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Primary Peritoneal Hydatidosis: Case Presentation

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ABSTRACT

Primary peritoneal hydatidosis is rare even in endemic areas and diagnosis is based mainly on imaging methods. We report a case of primary hydatidosis in a 5-year-old girl admitted for left upper quadrant pain. Ultrasound examination revealed a large, multicystic lesion occupying the left upper quadrant and the left lumbar region. Computed tomography showed a giant peritoneal cyst compressing the spleen and several smaller cysts without any organ involvement. The patient was surgically treated with partial cystectomy of the large cyst, adherent to the spleen and colon serosa, and complete excision with partial omentectomy for the other peritoneal cysts. Antihelmintic treatment (Albendazole) was administered prior to and after surgery. No recurrence was noted for 6 months after surgery. In conclusion, primary peritoneal hydatidosis remains a rare situation even in endemic countries, making the diagnosis difficult. It should be considered in the differential diagnosis of any abdominal pain or cystic lesion.

Keywords : primary peritoneal hydatidosis, pediatric patient, abdominal pain

INTRODUCTION

Hydatidosis, also known as echinococcosis or hydatid cyst, is a cosmopolitan anthropozoonosis due to the presence and development in humans of the larval form of the dog tapeworm of genus *Echinococcus granulosus*. The geographical distribution of *Echinococcus granulosus* is directly linked to human-dog-sheep contact and represents a real public health problem with worldwide distribution, being endemic mostly in developing countries.

In practice, all other organs or tissues can be affected once the hexacant embryo passes into the systemic circulation but liver and lungs account for more than 90% of cases of echinococcosis (Tsaroucha, AK. et al., 2005). Brain, kidney, spleen and bone usually represent secondarily involved organs in 1 to 5% of cases. Other rare locations cited in the literature are the thyroid gland, ovaries, pancreas, skin, muscle, and heart (0.5-2%) (Yuksel, M. et al. 2007).

Commonly, peritoneal hydatidosis may develop secondary to spontaneous or accidental rupture of liver or spleen cysts during surgery.

Primary peritoneal hydatidosis is rare and according to the literature it represents less than 2% of intra-abdominal hydatid disease cases. Clinical diagnosis of primary peritoneal hydatidosis is difficult due to absence of typical clinical symptoms, the clinical examination being unspecific (Sable, S. et al., 2012; Karavias, D.D. et al., 1996; Ramji, S. et al., 1987).

CASE PRESENTATION

We report the case of a 5-year-old girl admitted to the Clinic of Pediatric Surgery Clinic (Iasi Children's Emergency Hospital, Romania), for left upper quadrant pain for a month. The pain was initially dull, non-radiating, continuous, and later intermittent, without aggravating or relieving factors. The patient had no personal history of fever or chills, lived in a rural area and had close contact with family dogs.

Prior to hospital admission, the child had experienced 5-6 loose stools and several no-blood stained, watery and no-bilious vomiting episodes.

Clinical examination revealed a painless, immobile, and not tender mass, measuring 15 × 9 cm, cystic in consistency and occupying the left hypochondria and left lumbar region. The patient had a weight of 25 kg for a height of 108 cm (BMI 21.5), and no cardiovascular or respiratory pathology.

All blood test proved normal except a mild eosinophilia. Stool examination did not identify any parasites.

Abdominal ultrasonography revealed a large, multicystic lesion of 19 x 10 cm occupying the left upper quadrant and the left lumbar region, closely abutting the spleen, suggestive of a hydatid cyst.

Computed tomography (CT) has been indicated and identified a large cystic lesion of 18 x 12 x 15 cm with fine debris, two wall calcifications and multiple small cysts within the large cyst, an aspect compatible with daughter cysts. The large cystic lesion compressed the spleen with no fat plane separation. Other cystic lesions were identified in the peritoneal cavity notably in the left iliac fossa close to the descending colon (7.7 x 7 cm) and in the pouch of Douglas (5 x 2.5 cm). No cysts were identified in the liver, lung or other parenchy-

matous organs. CT examination raised suspicion of disseminated intraperitoneal hydatid disease.

IgG antibody test against *E. granulosus* (ELISA) revealed positive results.

DIFFERENTIAL DIAGNOSIS
Considering the age of the patient, hydatid disease should be included in the differential diagnosis of all omental cysts, especially in endemic areas.

TRAITEMENT
Antihelmintic treatment (Albendazole 200 mg orally twice a day) was administered for three weeks before surgery.

Laparotomy revealed a large cyst extending to the pelvic cavity, with upward and lateral displacement of the spleen and left kidney. All other cysts described in the CT scan report were identified during surgery (Fig. 1). The large cyst was adherent to the surface of the spleen and colon serosa, partial cystectomy with hypertonic saline (3%) irrigation of the pouch being preferred in this case. Complete removal with partial omentectomy was possible for the other cysts. Precautions were taken in order to prevent any spillage, the intestine and other organs being isolated with towels soaked in 3% saline solution. The peritoneal cavity was lavaged both with hypertonic and isotonic saline solution.

