Primary hydatid cyst of the left- sided colon presenting with lower gastrointestinal bleeding; an extremely rare location of extrahepatic hydatid disease

Mehmet Kafadar[1]

[1]Affiliation not available

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Abstract

Hydatid cyst is an endemic disease especially in underdeveloped and developing countries affecting mostly the liver and lungs. However, a wide range of unusual anatomical sites in the abdomen has been reported, including the spleen, pancreas, kidney, and ovaries as well as dissemination within the abdominal and pelvic cavities. The location of hydatid disease in the colon is very infrequent, and very few cases have been presented. The hydatid cysts located in other sites are mostly due to rupture or extrusion of primary liver or splenic cysts. In this article, we present a case of primary left-sided colon hydatid cyst, resected laparoscopically with the affected intestinal segment. The diagnosis of hydatid cyst was made by macroscopic and microscopic examination.

Introduction

Echinococcal disease remains an endemic parasitosis threatening public health globally. The liver (66%) and lung (25%) are the most commonly affected sites, but other organs (e.g. spleen, kidney, brain, muscle, adrenal glands, bone, heart, pancreas) can also be affected. Patients who have an extrahepatic hydatid cyst present mostly with discomfort and abdominal pain. Diagnosis can be challenging [1,2]. Herein, we present an unusual case of primary intestinal hydatid disease with lower gastrointestinal bleeding. As this is an extremely rare finding, we suggest that clinicians must be very suspicious of the disease when encountering a cystic mass in abdomen, and this is particularly true for cases in endemic areas.

Case Report

A 40-year-old man was admitted to the general surgery outpatient clinic of our hospital with complaints of episodic abdominal pain, discomfort with a 15-day history, and nausea, loss of appetite, blood in the stool during the last 3 days. The patient's medical history and family history were unrevealing. Physical examination revealed left lower quadrant tenderness. Vital signs were within normal limits. Laboratory tests resulted with; white blood cell: 12500/mm^3, hemoglobin: 11.2 g/dL, C-reactive protein: 10 mg/dL, and other biochemical parameters were also normal. In oral contrast-enhanced abdominal tomography (CT) (Figure 1a) a cystic lesion with a thick wall with a 5x4 cm size adhering to the intestinal wall was observed in the descending colon and sigmoid colon junction in the left lower quadrant. Enteric duplication cyst was considered in the differential diagnosis. It was reported that the mass did not disturb the intestinal passage, a benign pathology was considered and no pathological lymph node was detected in CT. Liver and spleen parenchyma was normal in CT (Figure 1b). Also no pathological appearance was detected in the thoracic tomography (Figure 1c). No abnormality (mass) was found during the colonoscopy in the intestinal lumen. Digital rectal examination revealed hematochezia. Hydatid cyst was not included in the differential diagnosis at the beginning. With these findings the patient underwent laparoscopic segmenter colon resection (4-port)

under general anesthesia with a stapling device. Abdominal examination revealed a 5 cm size mass associated with left-sided colon wall (Figure 1d). The excised mass was taken out of the abdomen inside a specimen retrieval bag, without being ruptured (Figure 1e). The two colonic ends were anastomosed. Intraoperative frozen section examination of the mass indicated a hydatid cyst (Figure 1f,g). The postoperative period was completed uneventfully and the patient was discharged 6 days after the operation on albendazole 400 mg bid for a period of 4 weeks. No recurrence was noted during the 6-months follow-up period. Microscopic examination confirmed the diagnosis of a hydatid cyst of the colon. Lamellar formations of adjacent hydatid cyst with large areas of fibrosis are seen (Figure 2a-c). Informed consent was obtained from the patient who participated in this case.

Discussion

Hydatidosis, a parasitosis which is largely caused by Echinococcus granulosus, should be taken into account in differential diagnosis both in endemic and non-endemic areas largely because the societies have currently been globalized more than ever. Humans are intermediate hosts, dogs and ships being definitive and intermediate hosts respectively. Infection occurs through contact with a definitive host or intake of contaminated food or water. The intake of contaminated food or water is followed by the entry of hexacanth embryos into liver, the terminal station for the parasites, via portal system. Although hydatid cyst is mostly of benign character, it may also show malignant features when pulmonary or other distant organ involvement takes place. Echinococcus granulosus is common to certain geographical areas of Mediterranean countries, New Zealand, Australia, Middle East and South America, where livestock raising is widespread. Extrahepatic hydatid disease usually remains asymptomatic for years. Clinical presentation occurs only after hepatic cysts become palpable or they lead to non-specific signs and symptoms such as sense of abdominal discomfort [3].

