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CASE REPORT

Rare isolated primary peritoneal hydatid cysts: A case report from Syria

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ABSTRACT

Hydatid disease caused by *Echinococcus granulosus* is endemic in many regions of the world. The major primary site for the disease in adults is the liver and the secondary site are the lungs. Secondary peritoneal cysts are relatively common and expected to occur after rupture of the primary hepatic hydatid cyst. Primary peritoneal hydatid cyst disease without any other organ involvement has been previously reported, and yet it is still considered rare even in endemic areas. A case of a large primary peritoneal hydatid multicystic lesion without other organ involvement in a 25-year-old girl seen at the gastrointestinal outpatient clinic in the University of Kalamoon Medical City is presented and discussed. The disease was very extensive but surgical intervention was refused due to the patient being a young unmarried female. The patient was treated and observed over a period of ten months.
She responded very well to medical treatment with albendazole. The case emphasizes the importance of hydatid disease being included in the differential diagnosis of any cyst in the abdominal cavity for patients living or coming from an area of endemic hydatid disease even without liver or lung involvement. This also goes to show that it can respond to medical treatment, which becomes even more valuable in conditions where surgical intervention might not be an option.

Keywords: primary hydatid cyst, peritoneal, *Echinococcus granulosus*

INTRODUCTION

Human hydatid cyst is a parasitic disease caused by the larvae of *Echinococcus granulosus* and was described and detailed as a human disease by the

contributions of many scientists such as Redi, Palas, Rudolfi and Premsek.[1] The disease is endemic in certain parts of the world including the Middle East.[2,3] Unfortunately, no clear epidemiologic data are available about disease prevalence in Syria. Due to the Syrian crisis however, the number of cases reported of this disease are increasing thereby constituting an escalating public health concern. Hydatid disease is now considered a re-emerging disease in Syria.

The major primary site for the occurrence of the disease in adults is the liver (75%), followed by the lungs (5 – 15%), and all other organs add up to 20% of cases.[4] These percentages are different for children as the lungs form the primary site (64%) followed by the liver (28%).[5]

Secondary peritoneal cysts are relatively common and expected to occur after rupture of the primary hepatic hydatid cyst.[4] Primary peritoneal hydatid cyst disease without any other organ involvement has been previously reported, and yet is still considered rare even in endemic areas.[5,6]

In this article, a case study of primary peritoneal hydatid cysts filling the cavity of the abdomen and pelvis without any other organ involvement is presented and discussed.

CASE REPORT

Case presentation

A 25-year-old woman presented to the gastrointestinal outpatient clinic in the University of Kalamoon Medical City (UOK-Medical City) with abdominal pain, diarrhea and distention of the abdomen. Abdominal pain had started three weeks earlier and it was colic and extended over the whole abdomen, increased after eating and decreased after defecation, induced nausea without vomiting, and did not awaken the patient at night. She gave no history of fever, chills, rash or itchy skin, no joint pain or stiffness or joint swelling, no loss of appetite or noticed weight loss. The patient had not undergone any previous surgeries and had no allergies or major medical complaints. She did not smoke or drink alcohol, and lived in a rural area with no direct contact with animals. At the time of examination, she was experiencing 4 – 5 episodes of loose stools daily without tenesmus or blood stains. She noticed a gradual extension of her abdomen over the month prior to her visit to the clinic. The patient was first treated with metronidazole that improved diarrhea initially; however, the symptoms relapsed after two days.

Clinical examination

The patient looked well; her vital signs were normal and she did not seem to be experiencing any pain. Examination of the head, neck, chest, heart and extremities were normal. Inspection of the abdomen showed no surgical scars or striae. The abdomen was slightly distended bilaterally and was moving with breathing. On auscultation, the bowel movements were found to be active. The abdomen was found to be soft on superficial palpation, while deep palpation caused generalized light tenderness. No masses or abdominal lesions were felt. Liver span on the midclavicular line was 12 cm.

Investigations

Ultrasonography (US) of the abdomen and pelvis, the liver, gall bladder, spleen, pancreas, kidneys, uterus and bladder were all within normal limits. The US showed a clear multicystic space-occupying lesion, which occupied the whole abdomen from the diaphragm to the pelvis with a large amount of free fluid (ascites) in both the abdomen and pelvis. Some of the cysts measured up to 40 × 35 mm (US images are unavailable).

