CASE REPORT

Advanced Form of Hepatopulmonary Hydatidosis in a Child: Case Report

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ABSTRACT

Hepatopulmonary hydatidosis (HPH) is a very rare condition in children with a prevalence of 11%. An 8-year-old girl with advanced HPH was successfully treated in our institution without complications. The coexistence of large numbers of high hydatid cyst makes this case very unusual and interesting.

Keywords: Children, hepatopulmonary hydatidosis, treatment.

INTRODUCTION

Hydatid disease is the most widespread potentially very dangerous human parasitic infection caused by the larval stage of *Echinococcus granulosus*. Hydatidosis mostly occurs in the adults, although, in the socially neglected and endemic areas, it is very common in the paediatric population. The liver is predominant site (70%), following lungs (15%) (1). Multilocular hydatidosis occurs at a frequency up to 11% (2, 3). Concomitant pulmonary and hepatic infection, which represents a distinct clinical entity called hepatopulmonary hydatidosis (HPH) (4), is more common in adults (5).

CASE REPORT

An 8-year-old girl was initially treated at the primary centre for bloated abdomen, fever up to 38°C and pain below the right rib. She had no vomiting, and stools were regular. Initial abdominal ultrasound confirmed sharply limited cystic formations that could fit hydatid cysts. Diagnosis was confirmed by the positive echinococcus test. The patient was sent to the Department of Infectious Diseases in tertiary centre. Multiple sliced computed tomography (MSCT) revealed multiple cystic changes in the liver (Fig. 1A, 1B), mesentery of the right lung (Fig. 1C), up to 90 mm in diameter, followed with mild intra and extrahepatic bile duct dilatation, with no kidney abstraction, but the right kidney shifted dorsally and caudally. According to recommendations, albendazole therapy was initiated by an infectious disease specialist for 1 month. She was treated with 2 × 100 mg per day with no changes in haemogram. The patient was referred to the paediatric surgeon who suggested surgery. Clinical examination revealed very distended abdomen, above the chest level, with several palpable large cysts (Fig. 2). Laboratory tests revealed direct bilirubin 9.48 umol/L, lactat dechidrogenase 501 U/L, aspartat aminotransferase 55.9 U/L, alanin aminotransferase 28 U/L,

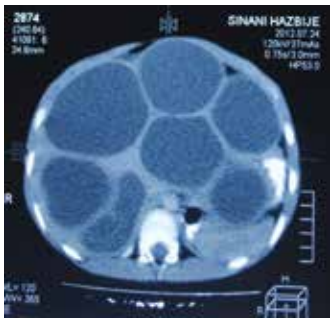

Figure 1A

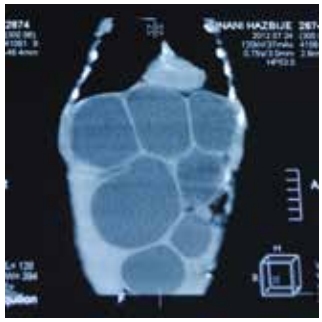

Figure 1B

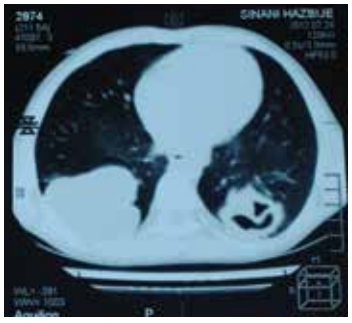

Figure 1C

Fig. 1 (A and B): Multiple sliced computed tomography examination revealed multiple cysts in the liver; (C) cyst in the lower right lobe, 3 cm in diameter.

