

Case report

Primary hydatid disease of diaphragm with subcutaneous extension

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Abstract

While diaphragmatic hydatid disease is a rare condition, subcutaneous extension of the disease is very rare. A 33-year-old female visited our clinic due to swelling on the right upper quadrant of the abdomen. Thoraco-abdominal CT scan and MRI revealed a hydatid cyst (110 x 98 x 78 mm) located in the costal part of the hemidiaphragm extending into the preperitoneal space and protruding intercostally to a subcutaneous area. Total pericyst resection without opening the cyst, including a margin of normal diaphragm, was performed. The patient was started on albendazole for three months. Hydatid cyst in the diaphragm should be considered in patients with preoperative imaging data indicating cystic lesions adjacent to the diaphragm, especially for cystic lesions encountered in patients who live in or who have come from endemic regions.

Key words: diaphragm; hydatid disease; subcutaneous extension; surgery

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Introduction

Hydatid disease (HD) occurs predominately in the liver (60–70%) [1], but it can also arise in any part of the body [2]. Diaphragmatic localization incidence is around 1%, and most cases are generally associated with liver hydatidosis [3]. Subcutaneous location or extension of HD is even more scarce [4]. We present a unique case of a primary large HD of the diaphragm protruding subcutaneously in the preperitoneal space.

Case report

A 33-year-old female visited our clinic with the main complaint of swelling on the left upper quadrant of the abdomen during the last ten years that had been gradually increasing in size, especially in the last year. On physical examination, a painless, fluctuant, 6 x 8 cm mass was palpated. The overlying skin was intact. A pre-operative indirect hemagglutination (IHA) test (Hydatidose, Fumouze, Levallois-Perret Cedex, France) detected positive serologic titers (1/256). Ultrasonography revealed a giant cyst measuring 11 x 10 cm which included multiple daughter cysts resembling HD. Thoraco-abdominal computed tomography scan revealed an HD cyst (110 x 98 x 78 mm) located in the

abdominal surface of the costal part of the hemidiaphragm extending into the preperitoneal space and protruding intercostally into a subcutaneous area (Figure 1); other organs were normal. Thoraco-abdominal magnetic resonance imaging revealed a giant HD cyst (11 x 10 cm) including multiple daughter cysts that was engaged to the diaphragm. The patient had no history of surgery for HD. The patient was diagnosed with primary superficial HD located in the diaphragm and after a preoperative evaluation she underwent surgery for removal of the cysts.

During surgical exploration under the midline incision, the preperitoneal areas were dissected to the left costal part of the diaphragm. When the skin and subcutaneous layers were incised, an 11 x 10 cm mass was reached (Figure 2). The cyst was easily dissected without damaging the peritoneum. Then the cyst was dissected from the diaphragmatic muscle, leaving a thin fibrous membrane on the diaphragm surface in a small area. There was no need to repair the diaphragm. A germinative membrane was noticed after opening the cyst (Figure 3). The surgical site was irrigated with 40% povidone iodine (Betadine, Kansuk, Istanbul, Turkey) and hypertonic saline (3% NaCl). The subcutaneous layers and skin were closed

Figure 1. Thoraco-abdominal computerized tomography scan showing hydatid cysts located in the left hemidiaphragm, which is protruding intercostally into subcutaneous space and preperitoneal extension

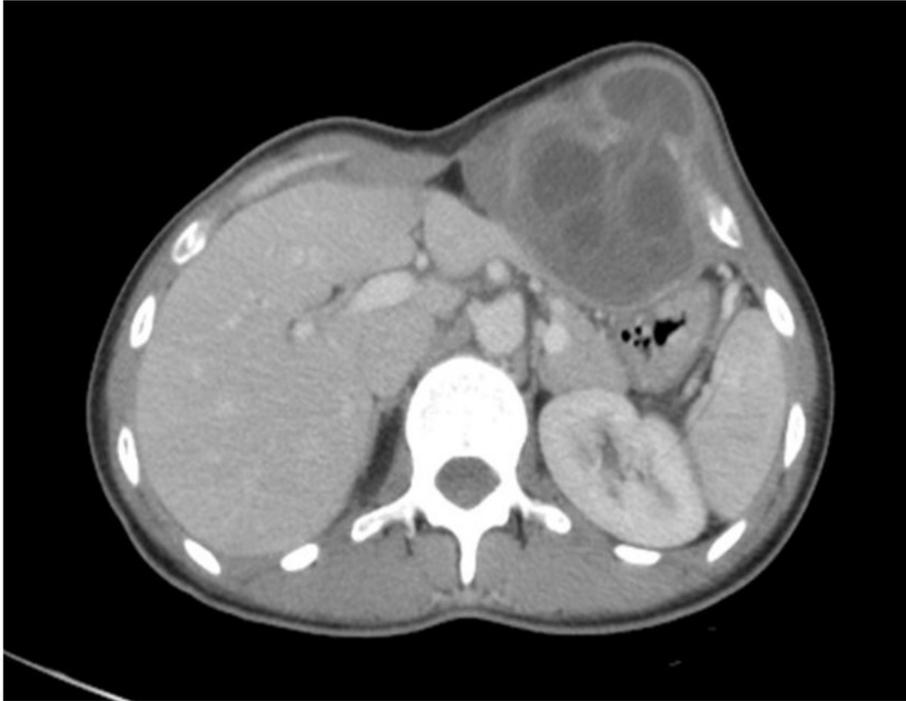


Figure 2. Surgical exploration revealing 11 x 10 cm cyst in the preperitoneal area

