



Unusual hydatid cyst in a 16 years old girl presented by shoulder pain

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ABSTRACT

Hydatid disease is caused by *Echinococcus granulosus* and is endemic in many parts of the world, including Iran. This parasitic tapeworm can produce cysts in almost every organ of the body, with the liver and lung being the most frequently targeted organs. However, the cyst tends to appear in different and sometimes unusual body sites in various geographical areas of the world. In this report we have presented a 16 years old girl that presented by abdominal pain and shoulder pain that was diagnosed as liver cyst and CT scan revealed a large cyst in liver and a large cyst in right lung that diagnosed as hydatid cyst.

Keywords: Hepatopulmonary, Liver, Hydatid Cyst

INTRODUCTION

The hydatid cyst is a zoonosis caused by adult or larval stages of tapeworms belonging to the genus *Echinococcus granulosus* [1]. The tapeworm stage is harbored in the intestine of carnivores such as dogs, which constitute the definitive host[2], and the eggs are passed in the feces of the infected carnivores and ingested by herbivores such as sheep, which comprise the intermediate host. Humans are the incidental intermediate host. Larvae emerge from the eggs in the intestine; and after invasion to the blood vessels, they can migrate into almost every part of the body[3]. The usual destination is the liver via the portal tract, but sometimes the larvae pass through the liver barrier and reach the lungs and all the other viscera, where they transform into small cysts[4]. Echinococcosis/hydatidosis is one of the most important zoonotic diseases inasmuch as it occurs in different parts of Iran[5]. Adult worms have been recovered from dogs, jackals, and wolves, but human cases have been reported from hospital archives by pathological reports of surgically proven cases in different geographical areas of the country.

Case report: A 16 years old single woman presented by ambiguous right upper quadrant pain without tenderness and intermittent right shoulder pain for 2 months recently abdominal pain has increased and onset of dyspnea, patient had no fever, nausea, vomiting, jaundice. On physical examination patient had puberty rashes on face, mild tenderness on right upper quadrant, no sign of previous surgery, In abdominal ultra-sonography reported that: 14x12cm cyst with irregular small area of hyperechoic material occupied most part of liver lobe, no evidence of calcification or daughter cyst, no intrahepatic or extra hepatic duct dilation and no gallstone or cholecystitis. Kidneys and bladder was normal and no evidence of free fluid, or adenopathy. And suggested for CT scan because of crowded ribs in the region Then performed a chest, abdominal, and pelvic CT scan with double contrast reported that: a large 14x11cm thin wall non-enhanced cyst is noticed in right liver lobe without gross internal septa, daughter cyst, or calcification, extended to the right diaphragm, same side also contain a large identical cyst measured 9x8 cm

with same characteristic as liver cyst, findings was highly suggestive for amebic cyst with trans diaphragmatic migration.

The cyst had two different arising regions in lateral and apical segment in right upper lobe of lung. Also reactive mild right-sided free fluid is seen. The rest of chest and abdomino-pelvic CT appeared normal. In ELISA test, echinococcus IgM was: 11.054(<20 is negative) and IgG was: 10.025 (<9.0: negative, 9-11: borderline, >11: positive).