The blood investigations including leucocytes, erythrocytes, platelets, hemoglobin and hematocrit levels were all normal. Blood tests showed elevated CRP levels (46 mg/l) (the normal range is up to 5.0 mg/l) with high ESR levels (30 mm/h) (the normal range is up to 20 mm/h). More importantly, the serum was strongly positive for *Echinococcus granulosus*'s antibodies (1/6400 with normal titer values up to 1/160). Stool investigations were found to be normal except for a few *Entamoeba histolytica* cysts.

CT scan of the abdomen and pelvis (Figure 1) before treatment shows a large number of multi-sized cysts filling the cavity of the peritoneum in the pelvis in front and behind the uterus reaching the left and right hypochondriac regions with free fluid especially in the area surrounding the liver. The liver, spleen, pancreas, kidneys and uterus all appeared to be of normal size, with no cystic or solid lesions in the liver or spleen. Chest X-ray was within normal limits without cystic lesions in the lungs. The free fluid was aspirated under the direction of the US; the aspirated ascites was observed to be yellow turbid fluid and the

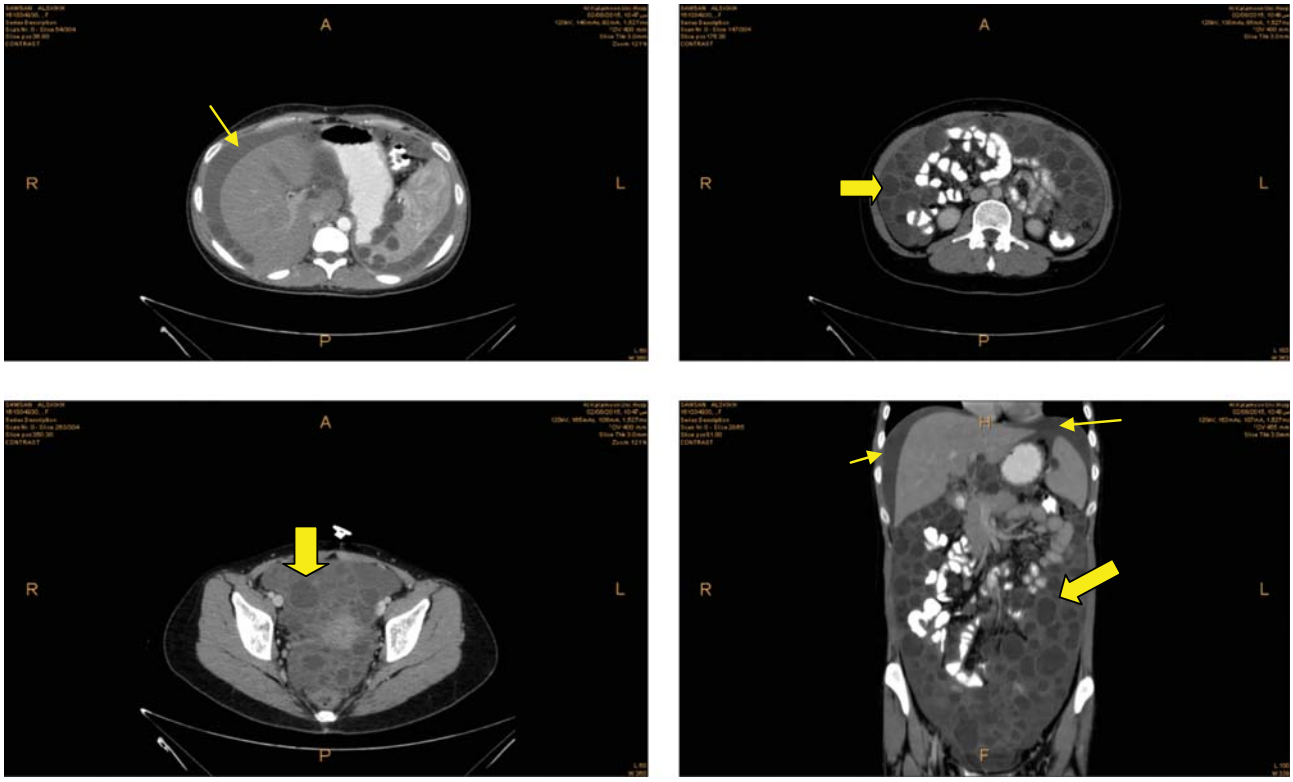

Figure 1. CT scan of the patient before treatment. The imaging shows a huge number of cysts occupying the entire peritoneal cavity with free fluid around the liver and spleen. The narrow arrows indicate free fluid, while the wide arrows indicate abdominal cysts.

