Mediastinal cystic lesions: a rare entity

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ABSTRACT Background: Hydatid cysts are caused by infection with a tapeworm parasite called Echinococcus granulosus. They are usually located in the liver and lungs. This is a case report of a mediastinal hydatid cyst. Although many uncommon locations of this disease are reported in the medical literature, only a few papers mention this type of lesion in the mediastinum. **Case summary:** We report the case of a mediastinal cyst in a 58-year-old Belgian female patient presenting chest pain, dry cough and dyspnea during the last four weeks. She was travelling to Tunisia two months before her admission. The diagnosis is suspected by Ultrasound coupled with CT-scan and confirmed by the surgical findings and the histopathological study. The therapeutic attitude was aggressive (cyst excision surgery), to avoid any complication or degeneration among the surrounding vital structures. **Conclusion:** Mediastinal hydatid cysts are extremely rare and must be considered in the differential diagnosis of the cystic masses of the mediastinum even in a non-endemic area, due to tourism and current important migration movements across Europe. Chest scan is the preferred imaging tool in diagnosis. Surgical removal of the cyst is the most common treatment.

KEYWORDS hydatid cyst, mediastinum, echinococcus granulosus

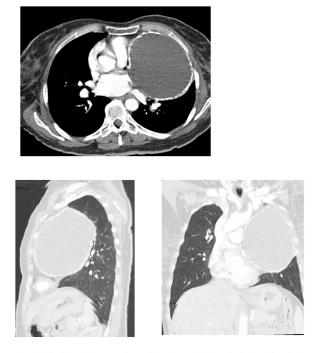
Introduction

Cystic echinococcosis (hydatidosis) is a parasitic disease caused by larval development of Echinococcus granulosus. It affects most frequently the liver and the lungs. Mediastinal involvement is rare. The frequency of the primary hydatid cyst in the mediastinum varies from 0.1% (in Western countries) to 4% (in endemic countries such as Tunisia) of all cases of hydatidosis [1, 2]. In endemic countries, the incidence of mediastinal hydatidosis varies between 0.5% and 2.6% of all chest locations [3]. The clinical problem of this localization is to differentiate it from other causes of mediastinal cysts. Bronchogenic, pleuropericardial, thymic, intramural esophageal, lymphangioma, anterior meningocele and enteric origins, as well as other rare types of cysts, may be found in the mediastinum of adults and children [4]. We report the case of a mediastinal cyst discovered in the setting of symptoms of mediastinal compression.

Copyright © 2019 by the Bulgarian Association of Young Surgeons DOI:10.5455/JJMRCR.Mediastinal-cystic-lesions First Received: October 26, 2019 Accepted: November 15, 2019 Editor-in-chief: Ivan Inkov (BG) ¹ Services des Urgences CHC Saint Joseph, rue Hesbaye 75, 4000 Li ge, Belgique; Email: deeba.ali@chc.be Figure 1.Transthoracic echography in sub-xyphoid: highlights the mediastinal mass which has a liquid appearance with a calcified wall which causes compression on the left side, at the level of the pulmonary vessels, the OG and VG seems to be repressed by this mass.



Figures 2: Enhanced CT scanner demonstrate a large cystic lesion develop from the anterior part of the left hilum with repression on the left lung. This lesion have <u>calcificated</u> walls, the HU of the content is compatible with serous liquid. No enhancement, partition or fleshy part are find inside the lesion.



Figures 3,4: Enhanced CT scanner demonstrate a large cystic lesion develop from the anterior part of the left hilum with repression on the left lung. This lesion have <u>calcificated</u> walls, the HU of the content is compatible with serous liquid. No enhancement, partition or fleshy part are find inside the lesion

Case report

A 58-year-old Belgian woman was evaluated in the Cardiology outpatient clinic for chest pain, dry cough and dyspnea evolving and progressively increasing during the last four weeks. Her past medical history was remarkable for mental retardation, swallowing and psychotic disorders. She visited Tunisia two months ago.

Initial clinical examination revealed sinus tachycardia (96 beats/min) and blood pressure was 90/70 mmHg. Respiratory rate was 25/min, oxygen saturation was 90% on room air, and she was afebrile. The cardio-respiratory examination showed normal heart sounds and marked hypoventilation in the left hemithorax.

Electrocardiogram showed diffuse alteration of repolarization. Complete blood count, electrolytes as well as liver, renal and coagulation studies came back within the normal range.

Transthoracic echocardiogram showed a cystic formation on the left side of the mediastinum. It should be noted that the cyst presented partially calcified wall. This formation of 11×13 centimetres displaced the left ventricle which then compressed the right ventricle itself (Fig. 1).

The diagnostic workup was completed with a chest CT-scan. The CT-scan showed a 12x13 cm non-contrast-enhanced liquid formation with calcified walls and a mass effect on the mediastinum and the pulmonary vessels (Fig. 2,3 and 4). Considering the location and the morphology of the cyst, the main diagnostic hypothesis was a pleuropericardial cyst or a cystic hamartoma. Surgical intervention was discussed and carried out. A brown liquid was obtained through the cyst aspiration, and it was subsequently removed with no postoperative complications. The histopathologic study was consistent with a hydatid cyst.

