CASE REPORT

Mediastinal hydatid cyst rupturing into the pleural cavity associated with pneumothorax: Case report and review of the literature

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Hydatid disease remains a serious health problem in Mediterranean countries. Living in a rural area is an important risk factor for the disease. Hydatid cysts are usually located in the liver, lungs and brain. Mediastinal hydatid disease is very rare and has been noted only anecdotally in the literature. The present article reports a case of a mediastinal hydatid cyst rupturing into the pleural cavity, which was associated with pneumothorax of the same side. The patient’s previous chest x-rays (posteroanterior and left lateral views) showed a well-defined mediastinal mass on the left side, and contrast-enhanced computed tomography of the thorax (taken a few days after the chest x-ray) showed multiple round-to-oval soft tissue opacities with partial collapse of the left lung. An indirect hemagglutination test for echinococcosis was positive. Even after two weeks of intercostal tube drainage, the patient’s condition did not improve. During thoracotomy, multiple daughter cysts were found in the pleural cavity, and the diagnosis of a hydatid cyst was confirmed after histopathological examination.

Key Words: Hydatid; Mediastinum; Pneumothorax

Hydatid disease is primarily an illness of residents in rural areas who frequently come into contact with carnivores, sheep and cows. Human hydatid disease, caused by the larval form of Echinococcus granulosus, has a worldwide distribution, and is endemic in many countries in the Mediterranean region, the Middle and Far East, and South America (1). Hydatid cysts have a predilection to locate in the liver, lungs and brain. Although many uncommon locations have been reported, the disease is rarely present in the mediastinum. Approximately 100 cases have been described in the English literature so far (2-5). Although the features of these mediastinal diseases have been described in small series and case reports, no large series (2-5). Although the features of these mediastinal diseases have been described in small series and case reports, no large series

CASE PRESENTATION

A 45-year-old woman presented to the emergency room with complaints of acute onset chest pain on the left side of the chest and breathlessness of sudden onset. She had been complaining of dull, aching chest pain, cough with expectoration and low-grade fever for the previous three months.

Un kyste hydatique médiastinal se rompant dans la cavité pleurale et associé à un pneumothorax : Rapport de cas et analyse bibliographique

La maladie hydatique demeure un problème de santé préoccupant dans les pays méditerranéens. Vivre dans une région rurale est un facteur de risque important de la maladie. Les kystes hydatiques se trouvent généralement dans le foie, les poumons et le cerveau. La maladie hydatique médiastinale est très rare et n’a été déclarée que de manière empirique dans les publications. Le présent article fait état d’un kyste hydatique médiastinal qui s’est rompu dans la cavité pleurale, en association avec un pneumothorax du même côté. Les radiographies thoraciques précédentes du patient (vue postéroanterior et latérale gauche) avaient révélé une masse médiastinale bien définie du côté gauche, et une tomodensitométrie du thorax avec injection d’un agent de contraste (prise quelques jours après la radiographie thoracique) ont permis de distinguer de nombreuses opacités ovalaires ou rondes des tissus mous et un collapsus partiel du poumon gauche. Une épreuve d’inhibition de l’hémagglutination indirecte était positive à l’échinococcose. Même après un drainage par sonde intercostale de deux semaines, l’état du patient ne s’était pas amélioré. Pendant la thoracotomie, de multiples kystes fils ont été découverts dans la cavité pleurale, et le diagnostic de kyste hydatique a été confirmé après un examen histopathologique.

On general examination, the patient was hemodynamically unstable, with a pulse rate of 100 beats/min and a systolic blood pressure of 70 mmHg. The patient was febrile and cyanotic. The use of accessory muscles of respiration was observed. On percussion, a hyper-resonant note was heard in the left side of the chest, with a normal percussion note in the right side. On auscultation, breath sounds were absent in the left side of the chest, whereas on the right side, normal vesicular breath sounds were heard without added sounds. Results from routine laboratory testing are shown in Table 1.

