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CASE REPORT

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Primary pelvic hydatid cyst in an infertile female, A case report

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Abstract

It is important to consider hydatidosis as a differential diagnosis in all suspected cysts in endemic regions. The wide range of hydatidosis presentations makes it difficult to differentiate from similar symptoms.

KEYWORDS

case report, *Echinococcus*, hydatic cyst, Iran, pelvic

1 | INTRODUCTION

A young woman was diagnosed with a multilocular primary pelvic hydatid cyst. Laparoscopic surgery confirmed the initial diagnosis of echinococcusis in pelvic. It is important to consider hydatidosis (echinococcusis) as a differential diagnosis in all suspected cysts, especially in endemic areas.

Echinococcosis refers to anthropozoonotic disease. The disease is endemic in Middle East, Eastern Europe, and South America.[1] Humans are the intermediate host and get infected through accidental consumption of parasites' egg with food or water.[2] The parasites can grow in two forms: unilocular cyst or cystic echinococcosis (CE) and multilocular cyst or alveolar echinococcosis (AE).[3] CE is the most common form of disease among nomadic area in which dogs play an important role in habitation. *Echinococcus granulosus* (*E granulosus*) can grow in every organ or body tissue and produce small cysts. Liver is the most infected organ (59%-75%), followed by lungs (27%), kidney (3%), musculoskeletal system (1%-4%), and pelvic (0.2%-2.25%) respectively.[4] In this study, we present an unusual case of primary localization of hydatid cyst in pelvis cavity.

2 | CASE REPORT

A 20-year-old infertile female married at 14 and had not been pregnant ever, referred to our hospital with the chief complaint of abdominal pain for several days. Clinical examination revealed low blood pressure (95/60 mm/Hg), pale mucosa, and general tenderness in abdomen with focus in lower quadrants of abdomen. Patient declared that had no past medical history but a 40-day-old abortion. The patient declared some cases of infection with *Mycobacterium tuberculosis* (TB) in first and second-degree relatives.

3 | DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS, AND TREATMENT

All laboratory studies such as complete cell blood count (CBC), blood urea, and creatinine were performed and results were normal. Also, BHCG test was negative. Ultrasonography and magnetic resonance imaging (MRI) of abdomen and pelvis performed. Ultrasonography findings of kidneys, bladder, and urinary tracts were

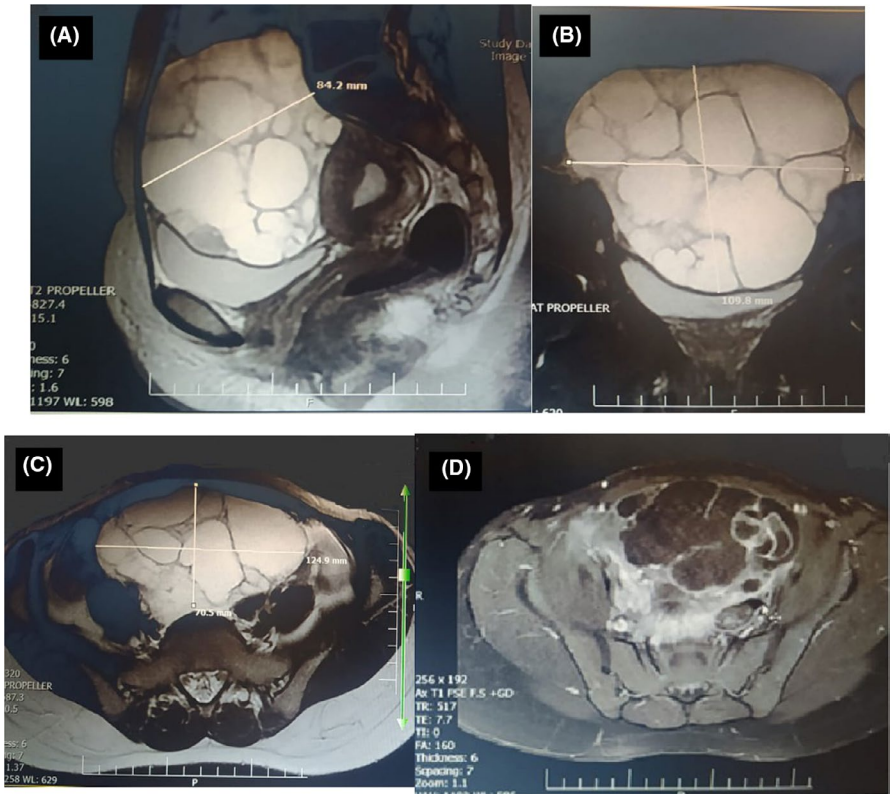

FIGURE 1 A, sagittal, B, Coronal and C, D, axial images obtained at T1.T2 fs and contrast-enhanced sequences large multilocular cystic structure without mural nodule and with enhancing septa about 70 × 84 × 124 mm is noted at Rt side of pelvic cavity

