large cystic hypo echoic lesions in right lower lung and spleen suggestive of HCs. CT scan was done which showed a well-defined 9.9 × 9 × 10 cm hypodense cyst involving the middle and lower lobes of right lung. It had thick enhancing walls and caused mild pleural effusion with adjacent atelectasis. A similar thick walled 11.2 × 9.9 × 10.5 cm hypodense cyst with enhancing walls and internal floating membranes was also seen, reported to be within spleen (Figure 1 and Figure 2). These cysts were causing displacement of surrounding vessels and adjacent viscera. No evidence of focal lesion was identified in liver parenchyma. Complete blood count, serum creatinine and liver function tests were within normal limits. Serology for echinococcus antibodies was negative. The patient was referred to cardiothoracic surgeon for further management. Thoracotomy was performed and right lower lobe cyst was removed. On the left side, however, presumed splenic cyst was surprisingly found to be lying within the diaphragm. It was successfully excised and the cavity was washed with hypertonic saline. Tissue diagnosis of HCs was confirmed in

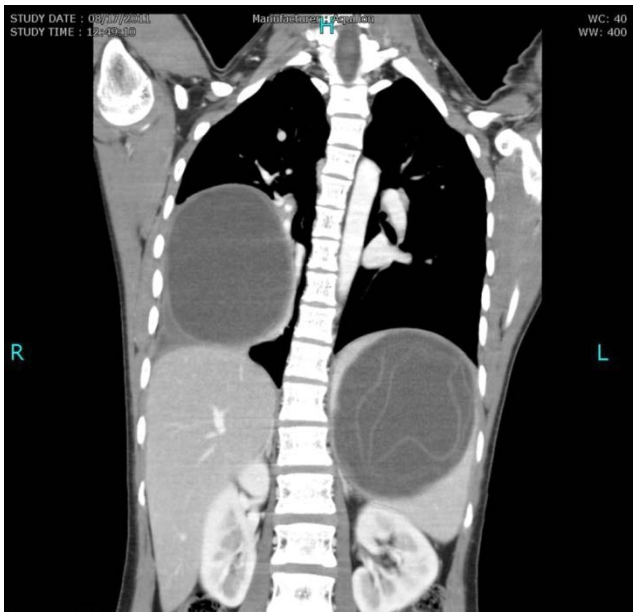

Figure 1: CT scan (coronal view) of bilateral hydatid cysts. One in right lung and the other 'reported' to be within spleen with floating membranes.

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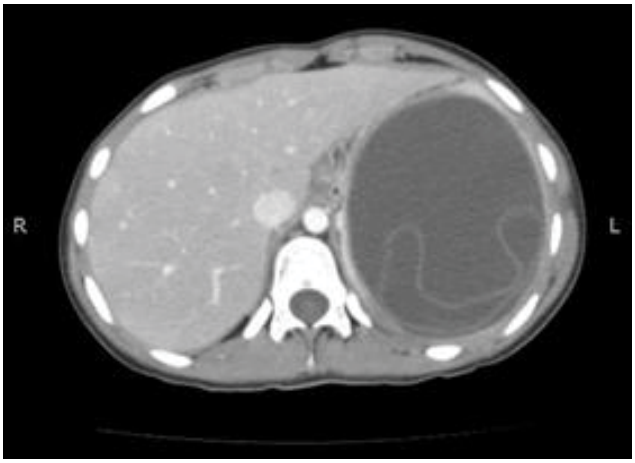

Figure 2: CT scan (sagittal view) of hydatid cyst 'reported' to be within spleen.

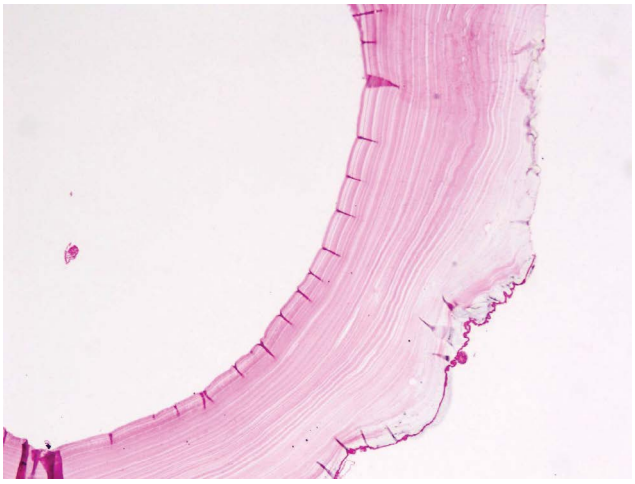

Figure 3: Histopathology section of HC showing thin germinal layer which gives rise to brood capsules.

both specimens (Figure 3). Postoperative recovery was uneventful and the patient was subsequently discharged on Albendazole (10 mg/kg body weight). On her last follow up visit, 2 months after surgery, patient was disease free and doing well.

Discussion

Hydatidosis is included in the differential diagnosis of space-occupying lesions. Although hydatid disease may affect any organ or soft tissue, the favoured sites of involvement include liver (70-75%) and lungs (20-25%) either alone or concomitantly (18%) followed by peritoneal cavity (10-16%). Less commonly, HCs may involve kidneys (1.5-4%), spleen (3%), brain (3%), musculoskeletal system (0.5-4%), heart (2 %), and retroperitoneum (1%). The HCs of the diaphragm is very rare [1]. In a review of 1,619 patients with thoracic hydatidosis, Jayashankar N *et al.* present the frequency of the intrapulmonary and extra pulmonary HCs as 94.6% and 5.62% cases respectively [3].

The symptoms of thoracic hydatidosis depend on the size and the site of the lesion. Thus, slowly growing HCs are generally asymptomatic until they are large enough to cause mass effect in an involved organ [3]. As in our case, the common presenting symptoms are cough, chest pain, and breathlessness [3].

More than one pathway has been proposed to explain the development of intrathoracic HCs. It is thought that small sized embryos (< 0.3 mm) initially gain access to the systemic venous system, as they can't be filtered out from the portal blood by virtue of their small size, and thereafter get pumped to all parts of body. Another proposed mechanism presumes that embryos gain access into the systemic circulation via intestinal lymphatics. Some researchers have supported a third pathway that involves direct exposure to lungs through inhalation of contaminated air [4]. After gaining access to the systemic circulation, further voyage to the diaphragm may involve pulmonary-bronchial anastomosis, arterial channels from the phrenic artery and branches from the intercostal arteries [1]. Left hemidiaphragmatic cyst in our case can be explained with the embryos bypassing the two filters i.e. liver and lungs.

Serological tests for hydatid disease have low sensitivity (64-87%) and imaging modalities are the mainstay for diagnosis [5]. Both sonography and CT scan have their pros in the form of cost and widespread availability but when it comes to cysts with atypical appearance on CT scan and peridiaphragmatic location, MRI is a better imaging modality due to its superiority in defining anatomical relationships and better chest wall and diaphragm delineation [1,6]. MRI may have helped in making correct localization of the cyst within the left hemidiaphragm. Eren *et al.* and Mekki *et al.* have also proposed MRI as a superior imaging modality in comparison with CT scan for evaluation of complex giant hydatid cysts, their topographical relationships and suspected diaphragmatic and hepatic involvement [2,7].

Conclusion

Hydatid cysts of diaphragm should be considered in patients with preoperative imaging data indicating cystic lesions adjacent to the diaphragm. MRI should be done if the cyst is found to be in close proximity of diaphragm, or elsewhere if in doubt, to determine accurate soft tissue relationship preoperatively.

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