CASE REPORT

A rare cause of pleural effusion: ruptured primary pleural hydatid cyst

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SUMMARY

Hydatidosis is an endemic parasitic disease in Mediterranean countries, often caused by the dog tapeworm *Echinococcus granulosus*. The disease predominantly affects the liver (60–70%) and lungs (30%), and the surgical management is considered as the gold standard for treatment. Besides anaphylactic reactions, the most frequent complication of the hydatid disease is rupture into neighbouring structures, often affecting the bronchi, gastrointestinal tract and peritoneal/pleural cavities, according to its location. Primary pleural hydatidosis is an extremely rare entity and we present a ruptured pleural hydatid cyst with unusual location.

BACKGROUND

Hydatid disease (HD) is endemic in many parts of world, especially in Mediterranean countries. It can be located in any part of the body, but in 85–95% of the cases, the liver and/or the lungs are involved.¹ Pleural HD is generally secondary to lung involvement and may be due to a hydatid cyst that arises in the liver and prolapses into the chest,² but primary pleural HD is seen in less than 1% of all cases. Imaging modalities and serological tests are useful diagnostic tools; surgery with post-operative anthelmintic therapy is the treatment of choice, and a close follow-up is required.

CASE PRESENTATION

A 73-year-old man presented to emergency department with shortness of breath for 3 h which developed abruptly. There was an incisive pain in the left side of the thorax with decreased respiratory sounds on the left hemithorax, on physical examination. All blood investigations were normal except for mild eosinophilia. In deepening his history, it is learned that he was a farmer and had habitual contact with dogs.

INVESTIGATIONS

A chest X-ray revealed pleural effusion on the left hemithorax.

A CT scan of the chest demonstrated pleural effusion of 4.5 cm thickness located in the left hemithorax within the thickened layers of parietal and visceral pleura. There were pleural irregularity and air bubbles within the effusion. Adjacent to the effusion, a moderate compression atelectasis in the lung parenchyma has also been seen (figures 1 and 2).

Blood investigations showed a mild eosinophilia, and ELISA for hydatid was positive therewithal in pleural fluid sampling.

Figure 1 CT reveals pleural effusion on the left hemithorax. There is pleural irregularity and air bubbles in the effusion and no calcification.

There was no other lesion in cranial and abdominal CT and ultrasonography.

DIFFERENTIAL DIAGNOSIS

As a left-sided pleural effusion and pleural irregularity was present, exudative tuberculosis, loculated empyema and other parasitic infections such as amoebiasis were considered in differential diagnosis. However, imaging and serological tests confirmed a hydatid cyst.

TREATMENT

The patient was treated with steroid and albendazole preoperatively which was followed by surgery. Lesion was aspirated, a scolical agent was injected, reaspirated and the patient was treated again with albendazole for 2 months. No lobectomy was needed as the lesion was only located in the pleura.

Figure 2 Adjacent to the effusion, a moderate compression atelectasis in the lung parenchyma is also seen.

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OUTCOME AND FOLLOW-UP

After surgery, the patient made an uneventful recovery. Serial ultrasound, abdominal, thoracic and cranial CT and clinical examinations were performed to follow-up the patient for 1 year, and he remained asymptomatic during this period.

DISCUSSION

HD is caused by larval cysts of the *Echinococcus* tapeworm, usually by the *Echinococcus granulosus* species. Infection results from ingestion of food or drink contaminated by tapeworm eggs or through close contact with dogs or other infected carnivores.^{3–4} It is an endemic parasitic disease in Mediterranean countries.⁵ *E granulosus* most commonly occurs in the liver as a space-occupying lesion; the second most common are lung cysts, whereas rupture into the pleural space occurs very rarely.⁶ Lung cysts are usually caused by transphrenic spread of larvae through the hepatic sinusoids³ or by haematogenous spread of metacystotodes following rupture of eggs in the stomach. After digestion of outer capsule of the egg, the freed embryo enters the portal vein through duodenal mucosa and locates and grows in the hepatic capillaries. Some of them pass through the capillaries and become lodged in the lungs and other organs,^{7,8} which may also result from translymphatic spread. Primary pleural HD is extremely rare² and multiple pleural involvements can be seen in cases of lung HC ruptures. Its initial phase is mostly asymptomatic and latent periods of several decades can be seen.^{9–10} This explains the late presentation of symptoms in our patient. A CT scan of the hydatid cysts is diagnostic with well circumscribed, rounded lesions with low attenuation and no contrast enhancement.¹¹ Calcification of the cyst wall is best seen on unenhanced CT. On plain radiography, the calcification can also be seen called as 'egg-shell calcification'. In addition, serological tests are reliable screening tools, including immunoelectrophoresis and ELISA. Immunoelectrophoresis is a more sensitive test for anti-hydatid antibodies, but ELISA is more specific.¹² In literature, treatment strategy is accepted as surgery with pharmacotherapy with benzimidazoles (mebendazole or albendazole).¹³ Percutaneous treatment with puncture, aspiration, injection and reaspiration is effective in liver cysts,¹⁴ but the technique shows more complications in pulmonary cysts as accidental intraoperative spillage of cyst contents may result in anaphylaxis and/or dissemination with later cyst recurrence, and therefore not suggested for pulmonary cysts. New surgical options include enucleation, wedge resection, segmentectomy, lobectomy and pulmonectomy, according to the location of lesion in the pulmonary parenchyma,¹⁵ with minimal visceral and parietal destruction.¹⁶ It is suggested that after diagnosis of the lesion, cysts should be surgically treated as soon as possible, preserving the maximum lung parenchyma regardless of their sizes.¹⁷ After surgery, a long follow-up is needed for recurrence and early treatment; our patient showed no signs of recurrence radiologically.

Learning points

- ▶ Primary pleural hydatidosis is a rare occurrence of hydatid disease.
- ▶ In endemic regions, cystic lesions with non-specific clinical signs and symptoms should alert for hydatid disease even if it is rare.
- ▶ For any recurrence, a long-term follow-up is required.

Competing interests None.

Patient consent Obtained.

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