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Intrathoracic multiple recurrence and bilateral endobronchial rupture of cyst hydatid disease; the rare cause of anaphylaxis

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1. Introduction

Hydatid cyst is a disease caused by Echinococcus Granulosus and Alveolaris. Often it is localized in the liver and lung. The disease is endemic in Turkey, Mediterranean countries, South Africa, the Middle East, South America and New Zeland [1]. The majority of patients are asymptomatic, with rarely seen signs of cough, dyspnea. Cysts may be symptomatic depending on its size, location and complications. Significant physical examination findings are not available. In pulmonary cyst hydatid disease, well defined round consolidations suggesting cystic lesions can be seen on chest X-ray and surgical treatment is the first choice in this patients. In endemic areas, pulmonary hydatid cyst is a rare cause of anaphylaxis. We present two cases who came to emergency department with anaphylaxis due to different presentations of hydatid cyst disease.

2. Case 1

A 28-year-old woman was brought to emergency department with anaphylaxis. She had hypotension (arterial blood pressure 70/45 mmHg), dyspnea, tachypnea (45/min.) and tachycardia (135/min.). On physical examination, lung sounds were decreased at the left lower zone and there was a left posterolateral thoracotomy scar. Erythrocyte sedimentation rate was measured as 57 mm/h in biochemical tests and there were no abnormality in hemogram and liver function tests.

Quickly we were started anaphylaxis treatment to the patient. Inhaled β2 mimetic, inhaled corticosteroid, methylprednisolone intravenous (120 mg) and intravenous 0.9% NaCl fluid support were given. Then patient was taken to the intensive care unit after improving the overall condition.

Chest X-ray showed left diaphragm is elevated, there were many well defined round consolidations suggesting cystic lesions at the left lower zone and adjacent to the mediastinum (Fig. 1a). Thorax CT scan showed multiple cystic lesions in the left lower lobe (3 × 3 cm), adjacent to the left pericardium (5 × 5 and 2 × 2 cm) and adjacent to the upper mediastinum (2 × 4 cm) (Fig. 1b). Indirect hemagglutination (IHA) test was positive. The abdomen ultrasonography showed no cyst in the liver.

The patient was operated after completion of preoperative preparations. Cysts adjacent to the upper mediastinum and left pericardium were removed by cystectomy (Fig. 2). Cystotomy and capitonnage were performed in the left lower lobe cyst. Pathological diagnosis was Echinococcus alveolaris. After expanded of lung in postoperative 4th day, thorax drain was removed. The patient was discharged on the 10th day of hospitalization.

3. Case 2

A 42-year-old female patient was admitted to our hospital emergency department with a saliva-like vomiting after a severe cough. There was no previous illness story in her anamnesis. On physical examination, body temperature was 38.7 °C, arterial blood pressure was measured 65/40 mm/Hg, there were occasional rashes in the body. She had tachycardia (132/min), wheezing respiration and dyspnea (oxygen saturation 87%). Crepitations were heard at the lower zone of right lung and upper zone of left lung. Hemogram, sedimentation and liver function tests were normal. On chest X-ray cystic lesions were detected in the left upper zone and right lower zone. Then anaphylactic treatment was started to the patient, adrenalin 0.5 mg intramuscular, methylprednisolone 120 mg intravenous, inhaler salbutamol, oxygen (5 L/min.) and bolus 0.9% NaCl fluid intravenous support were given. The patient was

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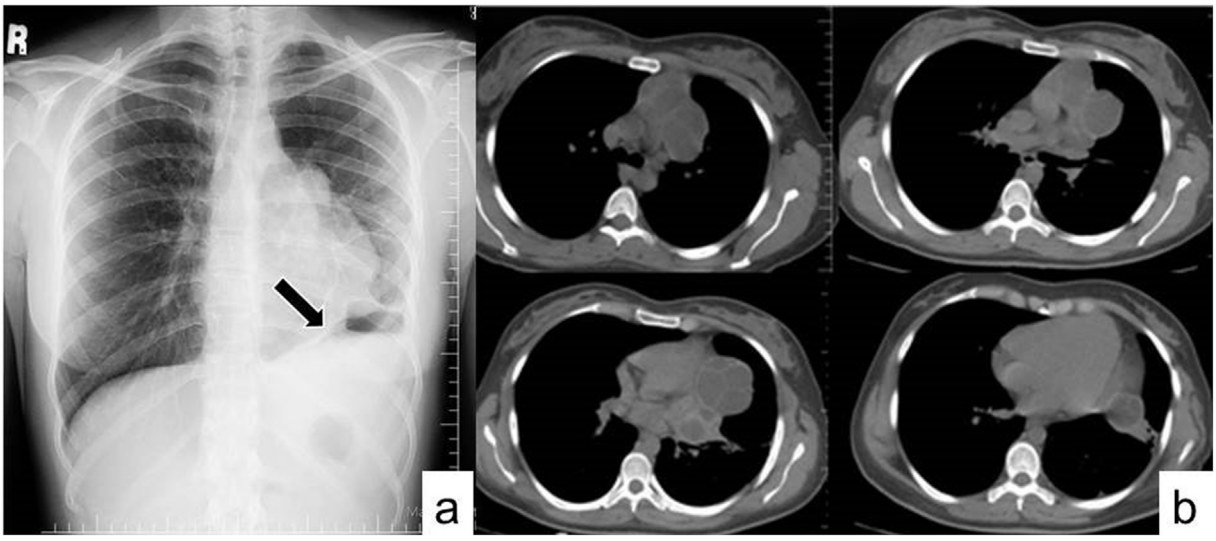

Fig. 1. Chest X-ray shows left diaphragm is elevated (black arrow), cystic lesions at the left lower zone and adjacent to the mediastinum (a). Non-contrast thorax computed tomography axial images shows cystic lesions in the left lower lobe, adjacent to the left pericardium and adjacent to the upper mediastinum (b).

