

Two patients presenting with mediastinal mass



Patient 1

A 32-year-old female from a suburb of Antalya (Turkey) was admitted with a mediastinal mass. She was a housewife and had no history of pet or other animal contact. She had no symptoms and her physical examination was normal, with no other remote pathological findings. She was subsequently referred to the Department of Cardiothoracic Surgery due to hilar and mediastinal enlargement incidentally found on her chest radiograph (figure 1) and a well-shaped mediastinal mass (diameter of 4x5x6 cm) in the left side of the middle mediastinum identified from her chest computed tomography (CT) (figure 2). Routine blood and liver ultrasound tests were normal. The preliminary diagnosis was mediastinal unknown mass. A left thoracotomy was performed and revealed a cystic mass with capsula, with approximate dimensions 4x5x6 cm, located in middle mediastinal area.



Figure 1
Pre-operative chest radiograph of patient 1, showing mediastinal enlargement.



Figure 2
Pre-operative chest CT of patient 1, showing a cystic and well-shaped mediastinal mass.

Patient 2

A 46-year-old male living in Antalya (Turkey) was admitted with mediastinal mass, cough and fatigue. He also had no medical history related to the presence of animals or pets in his home. His physical examination was normal and there were no other remote pathological findings. Routine blood and liver ultrasound tests were normal. However, chest radiography showed hilar and mediastinal enlargement. Additionally, chest CT revealed a well-shaped mediastinal mass (6x8x10 cm) in the anterior mediastinum. Median sternotomy was performed, and revealed that the capsulated tumour was cystic, measured 6x8x10 cm and was located in the anterior mediastinal area (figure 3).



Figure 3
The capsulated and cystic mediastinal mass from patient 2, which was surgical removed by left thoracotomy.

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Task

Study the similar case presentations for both patients and think of an underlying cause. Turn the page to find the diagnosis.

Answer

In both patients, the capsulated tumours were removed entirely and histopathological investigation diagnosed **primary mediastinal echinococcosis**.

Clinical course

Neither patient suffered complications, and they were treated with 15 mg·kg⁻¹·day⁻¹ of albendazole for 6 months post-operatively. Both patients underwent examination for cysts elsewhere in the body. CT scans of the abdomen and brain were performed on the 5th day after surgery and no primary origin of hydatid cyst was revealed. Echocardiography showed that the heart and great vessels were normal in both patients. Consequently, the patients were discharged on the 7th and 9th days post-operatively. One year after surgery, they were both in excellent health and asymptomatic.

Discussion

Hydatid disease, caused by *Echinococcus granulosus*, is an uncommon parasitic disease. However, it is frequently encountered in sheep-rearing regions of the world, including countries around the Mediterranean, in the Middle East, Central Europe, South America and Australia, and it causes major economic problems in endemic areas [1]. Turkey is an endemic area where hydatid disease can be studied in detail [2, 3]. To date, only small patient series and sporadic cases have been reported in the literature from the UK [4, 5].

In this article, two recently treated cases of primarily mediastinal hydatid cysts are presented. Although the features of this disease have been described previously in small series and case reports, no defining features of this disease have been published. In the present report, the clinical diagnostic and therapeutic method choices are discussed in more detail.

The adult *E. granulosus* is a worm that resides in the jejunum of the host (dogs and other canines) and produces eggs that are passed on through faeces. Accidental infection of human beings occurs after ingestion of the ova excreted by the host. The ingested ova hatch in the duodenum and then penetrate the intestinal mucosa. Subsequently, they enter the portal circulation, through which they are transported to the liver. Finally, they disperse to other organs and turn into hydatid cysts [6].

Hydatid cysts may occupy any place in the

body. However, >90% localise in both the liver and lungs. The remainder (<10%) are localised in other organs, such as the spleen, kidneys, pancreas, brain and heart [7]. Hydatid cysts are rarely found in the mediastinum [7].

Cystic lesions account for up to one quarter of all mediastinal masses identified incidentally or during work-up for symptomatic mediastinal abnormalities [6]. Cysts of bronchogenic, pleuropericardial, thymic, intramural oesophageal, lymphangioma, anterior meningocele and enteric origin, as well as other rare types, may be found in the mediastinum of adults and children. Thameur *et al.* [8] previously identified mediastinal hydatid cysts in eight out of 1,619 intrathoracic hydatid cysts (0.5%), and echinococcosis of the mediastinum was diagnosed in 0.5% of cases. Additionally, Rakower and Milwidsky [9] recorded >23,000 patients with hydatid disease in various large series. Among these, only 25 cases (0.1%) were in the mediastinal compartment and paravertebral sulcus. In one of the current patients the cyst was located in the anterior mediastinum, whereas, in the other one, it was located in the visceral region.

The region of Antalya in Turkey is endemic for echinococcosis [6, 7]; however, the current authors have only treated two sporadic cases within last 19 years, which does not reflect the echinococcosis population in the region. Neither patient had pet or other animal risks of hydatid cyst in their medical histories. In general, mediastinal echinococcosis is neither clinically nor radiologically distinguishable from other mediastinal cystic lesions [10]. Diagnosis can be obtained through the combined assessment of clinical, radiological, historical and laboratory data of patients, as in the cases presented here. Currently, chest radiography, chest CT and magnetic resonance imaging facilitate diagnosis. Chest CT is considered essential, and it is important for displaying the morphology, density and limits of these lesions. Moreover, it often accurately defines the relationship of the lesion with the adjacent structures. However, the definitive diagnosis can only be made by surgery with histopathological investigation. Some serological tests (haemagglutination) and some skin tests (the Casoni and Weinberg test) can also be used for diagnosis, although these tests have high rates of false-positive or false-negative results [6].

Symptoms and complications of the cysts depend on their size, location and involvement of neighbouring structures. If cyst-related symptoms are present, they are usually the result of

compression or erosion of the adjacent structures. And these symptoms range from retrosternal or parasternal pain, cough and dysphagia, to dyspnoea or severe compression of the trachea and superior vena cava [6, 10]. Here, both cases had small-sized mediastinal hydatid cysts, and, because of this, they had no severe symptoms.

The best therapeutic approach is to radically remove both the germinative membrane and pericyst through the appropriate thoracic incision [6, 9, 10]. When localisation of the cyst and invasion of vital structures prevent total excision of the cyst, partial pericystectomy is performed by removing the germinative membrane. Total excision was performed in both patients presented here, as the cysts were small in size. It has been proposed that better results are obtained by

combining surgical procedures with albendazole or mebendazole for post-operative prophylaxis, which reduces the incidence of recurrence [11, 12]. Post-operative albendazole was administered to the patients and recurrence was not encountered in the post-operative 6-month period.

In conclusion, although very rare, hydatid disease should be considered in the differential diagnosis of cystic lesion, especially in endemic regions. Chest CT is the most efficient method of diagnosing these lesions. Surgical removal was successful in both cases presented here and remains the best choice of treatment for mediastinal echinococcosis. To avoid recurrence, additional adjuvant medical therapy is recommended.

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