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Case Report

MANAGEMENT OF COMPLICATED HEPATIC HYDATID DISEASE WITH CONCOMITANT SPLENIC HYDATIDOSIS

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ABSTRACT

We report the case of a middle aged women presenting with complicated hepatic hydatid disease and concomitant splenic hydatid cysts. A 35 year old female presented in emergency department with complaint of breathing difficulty for 3-4 days. She had a long standing history of asymptomatic intra abdominal swelling. Radiological investigation was suggestive of transdiaphragmatic intrathoracic rupture of hepatic hydatid cyst with concomitant splenic hydatid cysts. She underwent chest drainage, ERCP and biliary stenting followed by definitive surgery. The patient recovered after surgery and continued prophylactic albendazole therapy. Follow up scan after 3 months showed no recurrence. Hepatic hydatid disease complicated by simultaneous intra thoracic and intrabiliary rupture is a serious condition. Successful outcome require multimodality approach with surgery as a mainstay therapy.

Key Words: Complicated Hepatic Hydatid Cyst, Biliopleural Fistula, Splenic Hydatidosis, Multiple Hydatidosis

CASES

Hydatid disease is a zoonotic disease with worldwide distribution. Hydatids cysts are most frequently seen in liver (50-75%) followed by lung (25%) and about 5-10% are distributed along the arterial system (Xiao *et al.*, 2006). The following case report is of a middle aged woman with concomitant liver and splenic cysts complicated by intrabiliary and transdiaphragmatic intrathoracic rupture of hepatic cyst.

A 35-year old female presented in emergency department of our hospital with the chief complaint of breathing difficulty for the last 3-4 days. On examination, the patient was dyspneic with Spo2 of 88% on room air. There was a marked decrease of air entry in the right side of chest. There were two well defined palpable lumps in the right and the left hypochondrium which collaborated with a long standing history of upper abdominal swelling. X ray chest revealed right hydropneumothorax with typical water lily sign.

Thoracoabdominal CT scan showed two large hepatic cysts in the segment 4 and caudate lobe of the liver and three large cysts in the spleen. Right pleural cavity showed collapsed right lung, hydropneumothorax, left sided mediastinal shift with the possibility of ruptured hepatic hydatid cyst into pleural cavity and associated bronchial fistula (Figure 1).

The patient was unable to maintain saturation despite adequate oxygen supplementation and required mechanical ventilation. In view of mediastinal shift and compromised respiratory function, a chest tube was inserted in the right pleural cavity. About 3.0 liters of biliopurulent fluid and a lot of hydatid membranes were evacuated. The patient was started on albendazole therapy through Ryle's tube. Subsequent fluid output from the chest tube was bilious, about 300-400 ml per day with associated air leak suggestive of both bronchial and bilious fistula. However, the endodtracheal tube secretions were nonbilious indicating absence of bronchiobiliary fistula. ERCP done on day 7 post chest tube insertion revealed fistulous communication between left bile duct and the right pleural cavity. A 7 Fr double pigtail stent was deployed in the left ductal system. However, there was no significant decrease in bile leakage post stenting. The patient underwent laparotomy on day 13 of admission for persistent sepsis and continuous leakage of bile from chest tube. The peroperative findings revealed collapsed cysts in segment 4a, 4b and caudate lobe of liver. Right hemiliver was adherent densely to diaphragm and on mobilization a 4.0 cm x 2.0 cm defect was visualized in the diaphragm (Figure 2).

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Figure 1: CT scan showing rupture of hepatic cyst into right pleural cavity with mediastinal shift and concomitant splenic cysts



Figure 2: Diaphragmatic defect

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On opening the cyst cavity, a 1.5 cm sized biliary communication with stent in situ was identified. There were 3 large cysts in the spleen arising from superiomedial, inferiomedial and inferiolateral surfaces respectively. Superiomedial cyst was retrogastric in location and adherent to pancreas whereas inferior cysts were adherent to transverse colon. Hepatic cyst deroofing, interrupted suture closure of the biliary defect followed by omental packing was done. Diaphragmatic rent was repaired with nonabsorbable suture. Splenic cysts were dealt with deroofing, evacuation of cyst contents and omentopexy (Figure 3).

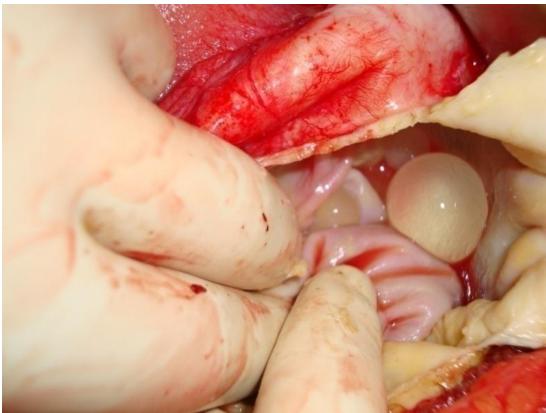


Figure 3: Splenic cyst deroofing and daughter cyst evacuation

The postoperative recovery was uneventful except for the wound infection and persistent air leak from the chest tube. The patient was discharged with chest tube in situ and was continued on prophylactic therapy with albendazole. On follow up, she remained asymptomatic; the chest tube was removed after cessation of air leak 2 months postoperatively.

DISCUSSION

Hydatid disease is endemic in farming areas of world. Overall 75% patients are identified incidentally (Tolgamuftuoglou *et al.*, 2005). Complications are observed in one third of patients with hepatic hydatid disease. Intrabiliary rupture has been reported in 5-25% of the cases (Wagholikar *et al.*, 2002). Intrathoracic rupture has been reported in approximately 3% of cases and is associated with high mortality (Crausaz, 1967). Hydatid disease in spleen is rare with reported incidence of < 5% (Meimarakis *et al.*, 2009). The patient was asymptomatic for years despite being aware of abdominal mass. The patient became symptomatic secondary to rupture into thoracic cavity with accompanying intrabiliary rupture. Usually transpleural approach has been advocated by many surgeons (Crausaz, 1967). However thoracic complications can be dealt by various other approaches like thoracophrenolaparotomy, laparotomy, thoracotomy and laparotomy. Surgery related mortality has been reported to vary between 0.5% and 40%

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Topuzlar (2009). Splenic hydatid is conventionally dealt by splenectomy Meimarakis et al., (2009). Splene preserving surgery is an alternative treatment option (Meimarakis *et al.*, 2009). Splenectomy would have been a formidable task in this case because of compromised general condition and accompanying perisplenic, pericystic adhesions. Therefore, only cyst deroofing, evacuation of cyst contents and omentopexy was done.

Hepatic hydatid disease complicated by simultaneous rupture into biliary and thoracic cavity is a serious condition. Surgical intervention is mainstay of therapy. Successful outcome requires multimodality approach in form of surgical, endoscopic and drug therapy.

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