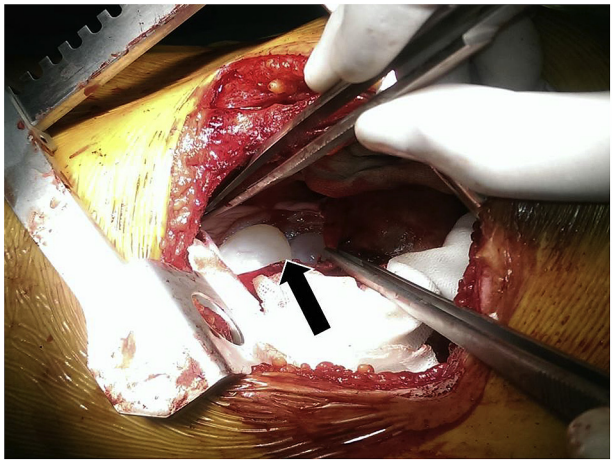

Fig. 2. Operation photo shows cysts adjacent to the upper mediastinum and left pericardium.

taken to the thoracic surgery clinic after improving the overall condition.

In the thoracic CT, perforated cystic areas were found in the left upper lobe posterior segment and right lower lobe superior segment of the lung (Fig. 3). Albendazole prophylaxis treatment was started to the patient (2 × 400 mg oral). Abdomen ultrasonography showed a 8 mm diameter cyst in the liver.

After one week of medical treatment, the patient was operated with bilateral Videothoracoscopy (VATS). Cystotomy and capitonnage were performed and parenchymal protective technique was applied to the bilateral pulmonary cysts. Pathological diagnosis was Echinococcus granulosus.

Thoracic drains removed on the postoperative 3rd day when bilateral expansion seen on chest X-ray. Albendazole therapy was started to the patient 2 × 400 mg and liver function tests were checked every 2 weeks. The patient's 6th months policlinic control was normal.

4. Discussion

Hydatid lung cysts are one of the most common helminth zoonotic diseases in humans, resulting in the infestation of the larval form of Echinococcus granulosus. The adult worm live in the small intestine of the dogs, and leaves a lot of eggs to the environment with the dogs feces. People who come into contact with contaminated food or water become sick. It is seen as endemic in

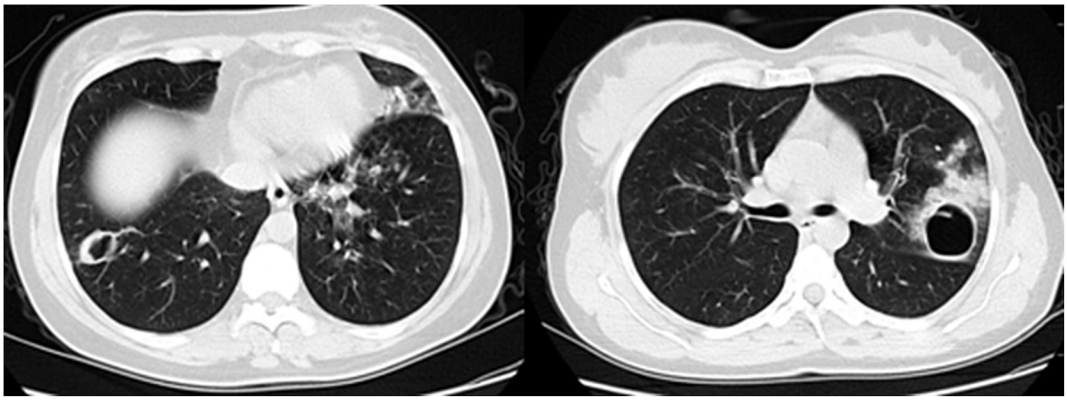

Fig. 3. Non-contrast thorax CT axial images shows perforated cystic areas in the left upper lobe posterior segment and right lower lobe superior segment of lungs.

regions such as Middle Eastern countries where animal breeding is common like eastern part of our country. There are also reports in the literature suggesting that family screening should be performed, especially when a newly diagnosed patient is identified in endemic areas [2].

Echinococci IHA, ELISA IgG, immunoelectrophoresis, indirect fluorescent antibody tests can be used in the diagnosis and follow-up of cyst hydatid [3]. Chest X-ray and thorax CT are the first step in the diagnosis of pulmonary cyst hydatid and MR can be used to evaluate the cyst adjacent to the diaphragm [4]. The majority of patients are asymptomatic, but they can be caused by compression findings according to the location of the cyst, and may also cause anaphylaxis and sudden death. There are many reports in the literature that it is the cause of anaphylaxis [5–7].

In all age groups, the first treatment of lung hydatid disease should be surgery. Recent developments have also been reported with minimally invasive techniques such as VATS [8]. Treatment of hydatid cyst with albendazole after surgery is recommended to prevent recurrences. In our first case, albendazole treatment was not given after the first operation, which posed a risk for recurrence.

5. Conclusion

Liver and lung are the most common sites of hydatid cysts and abdomen ultrasonography should be performed if lung cyst hydatid is detected. Surgery should be the first option in the treatment of lung cyst hydatid.

Treatment of hydatid cyst with albendazole after surgery is recommended to prevent recurrences. In our first case, albendazole treatment was not given after the first operation, which posed a risk for recurrence. When cyst perforation occurs, albendazole treatment should be given without time and anaphylactic precautions should be taken.

Conflict interest

We report no conflict of interest.

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