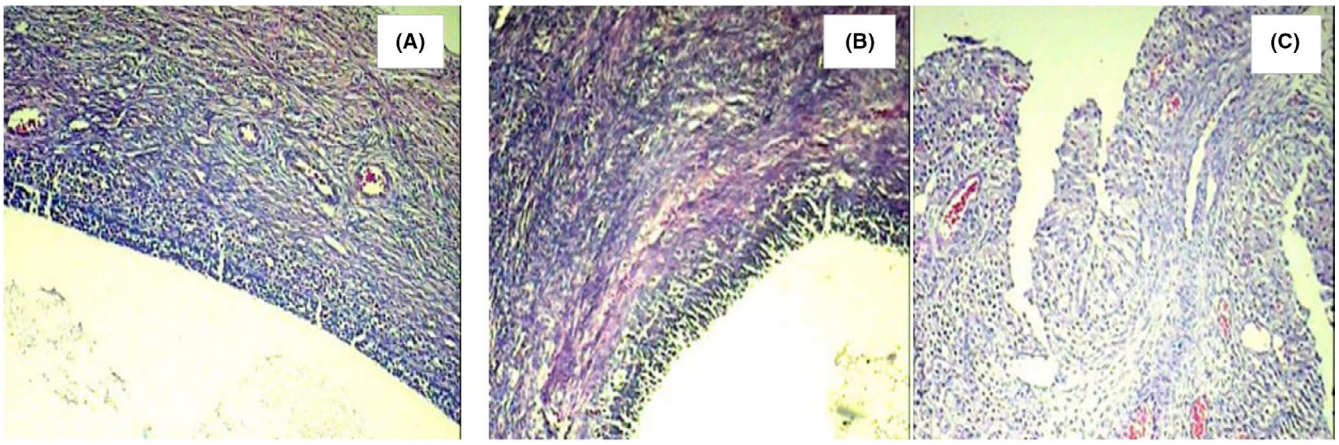

FIGURE 2 Left ovary (H&E staining). Containing numerous unilocular cysts, corpus luteum, and corpus albicans. A, B, Unilocular follicular cysts with inner layer of granulosa cells surrounded by theca interna in outer layer. Hemorrhage is visible in stroma of ovary. C, Unilocular follicular cyst. Note the luteinized cells forming the inner cyst layer and hemorrhage in stroma

normal. Ultrasonography suggested a large multilocular cyst (74 × 90 mm) attached to the right ovary (Figures 1 and 2). A large amount of free fluid around the cyst suggesting the cyst rupture observed. In addition, MRI of the abdomen and pelvis showed a normal liver, bile duct, pancreas, kidney, bladder, and uterus and in contrast-enhanced sequences a large multilocular cystic structure with enhancing septa about 70 × 84 × 124 mm was noted. Few amount (3 mL) of fluid was aspirated from pelvis and sent for pathologic study. The results of pathologic investigation demonstrated that the fluid contained mesothelial and inflammatory cells. Considering the history, physical examination, paraclinic and laboratory findings, and differential diagnosis of cystadenoma, multilocular cyst and hydatid cyst were considered.

Preoperative therapy with albendazole started for patient with dose (10-15 mg/kg/d) for 28 days. In laparotomy surgery, liver, spleen, mesentery, and omentum were normal but, a large tense hydatid cyst was noted in the pelvic cavity and multiple small daughter cysts adhered to pelvis organs were observed. Also, bloody fluid in abdomen and pelvis which were resulted from cyst rupture was noted. In the initial view, it was similar to abdominal tuberculosis which was in accordance with tuberculosis history in patient′s family. Cyst was completely excised after mobilization without

rupture and by packing the surrounding area with 1% cetrimide-soaked sponges then, abdominal and pelvis cavity washed to reveal adhesions. All daughter cysts and laminated membrane removed completely and a drain was placed in pelvis and abdomen sutured in layers. Final diagnosis was confirmed by pathological examination. Due to the familiar history of *Mycobacterium tuberculosis*, abdominal tuberculosis justified, nevertheless, pathology approved hydatid cyst in abdominal and pelvis cavity and follicular cyst in ovary (Figures 2-4). Patient was put on albendazole therapy for 3 cycles and the dose of the albendazole was adjusted according to the body weight of the patient. The duration of each cycle of albendazole therapy was 28 days and patient was advised to stop therapy for 2 weeks for assessing the liver function and complete blood counts. As, these tests were normal subsequently 2nd and 3rd cycles of therapy were completed. Patient followed up for 6 months and she had no symptom after this time.

