Ruptured pulmonary hydatid cyst: a case report

Maryam Karimi^{1,2} · Ali Rostami^{1,3} · Adel Spotin⁴ · Soheila Rouhani¹

Received: 24 April 2016 / Accepted: 10 January 2017 / Published online: 19 January 2017
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Abstract Ruptured pulmonary hydatid cyst (PHC) is an important clinical problem in endemic areas to echinococcal infection. Herein we present a rare case of ruptured PHC in an adolescent boy that was misdiagnosed as pulmonary tuberculosis in local health center. When sputum specimen was stained by acid-fast staining for detection of *Mycobacterium tuberculosis*, hooklets of *Echinococcus granulosus* were observed. A simple chest X-ray showed a multilobulated mass in the lower part of the left lung. Computed tomography scan verified existence of thick walled caviar lesion with irregular air–fluid level. The diagnosis was confirmed at the time of surgery. Misdiagnoses of PHC may even lead to irreparable damages. Therefore, accurate diagnosis is necessary to prevent severe complications.

Keywords *Echinococcus granulosus* · Pulmonary hydatid cyst · *Mycobacterium tuberculosis* · Lung

Introduction

Hydatidosis, due to infection with the metacestode of *Echinococcus granulosus*, is a silent zoonotic disease with worldwide distribution. The most common sites of cysts are liver and lung but it may also be located in spleen, brain, eye, heart, bone marrow, kidney (Rostami et al. 2015). It is believed that primary infection with *E. granulosus* occurs during childhood and lungs are the most common site of hydatidosis in children (Topçu et al. 2000; Lightowlers and Gottstein 1995). Due to slow growth of cysts, hydatidosis is often asymptomatic until the cyst grows and expands for several years after infection. In many cases, it is recognized incidentally during imaging studies (Smith and Rickman 2001). Symptomatic hydatidosis of the lung often follows the rupture of the cyst (Koul et al. 2000; Santivanez and Garcia 2010). Cyst rupture might be seen spontaneously or as a result of trauma. Pulmonary hydatidosis causes several symptoms such as unexpected onset of cough, fever, hemoptysis, and anaphylactic shock (Köksal et al. 2004). Pulmonary hydatid cyst (PHC) may rupture into the pleural cavity or bronchus (Balci et al. 2002). In such cases, PHC may be misdiagnosed with other common pulmonary diseases like tuberculosis and thus patients may not be receiving appropriate treatment. For example, misdiagnoses of PHC may even lead to major lung resection or pneumonectomy (Puri et al. 2011; Santivanez and Garcia 2010). Herein we present a rare case of ruptured PHC in an adolescent boy that was misdiagnosed as pulmonary tuberculosis in local health center.

Case presentation

A 16-year-old boy from a village presented with a 3-month history of fever, severe cough and a 2-month history of vomiting. There was no history of hemoptysis. Two months

✉ Soheila Rouhani
srouhani11@yahoo.com; srouhani11@sbmu.ac.ir

¹ Department of Parasitology and Mycology, School of Medicine, Shahid Beheshti University of Medical Sciences, Tehran, Iran

² Cellular and Molecular Research Center, Faculty of Medicine, Kurdistan University of Medical Sciences, Sanandaj, Iran

³ Infectious Diseases and Tropical Medicine Research Center, Babol University of Medical Sciences, Babol, Iran

⁴ Department of Parasitology and Mycology, Faculty of Medicine, Tabriz University of Medical Sciences, Tabriz, Iran

Fig. 1 Hooklet of *Echinococcus granulosus* in sputum smear, **a** $\times 10$ magnification, **b** $\times 40$ magnification

after initial symptoms and signs, severe dry coughing and itchy rose spot on the skin of throat and chest of patient were observed. Gradually he suffered from shortness of breath and weight loss. The patient had no known previous medical problems. Complete blood count revealed leukocytosis (12600 WBCs per microliter) with 6% eosinophilia and anemia (hemoglobin = 9.6 mg/dl,) with a hematocrit of 30.1%. Biochemical analysis of blood was normal except blood sugar level (134 mg/dl). Erythrocyte sedimentation rate was 90 mm/h.

The patient, who was an inhabitant of a rural area, (Shazand city in Markazi province of Iran) had a history of prolonged exposure to domestic dogs and sheep. Under National Tuberculosis control program, three sputum specimens were collected from patient. The direct smear and modified Zeihl Neelsen stained smears were sent to Shazand health center to observe acid-fast bacilli. However, the acid-fast bacilli were not seen on the smear but instead, many hooklets of protoscoleces in different shape and sizes were observed (Fig. 1).

The patient was referred to Shariati hospital in Tehran city. A simple chest X-ray showed a multilobulated mass in the lower part of the left lung (Fig. 2). Computed tomography scan verified existence of thick walled caviar lesion with irregular air–fluid level in favor of a hydatid cyst. In addition, adjacent parenchymal infiltration and fibrosis, pleural thickening and mild pleural effusion were observed in left lung. Right lung was normal. Bronchovascular patterns of both lungs were also normal. Abdominal tomography did not reveal any cysts in the liver. Finally, pulmonary hydatid cyst was confirmed by surgery. A posterolateral thoracotomy was performed and daughter-cysts were evacuated. Albendazole was prescribed in the dose of 15 mg/kg/day as a postoperative prophylactic measure to inhibit recurrence. The patient was discharged on the 10th post-operative day with good and stable vital signs. Six months later, chest X-ray showed a well expanded left lung. In follow up after 6 months and 1 year, the patient showed a complete resolution of all the symptoms with normal chest X-ray.