Serum electrolyte levels and liver function test results were normal. Ultrasonography of the abdomen was normal. A chest x-ray (posteroanterior view) showed pneumothorax with multiple rounded opacities (Figure 1); her previous chest x-ray showed a sharply circumscribed, homogenous mass visible in the left mid-zone, having a smooth but lobulated contour (Figure 2). Moreover, a chest x-ray (left lateral view) showed an opacity in the mediastinum, with compression atelectasis of the surrounding lung (Figure 3). Contrast-enhanced computed tomography of the thorax showed multiple, rounded, well-defined cystic masses, with partial collapse of the lung (Figure 4). An indirect hemagglutination test for echinococcus was positive.
Initial resuscitation of the patient from shock was followed by closed pleural drainage. Thoracotomy was performed after two weeks. At the time of thoracotomy, hydatid membranes with multiple daughter hydatid cysts of varying sizes were found lying in the pleural cavity. There was an opening in the left lower lobe with a bronchial communication. The membranes and daughter cysts were removed, and mediastinal pleura, along with the pericystic wall, was excised. Decortication was performed and the fistula was closed.

Histopathological components were the stroma, areas of liquified and nonliquified necrosis, metacestodal vesicles, entrapped host tissue, microcalcification and protoscolex in a brood capsule.

Postoperative recovery was uncomplicated and the patient was discharged on albendazole therapy.

**DISCUSSION**

Adult *E. granulosus* is a worm that resides in the jejunum of dogs and other canines, and produces eggs that are passed in the stool. The eggs, ingested by intermediate hosts (cows, sheep and humans), liberate an embryo in the duodenum, which passes through the intestinal mucosa to enter portal circulation. Most of these embryos get trapped in the liver; the rest pass through the liver and scatter to other organs and develop into hydatid cysts. Consequently, hydatid cysts may be found in every tissue and organ.

Echinococcal cysts are composed of tissue from both the host and parasite. A nonruptured cyst is surrounded by the pericyst – a layer derived from compressed host tissue and chronic inflammatory cells (6). The true cyst wall is derived from the parasite and is arranged in two layers. The acellular outer laminated ectocyst is 1 mm to 2 mm thick and is lined by a one-cell thick germinal membrane, the endocyst. The endocyst produces hydatid fluid and scolecis, which are infectious, miniature,
adult tapeworms. Intracystic fluid pressures of up to 58.8 mmHg (7) keep the parasitic membranes tightly applied to the pericyst. The endocyst is fragile (8,9) and is normally protected from rupture by the tougher pericyst.

Hydatid cysts are rarely present in the mediastinum. From collective statistics of all studies, the occurrence of mediastinal hydatid disease varies from 0% to 6% (10). Rakower and Milwidsky (10) examined more than 23,000 patients with hydatid disease; only 25 (0.1%) hydatid cysts were reported in the mediastinal compartment and paravertebral sulcus. Thameur et al (4) identified mediastinal hydatid cysts in eight of 1619 (0.5%) intrathoracic hydatid cysts.

Eroglu et al (11) reported 11 (2.6%) cases of primary mediastinal hydatid cysts in their 427 patients with thoracic hydatid disease in Turkey (endemic region for hydatid disease).

Symptoms and complications of mediastinal cysts depend on the size, location and involvement of neighbouring structures (12,13), and range from being asymptomatic to having retrosternal pain, cough, dysphagia, dyspnea, or severe compression of the trachea and superior vena cava (2,5). Marti-Bonmati et al (12) reported a case in which the cyst ruptured into the aorta. Eroglu et al (11) reported a case of a mediastinal hydatid cyst rupturing into the pleural cavity. Hydatid cysts in the lung can cause complications. The cyst may erode into the bronchus, with air entering the various layers of the parasite to produce any one of several x-ray appearances (14), or the cyst may rupture, with the patient coughing out the hydatid fluid and membranes. The incidence of pneumothorax in patients with hydatid lung disease is reported to vary from 0.5% to 3.2% (15,16). When a subpleural hydatid cyst increases in size, it incorporates the visceral pleura to form a pleuro-pericyst layer. An inflammatory reaction between the two pleural surfaces may precipitate pneumothorax in five of their cases of hydatid lung disease.

The gold standard for therapy is radical removal of germinal membranes and pericyst through appropriate thoracic incision (2,5,10). When the localization of the cyst and invasion to vital structures prevents total excision, partial pericystectomy is the treatment of choice after the removal of the germinal membrane. Postoperative prophylaxis with albendazole reduces the incidence of recurrence (23,24). The possibility of secondary hydatidosis due to seedlings in the pleural space is always present. The reported incidence varies from 10% to 61% (16); hence, there is a need for long-term follow-up.

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