INTRODUCTION

Primary spontaneous pneumothorax occurs commonly in young females. Spontaneous pneumothorax during pregnancy is quite uncommon, with only 56 cases reported till now.¹ Pneumothorax in a pregnant patient secondary to ruptured pulmonary hydatid cyst is extremely rare and only two such cases are reported in the literature.²³ During pregnancy, oxygen consumption is increased by about 20%. In addition, physiological anaemia of pregnancy and a relatively low partial pressure of oxygen in the umbilical vein of the fetus (approximately 35 mmHg) has little reserve if maternal partial pressure of oxygen starts to decrease.⁴ Any impairment of ventilation during pregnancy may have serious consequences for both the mother and her fetus. Herein, we report a case of ruptured pulmonary hydatid cyst leading to pneumothorax in a pregnant patient and its management.

CASE REPORT

A 19-year-old female patient presented to the emergency room with one-day history of progressive shortness of breath and pain on the right side of the chest. She also gave history of productive cough with on and off low-grade fever for the last 2 months. Chest radiograph taken with abdominal shielding showed a right-sided large pneumothorax (Figure 1).

A right-sided chest tube was inserted and connected to a closed suction device. It showed a large air leak followed by intermittent air leak. The patient had immediate clinical improvement, and her respiratory rate dropped to 20 / minute and pulse to 94 / minute and she was maintaining oxygen saturation of 98% at room air. She was admitted to the ward and treated as a case of right-sided spontaneous pneumothorax. Repeated chest X-ray showed incomplete lung expansion. Patient was assessed by the Obstetrician and obstetrical ultrasound examination showed a single viable fetus with parameters corresponding to a gestational age of 20 weeks.

On the 6th postadmission day, the patient developed low-grade fever along with a rise of white cell count to 18000/mm³. The drainage of the chest tube changed from serous to turbid fluid with persistent intermittent air leak. A CT scan of the chest with abdominal shield was performed. It showed a cavitating lesion in the right middle lobe, surrounded with patchy fibrotic changes. It measured 4.5 x 1.5 cm in transverse and antero-posterior diameters respectively. It was communicating laterally with the pleural space and medially with lateral segment of the right middle lobe, thus creating a ruptured focal lesion, with formation of bronchopleural fistula (Figure 2).

Based on these findings, the differential diagnosis was ruptured hydatid cyst, necrotizing infection (tuberculosis or fungal), or a cavitating malignant lesion.

ABSTRACT

Hydatid disease in pregnancy is a rare condition. Ruptured pulmonary hydatid cyst with pneumothorax during pregnancy is potentially serious for both the patient and the fetus. Diagnosis, treatment, and the mode of delivery of the infant all present complex problems related to this event. We describe here a case of pneumothorax occurring during pregnancy secondary to ruptured pulmonary hydatid cyst with a good outcome for both the mother and the fetus.

Key words: Pulmonary hydatid cyst. Rupture. Pneumothorax. Pregnancy.
Further investigations showed moderately high (1:1024) *Echinococcus* antibody titre and negative AFB staining in sputum.

The patient underwent a right thoracotomy under general anaesthesia. There was about 500 ml of pus in the pleural cavity. A perforated infected hydatid cyst measuring 5.4 cm x 2.3 cm was found in the lateral segment of the middle lobe along with hydatid cyst membrane. A middle lobectomy was performed. All infected debris was removed and thoracic cavity was washed with normal saline. Two chest tubes were inserted. The patient was shifted to the SICU post-operatively. Her postoperative recovery was uneventful and she was shifted to the general ward after 2 days. The patient was continued on broad spectrum antibiotics for 2 weeks.

Postoperatively, obstetrical ultrasound showed a normal viable fetus. On the 7th postoperative day, the output from the chest tubes was nil with no air leak, and repeated chest X-ray showed fully expanded lungs. The chest tubes were removed. The histopathology result also confirmed the diagnosis of hydatid cyst. Infectious disease team recommended continuation of antibiotics for 2 weeks and to start Albendazole after delivery. The patient did well and was discharged home after completing the course of antibiotics. Stitches were removed 2 weeks after surgery.