following levels were detected: RBC 1280 cells/μl, WBC 10,500 cells/μl (eosinophils – 91%, neutrophils – 4%, and lymphocytes – 5%), glucose 68 mg/dl, protein 5.8 mg/dl, LDH 166 IU and albumin 2.60 mg/dl. The type of ascites was found to be inflammatory with a low SAAG (serum-ascites albumin gradient) value of 1 g/dl, and high leucocytes count, predominantly eosinophils indicating a reaction due to a parasitic cause.

Treatment

The patient was given ciprofloxacin 500 mg bid and anti-spasmodics for one week for treating infectious diarrhea, in addition to albendazole 400 mg twice daily for six months. The abdominal pain and diarrhea were improved within a few days. The patient was closely monitored in the weeks following the treatment. The abdominal cysts were found to be gradually decreasing (according to US imaging and not CT scan). The patient was feeling very well without any abdominal complaint and clinical examination was within normal limits. Contrast-enhanced CT images after six months of treatment showed a major decrease in the number of cysts, with only two cysts remaining in front of the psoas muscle, which measured 28 and 20 mm in diameter (Figure 2); the remaining abdominal and pelvic cavity was totally free from cysts with complete absence of the free fluid.

DISCUSSION

Hydatid disease of the peritoneum is usually secondary to hepatic or splenic involvement in the disease.[6,7] Primary peritoneal infection with *E. granulosus* accounts for around 2% of reported cases.[6,8] The low prevalence of primary peritoneal infection may be assumed to be due to the physical barriers to the hematogenous diffusion of cysts created by the liver and lungs, which are the major primary sites for the disease in adults.[5] Various pathways have been suggested in the pathogenesis of peritoneal localization of the cysts. Up to 15% of parasites escape from being filtered in the liver and lungs and enter the systemic circulation to implant in various sites.[5,9]

The clinical course of peritoneal localization usually can be non-specific and depends on 1) the site of