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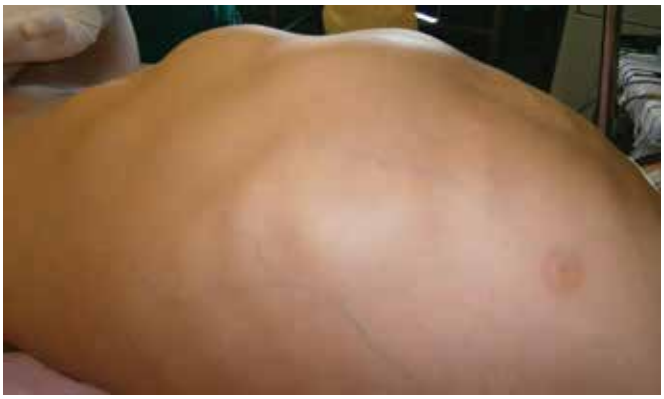

Fig. 2: Clinical examination revealed dilated abdomen, above the chest level, with several palpable large cysts.

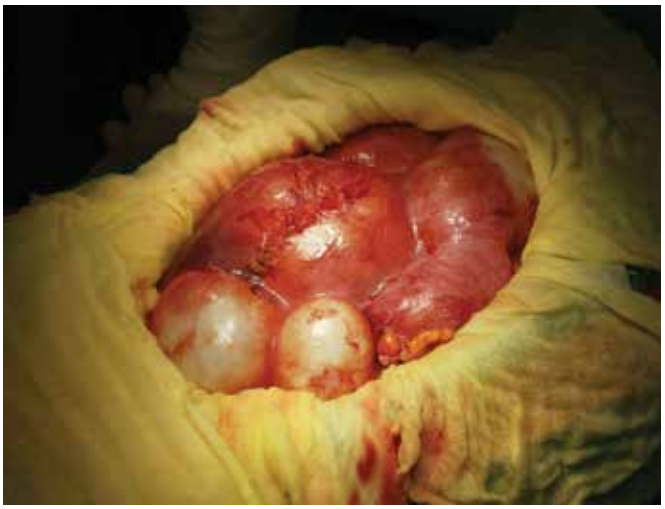

Fig. 3: Multiple cysts are localized and surrounded by sterile gauze sponged with rivanol solution in order to prevent the spillage of daughter cysts into the peritoneal cavity.

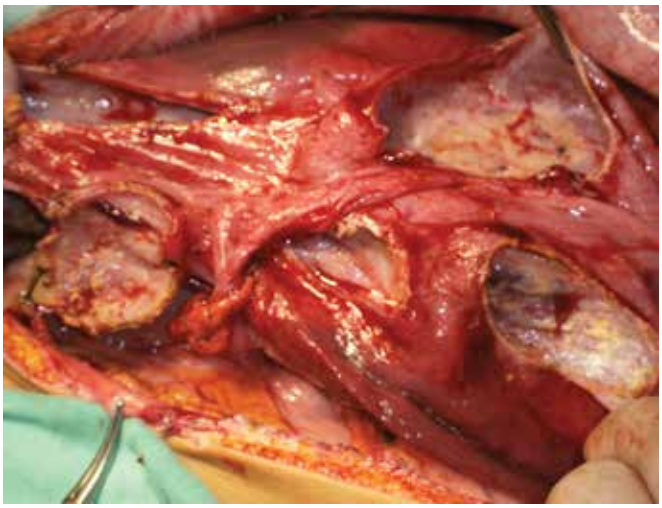

Fig. 4: Total cystectomy and partial pericystectomy of 11 individual cysts were performed. Pericystic remains were reapproximated with resorptive sutures. There was no biliary leakage.

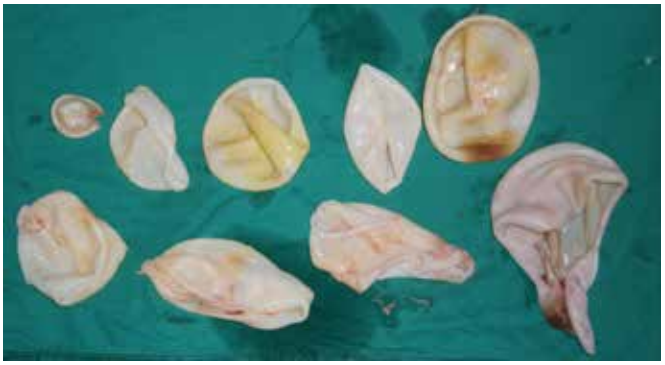

Fig. 5: Nine of eleven germinal membranes from the hydatid cysts.