The combination of clinical, radiological, and laboratory findings help for a preliminary diagnosis. Considering the available diagnostic imaging studies, ultrasonography (USG) offers a better visualization and assessment of hepatic cysts and hydatid disease compared to Magnetic Resonance Imaging and CT. In the literature, very high specificity (93-100%) and sensitivity (88-98%) rates are given for USG in the diagnosis of hydatid disease. Abdominal cystic lesions (i.e., mesenteric cysts, ovarian cysts, enteric duplication cysts, lymphangioma) must be considered in the differential diagnosis [4]. In other studies, CT offers the best diagnostic help in identifying the cysts and their complications; visualization of cysto-colonic fistulas, and identifying additional cysts in other organs and thus staging the disease [5].

Latatu-Córdoba MÁ et al. [6] have performed a systematic review of the literature focusing in diagnostic, epidemiological, and therapeutic tips about hydatidosis of the colon published to date. Nine case reports were found [five men, four women, median age 64.5 (21-81) years]. They have tried to find explanations about the low incidence of primary hydatidosis of the colon or affecting the colon. No clear explanation about low incidence has been found. In almost all cases several image studies were carried out, but abdominal CT was the most used (66.7%) in their study. In our routine practice, the workup of a patient with a mass located in the abdomen starts with a CT scan. Since the CT scan reported a mass originating from the left- sided colon wall, hydatid disease was not considered in the differential diagnosis, and we did not proceed with any other imaging modality in our case.

Enzyme-linked immunosorbent assay (ELISA), Indirect hemagglutination test (IHA), latex agglutination test and immunoblots are the widely available serological techniques to diagnose the hydatid disease. Inadequate sensitivity and specificity rates of such tests, however make these tests debatable on scientific level. In the studies, a wide range of sensitivity rates (50-100%) are given for the IHA test. The specificity rate is also reported to be 83% to 88. It has been reported that the combined use of ELISA and IHA tests boosts the overall sensitivity, reaching up to 94.7% [7]. Since the imaging study was also not supporting a diagnosis of hydatid disease, no further serological test was done for our patient.

Complete surgical excision of all cysts is the treatment of choice to eradicate the disease from the body, but depending on the location of hydatid cysts jeopardizing neighboring organs or tissues, partial or subtotal cystectomy can also be an option. An open approach by laparotomy is the usual management of this infrequent

clinical situation; in any case, the surgeon chooses the surgical technique in the light of the intraoperative findings [8]. In our case we laparoscopically resected the affected colon segment and took the specimen out in a specimen retrieval bag. Mebendazole or albendazole is given to the patient adjuvantly to prevent recurrence. In our case, we preferred albendazole treatment, and there was no recurrence in the 6-month follow-up period.

PAIR (puncture, aspiration, injection and reaspiration) and medical therapy also can be used for the treatment. Soft tissue hydatid disease is still treated best by surgical excision. Surgery is preferred mainly because it can prevent certain complications such as compression of neighboring tissues, infection, or cyst rupture. As soft tissue cysts are prone to rupture and recur, maximum care should be taken to prevent cyst rupture [9].

Based on our experience from our clinical practice, we administer and advocate administering prophylactic albendazole, at a dose of 400 mg bid for a minimum period of two weeks preoperatively and four weeks postoperatively, for any cyst of any location scheduled for surgical removal. When a patient has a ruptured cyst or multiple cysts at different locations, postoperative albendazole treatment should be extended to at least two months [10].

Conclusion

Hydatid disease is a significant public health problem in underdeveloped and developing countries affecting mostly the liver and lungs. Primary large intestine hydatid disease is a very rare clinical entity. It was highlighted by this case report that the differential diagnosis of masses in abdomen should include hydatid cyst, particularly in areas where the disease is endemic like in Sanlıurfa- Turkey. There is a paucity of clinical cases in the literature largely hindering a thorough and evidence-based description of the condition. More robust implications can be reached by further studies.

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Figure Legends:

Figure 1. CT image of the patient (a), normal liver and spleen on CT scan (b), normal lung parenchyma on CT scan (c), intraoperative appearance of the cyst (d), excised cyst (e-g), resected colon segment (h).

Figure 2. Microscopic view of sections of surgical specimen; lamellar formations of adjacent hydatid cyst with large areas of fibrosis (a-c). (H&E: ×40) (a), (H&E: ×100) (b,c)