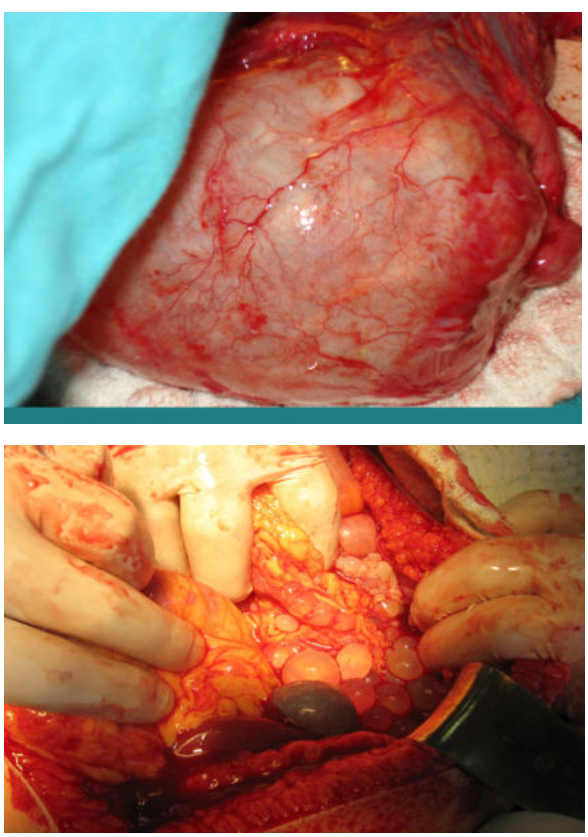

Fig. 1. The large left upper quadrant cyst and the other omental cysts

Histopathological examination of the samples revealed a structureless, laminated, eosinophilic membrane consistent with hydatid cyst wall.

After surgery the patient continued the antihelmintic treatment (albendazole 200 mg twice daily) for 3 months to prevent recurrence.

RESULTS AND FOLOW-UP
The patient was released the 10th postoperative day after an uneventful evolution. Monthly evaluations (clinical and ultrasound) showed no sign of recurrence for 6 months and the girl remained asymptomatic throughout this period.

DISCUSSION
Peritoneal hydatidosis accounts for 10-16% of the intraperitoneal hydatid cysts, occurring mainly secondary to spontaneous or accidental rupture of a hepatic or splenic cyst during surgery (Karavias, D.D. et al., 1996; Acharya, A.N., & Gupta, S. 2009).

Primary peritoneal hydatidosis is rare representing only 2% of cases of abdominal hydatid disease. The mechanism of primary peritoneal involvement is unclear; dissemination by lymphatic or systemic circulation being suspected.

The diagnosis of hydatid disease should be suspected when a cystic mass is palpated in the abdomen, especially in endemic areas with a history of livestock or owning pets. Patients usually remain asymptomatic for many years the diagnosis being made after a series of clinical, imaging, and laboratory tests (especially serological). As hydatid disease can present with a wide range of symptoms it has to be considered in the differential diagnosis of any abdominal pain. The differential diagnosis of hydatid cyst includes mesenteric, ovarian and renal cysts, lymphangioma, intra-abdominal abscess, hematoma etc. (Ionescu, A., et al., 1985; Prousalidis, J., 1998).

In primary peritoneal hydatidosis, the hemogram may reveal mild hyper eosinophilia. Liver and kidney tests usually prove normal.

Ultrasound and abdominal CT may confirm the diagnosis of hydatid cyst, identify the affected organs and describe the anatomical relationship with surrounding structures with a sensitivity of 90 - 95% (Daali, M., et al. 2000; Majbar, M., et al. 2012).

In most cases, ultrasound identifies single or multiple well-defined unilocular cystic lesions or cystic lesions with daughter cysts or hydatid sand. Changing the patient position during examination can help visualizing hydatid sand (made of hooks and scoleces). When fluid pressure in the cyst rises, it may lead to detachment of the inner membrane (endocyst) seen within the original cyst - the snake (or serpent) sign. Collapse of the endocyst gives the aspect of debris floating in the remaining fluid at bottom of the original cyst - the water lily sign. According to the WHO Informal Working Group on Echinococcosis (WHO-IWGE) Classification based on ultrasound images (more recent and standardized than the older Gharbi classification), our patient displayed type 2 hydatid cysts.

CT is very sensitive (95-100%) in detecting hydatid cysts and shows rounded, well defined hypodense lesions, with a thick wall and peripheral contrast enhancement. Fine calcifications in the wall of the cyst can be identified on CT.

