Seronegative, complicated hydatid cyst of the lung: A case report

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ABSTRACT

Cystic echinococcosis (CE) is an important helminthic zoonotic disease that commonly affects the liver and lungs. Imaging methods and serology establish the diagnosis in most cases. Chest x-ray can diagnose uncomplicated pulmonary hydatid cysts, whereas superinfection and/or rupture of the hydatid cyst (complicated cysts) may change the radiographic appearance and lead to delayed diagnosis and treatment. We report the case of a patient with hemoptysis and chest pain, where computer tomography scan of the lung suggested a large, ruptured hydatid cyst. However, serological tests with indirect hemagglutination (IHA) for Echinococcus granulosus antibodies were negative, and there was massive growth of Streptococcus pneumoniae in sputum. Based on this, we concluded that the patient had a bacterial lung abscess. The diagnosis of CE was only made after surgical removal of the cyst followed by microscopy and polymerase chain reaction.

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suggested a hydatid cyst. The air filling indicated that the cyst communicated with the airways. The rest of the lung fields and mediastinum appeared without pathology. (Fig. 2). A CT scan of the abdomen revealed no abnormality. The patient was prescribed antihelmintic treatment with albendazole 400 mg, two times daily.

Blood samples tested with enzyme-linked immunosorbent assay (ELISA) for *E. multilocularis* and with indirect hemagglutination (IHA) for *E. granulosus* antibodies came out negative. Microscopy of sputum showed massive growth of *Streptococcus pneumoniae*, and it was concluded that the patient had a lung abscess. The patient was prescribed broad-spectrum antibiotics and albendazole was discontinued.

Few days later the patient developed acute chest pain; chest X-ray (Fig. 3) showed atelectasis of the lower part of the left lung with an air compartment above the abscess. The patient was prescribed broad-spectrum antibiotics and albendazole was discontinued. 600 mL yellow liquid was removed and sent for microscopy. Due to respiratory distress the patient was transferred to our intensive care unit (ICU). After 24 hours of care in the ICU, the now stable patient was transferred to the pulmonary ward.

A multidisciplinary team conference was held to discuss differential diagnoses, further investigations and management. The team concluded that further investigation with bronchoscopy was necessary. Microscopy and polymerase chain reaction (PCR) tests for *E. granulosus* bronchio-alveolar lavage (BAL) fluid were negative. However, tests for *Aspergillus* galactomannan antigen were positive. On suspicion of invasive aspergillosis the patient was prescribed antifungal treatment with IV voriconazol 200 mg twice daily.

After presentation of the case to the department of thoracic surgery, lateral thoracotomy at IC5 was performed and the cyst was removed by lobectomy of the superior lobe of the left lung. Microscopic examination and polymerase chain reaction (PCR) for DNA showed the presence of *E. granulosus*. Thus, the diagnosis was changed accordingly, and the patient was prescribed albendazole 400 mg, twice daily for 6 months.
The patient came to regular follow-up in the outpatient clinic for infectious diseases for 12 months. The patient was asymptomatic during this period and no recurrence was observed.

3. Discussion

CE is one of the major zoonotic diseases in the world, and it is found on every continent except for Antarctica. High prevalence of E. granulosus is reported in the Middle East [4]. In our case, the patient came from rural Syria, where CE is endemic.

The clinical presentation of CE differs between simple and complicated cysts. A complicated hydatid cyst of the lung is defined as a cyst that has ruptured into the pleural or bronchus cavity, with or without secondary infection of the cyst [5]. Those cysts regularly cause cough, chest pain and shortness of breath. Small hydatid cysts are usually asymptomatic [6]. Our patient presented with hemothysis in addition to the typical symptoms. Parasitic aetiology of hemothysis is rare, as it is more common in tuberculosis, bronchiectasis, bronchogenic carcinoma or trauma. In our case tuberculosis was ruled out by a negative IGRA and negative sputum cultures. Hemothysis in patients with CE may be caused by pressure erosion of a bronchus, an obstructive effect with bronchial infection or rupture of cysts into the bronchus, which can lead to massive hemothysis [6].