Fig. 2 Chest X-ray showing multilobulated mass in the lower part of the left lung and a relatively large pneumothorax

Discussion

Hydatid disease is endemic in developing countries especially in sheep raising area, like Iran (Rostami et al. 2016a, b). The clinical manifestations of hydatidosis are variable and depend on the size, location of the lesion and pressure effect of the slowly enlarging cyst in the involving organ (Rostami et al. 2015; Santivanez and Garcia 2010). Hydatid cysts must be differentiated from benign cysts, mycoses, abscesses, caviar tuberculosis, and benign or malignant neoplasms (Moro and Schantz 2009). The liver is the most effected organ followed by lungs (almost 25% of cases) in patients with hydatidosis (Moro and Schantz 2009). Rupture of the pulmonary hydatid cyst (PHC) can occur into the pleural cavity and/or bronchus (Balci et al. 2002). In the present case, cyst rupture was observed into bronchus. Rupture of PHC into bronchus is a significant complication and produce severe cough, chest pain, fever and weight loss (Doğan et al. 1989), as was observed in the present case. Rupture into pleural or pericardial cavity is severe, life-threatening illness and may produce several complications such as bronchopleural fistula, tension

pneumothorax, simple pneumothorax, collapsed lung, pleural thickening, empyema and large residual cavity (Aribas et al. 2002; Balci et al. 2002; Puri et al. 2011). In the present report, patient had rose spots (rash) on the skin of throat and chest and severe coughing. It is reported that hydatidosis rarely cause acute dyspnea and rash. Zingg et al. (2003) reported in a 7 year old girl with diagnosis of ruptured PHC that the respiratory symptoms resolved and the rash disappeared after treatment with albendazole. In hydatidosis, it is reported that eosinophilia is present in <25% of infected patients (Moro and Schantz 2009). Also, it is described that eosinophil rate increases significantly in rupture of cyst (Basavana et al. 2007; Doğan et al. 1989). In the present case, moderate eosinophilia was seen (6%).

The imaging techniques like plain X-ray and Computed tomography (CT scan) are the most important diagnostic tools in lung hydatidosis (Koul et al. 2000; Sahin et al. 2003; Rouhani et al. 2013; Lightowers and Gottstein 1995). Round or oval shape and sharp-edged homogeneous densities are observed for intact cyst in plain chest radiograph, but meniscus sign, water-lily sign, cavity with or without air–fluid levels, pneumonitis and atelectasis of the lung are the most significant features of ruptured cysts. The presence of an air–fluid level is the most prominent radiologic finding in patients with complicated cysts (Aribas et al. 2002). In our patient, we observed a multilobulated mass in the lower part of the left lung in simple X-ray and computed tomography scan verified existence of thick walled caviar lesion with irregular air–fluid level. In addition, adjacent parenchymal infiltration, pleural thickening and mild pleural effusion in the left lung were noticed.

In few cases with diagnosis of suspected PHC, sputum specimen examination represents a non-invasive technique and hooklet/scolices may be observed in smears. This approach could be used as an alternative to biopsy technique under certain conditions (Garcia et al. 1986; Ramos et al. 2001).

The main treatment of PHC is surgical intervention accompanied with chemotherapy as a complementary treatment. This procedure is best choice to remove all of the cysts and avoid the recurrences (Aribas et al. 2002; Balci et al. 2002; Kuzucu et al. 2004). The most appropriate surgical procedures are removal of the cyst membrane, closure of the bronchial openings and capitonnage, but in complicated hydatid cysts decortication, segmentectomy, thoracotomy and lobectomy may be required (Balci et al. 2002; Ramos et al. 2001). In our patients decortication and thoracotomy was performed to remove all of the cysts.

In surgery, patients are exposed to various complications such as, unexpanded lung, empyema, pneumothorax, bronchobiliary fistula and spillage of hydatid contents.

However, our case had an uneventful postoperative recovery and remained asymptomatic in a follow-up of 2 years.

Limitations in the present study are mainly that sensitive serological tests (ELISA or western blot) before surgery and histopathological analysis of the specimen after surgery were not done, which could have been helpful to arrive at definitive diagnosis of PHC.

Conclusion

Hydatid cysts in lung are common in endemic area. In the present report, we described details of ruptured PHC in a boy with severe dry cough and itchy rose spot on the skin of throat and chest, as the most prominent symptoms. The disease was misdiagnosed clinically, as pulmonary tuberculosis. Further, hooklets observed in sputum specimen suggested diagnosis of hydatid cyst. CT scan and chest X-ray confirmed ruptured PHC in this case. After definitive diagnosis, surgery was performed followed by specific drug treatment. Patient was discharged with good and stable vital signs. This report emphasizes that the ruptured PHC should be considered in the differential diagnosis of tuberculosis especially in endemic areas for both infections.

Compliance with ethical standards

Conflict of interest None.

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