Immunoelectrophoresis is a more sensitive test for antihydatid antibodies but ELISA is more specific.

Although hydatid disease is widespread and treatment is well known, patients may present complications such as compression of various structures, infection, and rupture with secondary spread or anaphylactic shock, fistulisation to adjacent cavitary organs or portal hypertension.

The aim of treatment for hydatid disease is to remove the existing cysts and prevent recurrence and complications. Medical treatment implies the administration of albendazole/praziquantel alone or adjuvant to surgery, depending on the size, location and distribution of cysts.

Surgery, either radical or conservative, remains the treatment of choice, especially in case of large or multifocal hydatic cysts. The total or partial cystectomy with or without drainage or omentoplasty forms the basis of hydatid surgery. In case of a cyst adherent to the abdominal wall, a percutaneous aspi-

ration of cystic content followed by injection and aspiration of alcohol (PAIR) may be used.

The type of surgical treatment needs to be individualized for each patient. Although excision of the intact cyst is ideal, it is not always possible. In such cases, partial excision (if the cyst is close to vital structures) with enucleation and omentoplasty remains the treatment to choose. The new PAIR therapy shows a good success rate, but patients have to be monitored for a longer period of time in order to prevent recurrent disease. During the first 6 months after surgery for peritoneal hydatidosis, our patient registered no clinical or ultrasound recurrence.

CONCLUSIONS

Primary peritoneal hydatidosis is very rare, accounting for 2% of cases of abdominal hydatidosis. Clinical polymorphism, latency and severity of complications represent its main features. Hydatidosis registers a slow but potentially life threatening evolution. Physicians must maintain a high degree of clinical suspicion in all cases of cystic abdominal masses especially in endemic areas, because the clinical signs and symptoms may be nonspecific for a long period of time.

The diagnosis of hydatic cyst is made after several clinical, imaging and immunological tests. The severity of the natural evolution of hydatidosis requires surgical treatment in a short period of time after diagnosis, but the choice of the surgical procedure depends on the final assessment of the lesions. A careful and complete surgical excision of the cysts represents the only curative treatment. As always in medicine, the best treatment is prevention, which is essential for the control and reduction of the prevalence of human hydatidosis. Adequate preoperative and postoperative medical care prevents recurrence. Long-term follow-up is needed for detection and treatment of recurrent disease.

The particularity of our care consists in primary peritoneal location and the fact that the complete involvement (adherences) was not established by imaging techniques.

The authors declare that there are no conflicts of interest.

REFERENCES

[1] Tsaroucha, A.K., Polychonidis, A.C., Lyrantzopoulos, N., et al. (2005). Hydatid disease of the abdomen and other locations. World J Surg, 29, 1161–1165. doi: 10.1007/s00268- 005-7775-3 | [2] Yuksel, M., Demirpolat, G., Sever, A., et al. (2007). Hydatid disease involving some rare locations in the body: a pictorial essay. Korean J Radiol, 8, 531–540. doi: 10.3348/kjr.2007.8.6.531 | [3] Sable, S., Mehta, J., Yadav, S., et al. (2012). Primary omental hydatid cyst: a rare entity. Case Rep Surg, 2012, article ID: 654282. 10.1155/2012/654282 | [4] Karavias, D.D., Vagianos, C.E., Kakkos, S.K., & Panagopoulis, C.M. (1996). Peritoneal echinococcosis. World J Surg, 20, 337-340. | [5] Acharya, A.N., & Gupta, S. (2009). Peritoneal hydatidosis: A review of seven cases. Tropical Gastroenterology, 30(1), 32-34. | [6] Ionescu, A., Trufin, R., Jakab, A., & Jutis, T. (1985). Primary hydatid cyst of the great epiploon with spontaneous rupture: hydatid peritonitis. Rev Chir Oncol Radiol O R L Oftalmol Stomatol Chir, 34, 53-56. | [7] Prousalidis, J., Tzardinoglouk, K., Katsokis, C., & Aletras, C. (1998). Uncommon sites of hydatid disease. | World J Surg, 22, 17-22. doi: 10.1007/s002689900343 | | [8] Daali, M., Hssaida, R., Zoubir, M., Hada, A., & Hajji, A. (2000). Peritoneal hydatidosis: a study of 25 cases in Morocco. Sante, 10(4), 255-260. Retrieved from http://www.jle.com/fr/revues/sante_pub/san/e-docs/00/03/5B/91/article.phtml | [9] Majbar, M., Souadka, A., Farid, S. et al.(2012). Peritoneal echinococcosis: anatomoclinical features and surgical treatment. World J Surg, 36(5), 1030-1035. doi: 10.1007/s00268-012-1475-6