Most cases of CE are diagnosed by a combination of history of exposure, serological testing of serum or hydatid fluid and imaging [3]. Chest X-ray is usually the initial investigation for a patient with lung symptoms. An intact single hydatid cyst appears as a homogeneous spherical opacity with definite edges. It is often surrounded by normal lung tissue [7]. Images of a complicated hydatid cyst vary and may resemble a lung abscess, malignant tumor, tuberculosis, and other cystic lesions of the lung [6]. Large cysts can cause atelectasis of adjacent parenchyma, induce a pleural reaction or shift the mediastinum [8] as seen in our case.

For complicated hydatid cysts, CT scan has proven to be an important imaging modality. Various signs are associated with hydatid cyst, e.g. both the serpert sign and the spin sign indicate a collapsed parasitic membrane [9]. Other classical radiographic signs are: signet ring, water lily, crescent or meniscus, inverse crescent, air bubble and dry cyst [10]. Superinfection with bacteria of ruptured hydatid cysts can change the appearance of the cyst on X-ray and CT scan [11].

Serological methods such as ELISA, IHA, latex agglutination test and immunoblot test (IB) are commonly used to detect hydatid cyst [12]. However, commercially available serological tests have shown unsatisfactory performance in terms of both sensitivity and specificity. Factors such as cyst size, stage, location and patient characteristics have been suggested to affect the test results [13]. A negative serological test normally does not rule out CE [14]. IHA test for serum antibodies for E. granulosus is positive in approximately 50% of patients with pulmonary CE [15]. However, complicated cyst usually causes positive serology. In a series of 100 cases, IHA showed a sensitivity of 100% in patients with ruptured cysts [16]. In our case, the CT scan indicated a large, ruptured hydatid cyst. Therefore, negative E. granulosus serology strongly suggested the absence of CE. However, in patients with initial negative serological testing a combination of several tests may increase diagnostic accuracy [17]. This was not performed in our case because massive growth of S. pneumoniae in sputum instead led us to conclude that the patient had a bacterial lung abscess. Later, positive Aspergillus galactomannan antigen in the BAL fluid indicated that the patient had invasive aspergillosis. Investigation after the removal of the hydatid cyst revealed a secondary S. pneumoniae infection of the cyst and false positive results for aspergillosis.

Peripheral blood eosinophilia is present in <25% of persons with hydatid cysts and are more common in cases of ruptured cysts [12]. There was no peripheral blood eosinophilia in our case.

Protoscolices may be detected in the sputum or bronchial washings in patients with ruptured pulmonary hydatid cysts [12]. Sputum was not evaluated for protoscolices, membranes or parasite hooks in our case. Considering our suspicion of a complicated hydatid cyst and that the patient came from a country where E. granulosus is endemic it would have been appropriate to investigate sputum with microscopy and PCR. Though we evaluated BAL fluid for E. granulosus, an evaluation of sputum may have increased the chance of detecting the infection at an earlier stage.

Treatment of lung hydatid cysts is essentially surgical. Surgery is associated with low mortality (1–2%), acceptable morbidity rate, and low recurrence rates (1–3%) [18]. The surgical options include lobectomy, wedge resection, pericystectomy, intact endocystectomy, and capitonnage [19]. There are only two antimicrobials effective against CE: albendazole and mebendazole. Albendazole is the drug of choice because its systemic absorption and penetration into hydatid cysts is higher than mebendazole. When surgery is necessary, presurgical use of albendazole reduces risk of recurrence and facilitates the removal of the cyst by reducing intracystic pressure [18]. Albendazole is effective at a dosage of 15 mg per kg body weight in 2 divided doses, and the usual dose is about 800 mg daily. Therapy is often indicated for a minimum of 3–6 months [20].

Declaration of interest

The authors declare no conflict of interest. The study received no financial support.

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