gamma-glutamyl transferase 64 U/L , protein 46.5 g/L, albumin 24.7 g/L, and C-reactive protein 105.6 mg/L. Blood tests were almost in the normal range, except slightly elevated white blood cells (18.8×10^9/L). A single-stage surgical cystectomy was considered. After liver mobilization, cysts were localized and then surrounded by sterile gauze sponged with rivanol solution in order to prevent the spillage of daughter cysts into the peritoneal cavity (Fig. 3). All the cysts were initially punctured with the germinal membrane shriveling, and releasing a yellow fluid and the membrane from cyst cavity. Cavities were irrigated with saline solution. Total cystectomy and partial pericystectomy of 11 individual cysts were performed (Figs. 4 and 5). Pericystic remains were reapproximated with resorptive sutures. There was no biliary leakage. The abdominal cavity was drained with two drains. After 3 days in the intensive care unit, and 15 more days in the paediatric surgery department, the patient was released home. Postoperative course was uneventful, and albendazole treatment was continued for another 3 months. Control MSCT scan of the liver and lung confirmed the absence of cysts. To date, there has been no recurrence of the disease.

DISCUSSION

Hydatid disease is a consequence of parasitic infestations of *Echinococcus* that occurs by hand-to-mouth transfer of tapeworm eggs from dog faeces. Logically, the infection is more common in the paediatric population due to the poor hygiene. Ingested ova passes through the intestinal wall into the portal system and settles in the liver, or enters into the systemic circulation to invade lungs, brain or other organs (6). Daughter cysts usually invade the lungs, and rarely brain tissue, kidneys, spleen, and intestines, posing multiorgan hydatidosis. Symptoms are mostly consequence of the space occupying effect of

the enlarging cyst in a confined space (jaundice, biliary obstruction and pneumothorax), or anaphylactic shock. Distinct clinical entity named HPH represents synchronous occurrence of pulmonary and hepatic cysts observed in less than 10% of cases (7). Hepatopulmonary hydatidosis typically does not occur in childhood. The most frequent complaints reported in symptomatic patients with HPH are cough, chest pain, dyspnoea, and haemoptysis (8). In our case, 'mute' clinical presentation of HPH was atypical.

Diagnosis is based on clinical examination, serological tests, ultrasound, chest radiography, and computed tomography scans. Serological test of indirect haemagglutination represents highly specific test but not sufficiently trusted (sensitivity rate is 60%–80%). Surgical resection and cyst enucleation from infected organ is the mainstay of treatment. Generally accepted attitude is based on surgery with concomitant pre- and post-interventional chemotherapy (albendazol) (9). Giving albendazole is based on the fact that it reduces the tension in the cyst wall (low risk of intraoperative rupture), reduces the possibility of anaphylaxis and sterilizes cystic content. Lung cysts, especially in young people, are more responsive to carbamate therapy (10). Carbamates significantly reduce risk of recurrence and post-operative complications (11, 12). In the post-operative course, regular follow-up is the key to detect any recurrence. Introduction of radical surgery for HPH in a child is the subject of debate. Conservative approach with maximum preservation of the affected organs is more acceptable. In the past two decades, percutaneous minimally invasive procedure is developed as the alternative to aggressive surgery, but only in cases of solitary cystic formations. It represents a method of cyst puncture and evacuation of cyst fluid under sonografic guidance. Injection of a scolicidal agent (to destroy germinal layer) and reaspiration of cyst remains (endocyst) follow (13). In our case, due to multiple cysts in the liver, the conservative treatment was not taken into consideration.

CONCLUSION

Simultaneous involvement of both liver and lung (HPH) is less frequent in children. With early diagnosis and proper managements, HPH in children has an excellent outcome. Surgical resection and cyst enucleation is the mainstay of treatment with concomitant pre- and post-interventional chemotherapy (albendazol).

Hence, more extensive follow-up studies are necessary for better understanding of HPH, its recurrence and complications, and need for use of scolicidal agents and drugs.

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