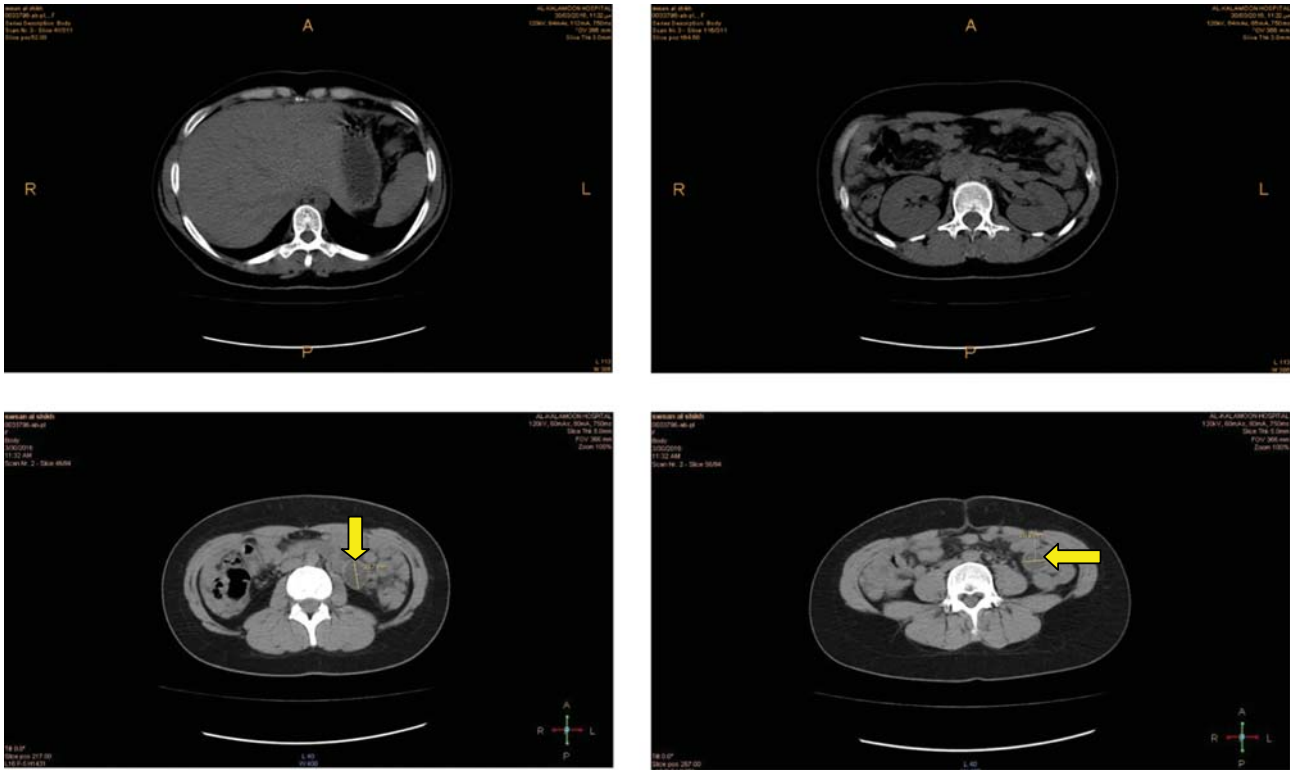

Figure 2. CT scan of the abdomen and pelvis after treatment. Images indicate very good improvement with no ascites and only two small cysts present in the abdomen. The yellow arrows indicate pelvic cysts.

involvement, 2) cyst size and 3) the effect of the enlarged cysts on adjacent organs. Therefore, it is not unusual to come across the presence of an inert mass without any symptoms or deterioration in the patient's condition.[10] Similarly following this scenario, a painless multicystic mass was detected in our patient with no major complaint other than abdominal pain that increased on eating and decreased with defecation, which brought her to the clinic. As in other reported cases, imaging evaluation including US and CT were very useful in diagnosis and treatment follow-up.

Some reports have documented that serology may not always be helpful in diagnosing primary and nontraditional (other than the liver and lungs) hydatidosis[5]; however, this was not the case in our report. Serology assisted in the differential diagnosis of the lesion and helped avoid biopsy or partial excision, which may have caused harmful effects, such as an anaphylactic reaction. In the literature, medical treatment varies from percutaneous aspiration, injection, and reaspiration of certain types of hydatid cysts to surgery, which remains the treatment of choice.[2] In similar published cases,[5,6] the chosen therapy was laparotomy and cyst deroofing with pre- and postoperative albendazol treatment. In this report, the disease was very extensive and with the patient being a young unmarried female, surgical intervention was refused; therefore, medical treatment was adopted. The patient responded very well and after six months of albendazole (400 mg twice daily), only two cysts were left that measured less than 30 mm in diameter. At this current time, the remaining abdominal and pelvic cavity is totally free from cysts with complete absence of the free fluid.

CONCLUSION

The number of reports of primary peritoneal hydatid cyst disease without any other organ involvement are still very few. To the best of our knowledge, this is the first time such a case is reported in Syria.
The presented case emphasizes that hydatid disease should be considered and included in the differential diagnosis of any cystic mass for patients living or originating from an area of endemic hydatid disease, regardless of the presence of primary cystic lesions in the common sites. Furthermore, what makes this case

worth reporting is the successfulness of the unconventional management. This good response to medical treatment becomes even more valuable in rural or unstable areas where surgical intervention might not be an option.

CONFLICTS OF INTEREST

The authors declare that there are no conflicts of interest regarding the publication of this paper.

CONSENT

Informed consent was obtained from the patient and the research was conducted according to appropriate institutional ethics followed at the UOK-Medical City.

AUTHORS' CONTRIBUTIONS

BA conducted the clinical part of the research according to the institutional ethics followed at the UOK-Medical City. LA contributed to the conception and design of the study and to the acquisition, analysis and interpretation of data. LA also drafted the manuscript and both LA and BA revised it critically for content. All authors read and approved the final manuscript.

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