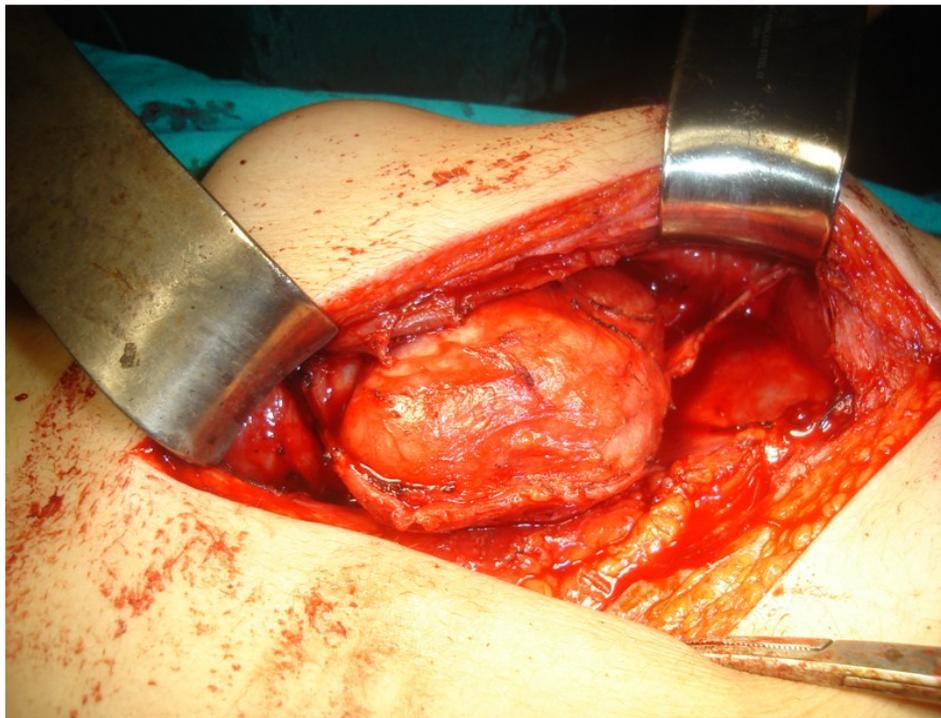
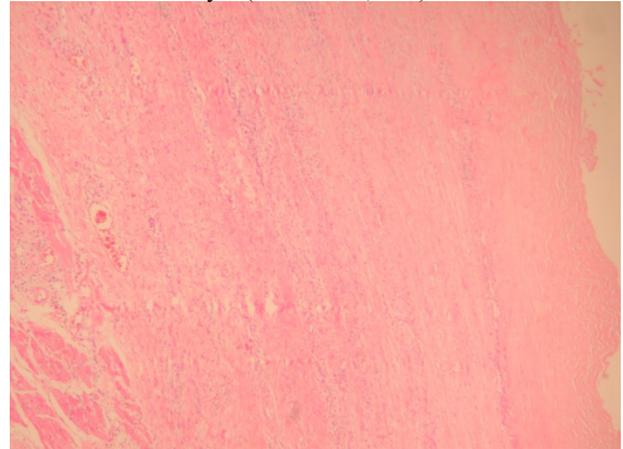


Figure 3. A germinative membrane in the cyst**Figure 4.** Fibromuscular tissue covered with cuticular membrane of the cyst (H&E stain, x40)

in the standard manner. Histopathological examination revealed a hydatid cyst (Figure 4). No postoperative complication developed. The patient was started on albendazole for three months (10 mg/kg/day). No findings associated with local or systemic HD were detected during the follow-up period.

Discussion

The mechanism of primary diaphragmatic HD is unclear. After oral ingestion, the oncosphere penetrates the intestinal wall then joins the portal system and reaches the liver. Parasite eggs can pass into the systemic circulation and cause disease in other end organs. Larvae must pass through two filters (liver and lung) to form a solitary HD. It is very possible that systemic dissemination via the lymphatic route accounts for cases with solitary cysts in uncommon sites [5]. Direct spread from adjacent sites may be another mechanism of infection provided a microrupture has occurred [6]. In our case, the first explanation is more acceptable since neither hepatic nor pulmonary HD was detected.

Familiarity with imaging findings, especially in patients living in countries where this disease is endemic, provides important advantages in making the diagnosis. Despite the characteristic imaging findings, HD in unusual anatomic locations may make differential diagnosis difficult, even in patients from endemic regions [2]. Our case was diagnosed according to the appearance of the mass on different imaging techniques.

Total cyst resection including a margin of normal diaphragm gives the best chance for cure since it removes the parasite and prevents hydatid dissemination [7,8]. This approach is suitable for small cysts. Total cysto-pericystectomy, removing the pericyst without opening the cyst, is feasible in superficial HD of the diaphragm where dissection in a correct plane of cleavage leaves thin fibrous membrane on the diaphragm surface. However, in large cysts, a diaphragm defect is usually difficult to reconstruct and prosthetic materials may be needed to prevent diaphragmatic hernia [8,9]. In the present case, the mass was rough, compact, multinodular, and linked with thin fibrous membranes on the diaphragm surface in a small area. Total cyst excision was performed in this case and the surgical area was irrigated with protoscolicidal agents. Diaphragm repair was not necessary.

Albendazole (10 mg/kg) treatment for a three-months period has better results for preventing postoperative recurrence of the HD [10]. We prescribed our patient a dosage of 10 mg/kg per day for 3 months.

HD in the diaphragm should be considered in patients with preoperative imaging data indicating cystic lesions adjacent to the diaphragm, especially when encountered in patients who live in or have come from endemic regions and if any of the specific imaging features such as calcification, daughter cysts and/or intracystic membranes are seen. Total excision of the cyst is the treatment of choice in diaphragmatic HD.

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