No specific anti-parasitic drug therapy (such as albendazole or mebendazole) was given to the patient. The follow-up with the cardiologist and the cardiothoracic surgeon with a chest CTscan at three months did not show any complication, and the symptoms were relieved.

Discussion

We report the case of a mediastinal cyst in a psychotic patient with swallowing disorders who was travelling to Tunisia 2 months prior to her admission, turning out to be a hydatid cyst.

Hydatid cysts are caused by infection with a parasite called Echinococcus granulosus. The adult worm resides in the jejunum of canines and produces eggs that are passed in the stools. Intermediate hosts (cow, sheep, human) are infected through ingestion of parasite eggs in contaminated food, water or soil. Eggs then liberate an embryo in the duodenum, which passes through the intestinal mucosa to enter portal circulation. Most of these embryos are trapped in the liver [4]. The rarity of finding the parasite in other locations than the liver and lungs can be explained by the fact that these organs act as a blood filter against the dissemination of the parasite. The pathogenesis of mediastinal localization of hydatid cyst remains controversial. Some findings argue in favour of the hypothesis that the parasites cross the hepatic and pulmonary filter subsequently reaching the systemic circulation. Mediastinal involvement could also result from the lymphatic circulation [1, 3].

Hydatid disease is primarily an illness affecting residents of pastoral and rangeland areas who frequently come into contact with carnivores, sheep, and cows. It has a worldwide distribution with highly endemic areas in the Mediterranean region, northern Africa, the Middle and the Far East, South America, Central Asia, Siberia and western China [4].

Mediastinal cystic lesions are rare and represent 12 to 30% of the mediastinal masses, most of them of congenital origin. Their multiple etiologies are mainly dominated by bronchogenic cysts (accounting for 50 to 60% of all mediastinal cysts), springing from division abnormality from the embryonic foregut. Mesothelial cysts (about 30% of all mediastinal cysts) are arising from abnormal coelomic development. Pleuropericardial cysts, thymic cysts and cystic lymphangiomas have also been described. They are frequently found unexpectedly in routine exams as patients are often asymptomatic [5].

These mediastinal cystic lesions are very rare and usually evolve as a mass of progressive onset. Symptoms and signs are unspecific and may include chest pain, dyspnea, venous compression, cough, nerve compression and dysphagia.

Diagnosis workup must include imaging investigations. Chest X-ray often shows widening or deviation of the mediastinum, a round-shaped opacity of a homogeneous watery tone, and sometimes the presence of arciform calcifications. Transthoracic ultrasound is a very sensitive exam because it identifies the liquid nature, characterize the wall of the cyst and its location [2]. Ultrasounds sometimes determine the diagnosis alone, but CT-Scan is necessary to precise the localization of the cyst and establishes its relationship with the surrounding structures. CT-scan usually shows a non-contrast-enhanced mass of liquid density, with well-defined margins. The MRI has no advantage over CT-scan for pulmonary hydatid cyst and is less cost-effective [6,7]. These exams (ultrasounds, CT-scan, MRI) allow in most cases to differentiate hydatic mediastinal cyst from other mediastinal cystic masses.

Treatment is indicated in all symptomatic cysts, big size asymptomatic ones or case of diagnostic doubt. Surgery is the treatment of choice. Theoretically, radical excision of the pericyst would be the ideal curative operation. However, the extensive resection may be difficult due to dense adhesions to vital structures around. Therefore, partial pericystectomy and removal of the germinative membrane are recommended [8]. Surgery relieves from all the symptoms, helps to obtain a precise diagnosis with histopathologic study and prevents complications.

Other options for the treatment of cystic echinococcosis consist in percutaneous treatment of the hydatid cyst with the PAIR technique (Percutaneous puncture of cysts using US guidance, Aspiration of cyst fluid, Injection of protoscolices for 10–15 min, Re-aspiration of the fluid) and anti-infective drug treatment. Anthelmintics drugs can be used to prevent recurrence following surgery or PAIR. Albendazole is currently the drug of choice to treat cystic echinococcosis, either alone or together with percutaneous treatment. Given orally, it should be administered continuously at a dosage of 10–15 mg/kg/day. However, optimal dosage and optimal duration have never been formally assessed [9].

Conclusion

Mediastinal hydatid cysts are extremely rare but should be considered in the differential diagnosis of mediastinal cystic masses even in the non-endemic area because of population movements. Chest scanner is the preferred imaging tool in diagnosis. Surgical removal of the cyst is the most common treatment. Postoperative anthelmintics drugs therapy can be administered.

Learning points

- Hydatid cysts should be considered in the differential diagnosis of mediastinal cystic lesions.
- Chest scanner is the preferred imaging study.
- The clinical approach of mediastinal cysts lesions should consider the epidemiological data,

Conflict of Interest

There are no conflicts of interest to declare by any of the authors of this study.

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