**DISCUSSION**

Hydatid disease is caused by the tapeworm *Echinococcus (E).* The vast majority of infestations in humans are caused by *E. granulosus*. Humans may serve as intermediate hosts, being infected by contact with infected dogs, or by ingestion of eggs from contaminated food, water or soil. Hydatid cysts form in the lungs in 10-30% of the cases. They are multiple in 30% of the cases, bilateral in 20%, and located in the lower lobes, mostly right, in 60%. Most cysts are acquired in childhood and remain asymptomatic for a long period of time. But cysts may enlarge to more than 20 cm in diameter and cause symptoms by compressing adjacent structures. Cysts may rupture spontaneously, or more commonly precipitated by a sudden rise in the intrapulmonary pressure.\(^5\) The cause could be trivial such as coughing or sneezing, or it may follow an increase in intra-abdominal pressure as in pregnancy.\(^2\) In a communicating rupture, the contents of the cyst escape into the tracheobronchial tree through bronchioles that have been incorporated into the pericyst, with expectoration of a clear salty or peppery tasting fluid containing fragments of hydatid membrane and scolices. Fever and acute hypersensitivity reactions ranging from urticaria and wheezing to life-threatening anaphylaxis may be the principal clinical manifestations.\(^5\) Rupture of hydatid cyst in the pleural cavity can lead to pneumothorax and bronchopleural fistula.\(^3\) The most important diagnostic tools in pulmonary hydatid cyst are plain chest X-ray, contrast enhanced CT scan of the chest, and immunological tests. Radiologically an intact cyst typically present as a round or oval homogenous density with sharp margins. In case of ruptured cyst, the solid component will fall into the most dependent part of the cavity (mass within a cavity).\(^6\) Immunological screening is positive in only 50% of the patients with pulmonary hydatidosis.\(^7\) For standard chest radiography with a shielded maternal abdomen, the dose to the uterus is estimated to be 1 mard per examination. A single series of chest CT scan at 1 cm interval exposes the conceptus to an upper limit of less than 1 rad, and the actual dose is likely closer to 0.5 rad. No increased rate of anomalies have been reported in children exposed to less than 1 rad of ionizing radiations in utero.\(^8\)

Pregnancy and pulmonary hydatidosis occurring together is very rare and there is no consensus on standard approach.\(^2\) Chemotherapy (Albendazole, Mebendazole) is used as a complement to operative treatment to avoid recurrence but both drugs are contraindicated in pregnancy (especially during the first trimester) because of possible teratogenicity. However, the treatment of pulmonary hydatidosis is mainly surgery. The management options for lung cysts include lobectomy, wedge resection, pericystectomy, intact endocystectomy and capitonnage.\(^4\) In cases of ruptured cyst, thoracotomy is undertaken for the removal of the residual wall of the ruptured cyst, and other ipsilateral cysts.\(^10\) The lung cavity that remains after removing the cyst may be left as it is or obliterated by sutures from within the cavity in regard to the size and location of the cyst. The bronchial openings in the cavity must be closed by non-absorbable sutures e.g. prolene 3/0 in all cases.\(^11\) In this case, most of the middle lobe was destroyed, so a middle lobectomy was performed.

Pneumothorax should be considered in any pregnant woman with chest pain and/or dyspnea and must be confirmed radiographically. Perforated pulmonary hydatid cyst should be kept in mind in the differential diagnosis of pneumothorax in a pregnant woman in endemic areas. Initial management of large pneumothorax during pregnancy is by tube thoracostomy. Definitive management of ruptured pulmonary hydatid cyst is surgery. A multidisciplinary team approach is needed to optimize the maternal and fetal outcome in these critical situations.

**REFERENCES**


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