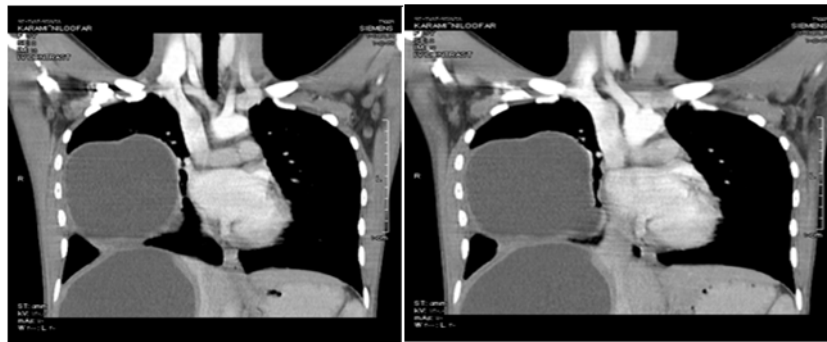


Fig. 1. Coronal section of thorax and abdomen CT scan

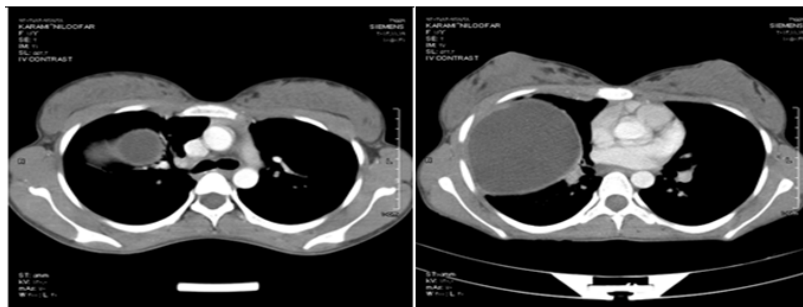


Fig. 2. Axial section of thorax and abdomen CT scan

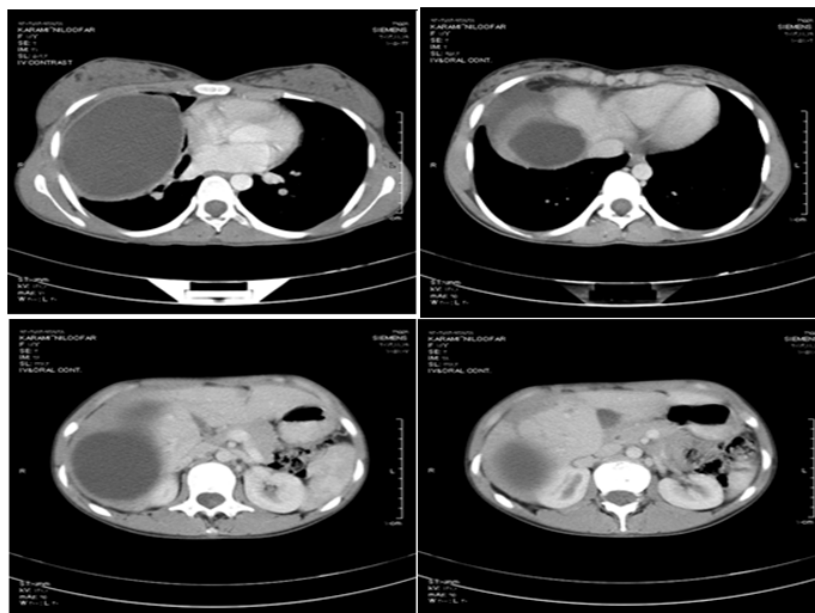


Fig. 3. Different sections of thorax and abdomen CT scan

The patient underwent drain the liver cyst under guide of ultra-sonography and injection of hypertonic saline that during the procedure developed by anaphylactic shock, so administered infusion of dopamine and a drain was inserted and transferred to ICU, after stability prepared for operation and patient under general anesthesia after a long thoracoabdominal incision and splitting of diaphragm entered abdominal cavity and pleural cavity previous catheter was extracted then slide off all around pulmonary and peritoneal cyst then hepatic cyst and germinal layer was resected and sent for pathology then pneumolysis was done and cyst was resected with segmentectomy of involved lung with linear staph after irrigation and hemostasis chest tubes was placed and mesh was placed adjacent to liver and diaphragm was repaired. In pathology report hydatid cyst confirmed. The patient had a normal uneventful postoperative recovery and was discharged 4 days after the operation. The patient provided informed consent for this case report.

DISCUSSION

Hydatid disease is a unique parasitic disease that is endemic in many parts of the world [6]. In hydatid disease, the liver and lung are the most common involved organs, but the disease can be seen in any organ of the body [7, 8]. The rates of the localization of hydatid disease in different body organs vary in the literature [9]. The liver in adults and the lungs in children are the predominant sites [10]. It develops outside these organs in 2.1% of the patients [4]. Extrapulmonary intrathoracic localization is seen in 8% [11]. Cysts in two-thirds of patients are unilateral and/or solitary [2,3,5,12,13]. However, recently it has been noted that there is an increase in the patients with multiorgan localization and multiple cysts [5]. Single pulmonary localization of the hydatid disease is more common in children and youth than adults [3]. Although single pulmonary cyst in adults is seen in both sexes, it is more frequent in males (53–70%) in the second and third decades [2,10,15]. Although pulmonary cysts are asymptomatic in 8–32% of the patients with hydatid disease, the symptoms are generally related to the cyst size or complicated [1–3]. The pulmonary cysts may grow fast due to the elastic structure of the lungs and they may reach huge sizes especially in children.

In that occasion, they may cause the symptoms such as cough, chest pain, dyspnea, expectoration and hemoptysis [2,6,12,16] in our case shoulder pain was an extraordinary symptom. However, the cysts may grow slowly in the liver because of the organ solidity [1,13,16]. Although pulmonary cysts may establish in every lobe of lungs, they are more frequent in lower lobes and mainly in the right hemithorax [1–3,12,15,16]. The hepatic cysts also prefer the right lobe (73%) of liver [1,7]. The most important diagnostic tool in hydatid cysts is chest X-ray [1,11,15,17]. It is typical for an intact cyst to present as round or oval homogenous densities with sharp contours. Computed tomography of the chest clearly reveals especially cysts, which are complicated, behind or in the heart or mediastinum [11,15,19]. Ultrasonography and echocardiography are two indisputable methods to evaluate the hepatic and/or pericardiac–cardiac cysts [11].

A rupture of the cyst into the pleural cavity is a severe complication but rarely seen and is reported 0.5–18.2% in the literature [13,14,20,21]. This complication may be associated with both pulmonary and hepatic cysts [1,2,4,13,14,20,21]. This rarity is due to the dense adhesions between the cyst and the parietal pleura, not letting the cysts to open into the pleural cavity [20]. Several procedures have been described for the treatment of hepatic echinococcal cysts, ranging from simple puncture to liver resection and transplantation, although the most commonly used technique is total or partial cystopericystectomy. Usually, radical surgery (total pericystectomy or partial hepatectomy) is indicated for liver cysts. Conservative surgery (open endocystectomy with or without omentoplasty) or palliative surgery (simple tube drainage of infected cysts or communicating cysts) is also an option. More radical interventions have higher intraoperative risks but less numerous relapses. With the inclusion of chemotherapy prior to or after surgery, less-aggressive surgery may be possible. Surgery for pulmonary cysts includes extrusion of cysts using Barrett technique (intact endocystectomy without preliminary aspiration), pericystectomy, and lobectomy [10]. The puncture of echinococcal cysts has long been discouraged because of risks of anaphylactic shock and spillage of the fluid; however, as experience with ultrasonography-guided interventional techniques has increased since the early 1980s, an increasing number of articles have reported its effectiveness and safety in treating abdominal, especially liver, echinococcal cysts.

A recent systematic review of the literature found that the overall fatality rate due to lethal anaphylaxis from puncture of echinococcal cysts is 0.03% (2 in 5943 procedures) for procedures and 0.04% (2 in 5517 cysts) for cysts respectively [11].

COCLUSION

In this case, the girl's clinical presentation was atypical and before aspiration of the cyst fluid all immunologic markers for hydatidosis was negative, but The diagnostic suspicion of hydatid disease was raised after abdominal ultra-sonography and the history of frequent contact with dogs and sheeps. After aspiration under sonography guide anaphylacticraction happened and at the ICU course all the markers for hydatidosis came positive. Another wondering finding in this case was type and position of the cyst. Despite of other hepatopulmonaryhydatidosis Cysts this cyst had two arising area; one in right lobe of liver and the other arising was hanging on apex of right parietal pleura.

Hepatopulmonaryhydatidosis (HPH) can cause greater economic and social burden compared with hydatidosis with single-organ involvement because the management of the former may require numerous surgical interventions and prolonged postoperative care. Therefore, appropriate diagnosis and an optimal surgical approach in this group of patients are issues of paramount importance [12, 13]. A combined resection of hydatid cysts at both the sites during the same operation when feasible with a maximum preservation of the parenchyma in uncomplicated cysts (regardless of their dimension) appears to be a safe and optimal approach [13, 14].

In endemic regions, physicians require a high index of suspicion for hydatid disease, especially when circular cystic lesions are occasionally visualized on routine chest radiography. Additionally, because there are many potential sites of cyst formation and the presence of multifocal disease affects the therapeutic strategy, it is necessary that patients with suspected pulmonary hydatid disease be investigated for the possibility of hepatic involvement [13]. Physicians should be familiar with the diagnosis of patients with clinical manifestations of hydatid disease, such as HPH. HPH represents a separate clinical entity that requires a different surgical approach may result in beneficial outcomes in terms of reducing its financial and occupational loss.

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