4 | DISCUSSION

Echicoccosis known as an important socioeconomic concern in countries like Iran.[5] The hydatid cyst is larval stage of genus *Echinococcus granulosus*. Adult worms live in intestinal lumen *of* definitive host of carnivores such as dog and the eggs are passed with feces of infected dogs. Herbivores as intermediate hosts get infected through digestion of egg by consumption of contaminated food or water. Consequently, oncospheres hatched out from the eggs and released into the blood stream. Human incidental intermediate host.[6] Liver is the most common destination of oncospheres via the portal vein, but in some occasions they can pass through the liver barrier and reach to other organs where they can convert to the hydatid cysts.[7] Kiresi et al,[8] claimed that hydatid cysts formation rarely occur in peritoneal cavity, pelvic sites, brain, cavernous sinus and bones. Pelvic hydatidosis could be primary or frequently secondary to the liver cysts or rarely in spleen. Sudden trauma/pressure can rupture cysts and Hydatid fluid can release and cause symptoms such as fever or syncope due to anaphylactic reactions.[9] Routine laboratory tests are usually normal except in cases of renal failure. Several serological tests are available but they mostly are used in epidemiological data, clinical features, and especially imaging investigations.

Imaging techniques and serological surveys are the main diagnostic tools for cystic lesion. Ultrasound is the preferred first line imaging, but CT scan and MRI give more information about morphology (location, size, neighborhood, and number) of the cysts. Imaging techniques are still more sensitive than serologic tests moreover negative serology results have been reported in affected cases.[10,11] Also, histopathologic examination on specimens after surgery revealed ruptured hydatid cysts in abdomen and pelvis and demonstrated the MRI results as well. As shown in Figure 4, the chitinous outer layer of hydatid cyst surrounded by fibro inflammatory cells demonstrated that why patient complained of abdominal pain (Figures 2-4).

Pelvic hydatid cysts generally have nonspecific presentations like mass making pressure on close organs (urinary bladder or rectum) and may obstruct urinary canals and leads to renal failure. In our case, the presence of cyst may be the main reason of patient's infertility for seven years. There are evidences of infertility in patients with pelvic hydatidosis.[12] Sometimes, the cysts rupture spontaneously and releasing the hydatid fluid containing proto scolex would produce later cysts.

Surgery is the most acceptable treatment of the pelvic hydatid disease, although albendazole therapy seems to be effective in some cases but, generally it does not use in all cases except in small cysts or in patients who are not suitable to undergo surgery.

Preoperative albendazole therapy reduces the intracystic pressure, while postoperative albendazole therapy decreases the risk of recurrences of hydatidosis. Combination of pre and postoperative albendazole therapy with surgery is the best choice. In this case, we used this procedure. Postoperative

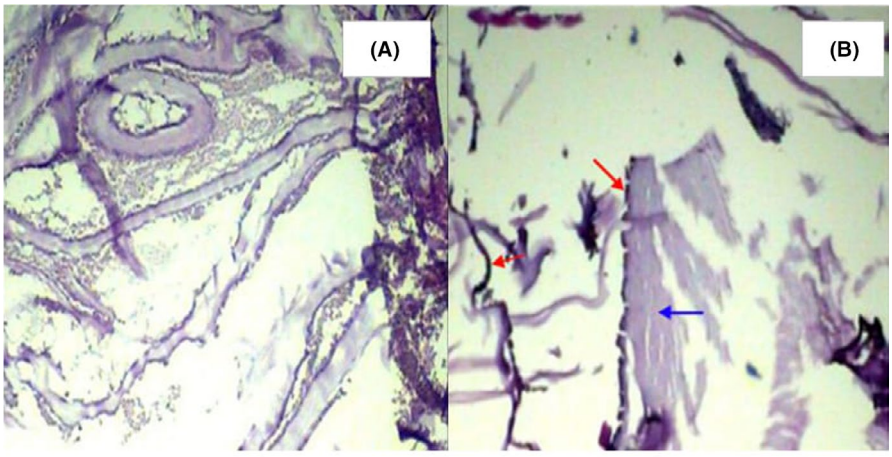

FIGURE 3 Ruptured hydatid cysts in abdomen and pelvis. (H&E staining): A, Chitinous outer layer is an acellular laminated and thin layer surrounding internal germinal (nucleated) layer. B, Ruptured chitinous layer (blue arrow) and thin germinal inner layer (red arrow)

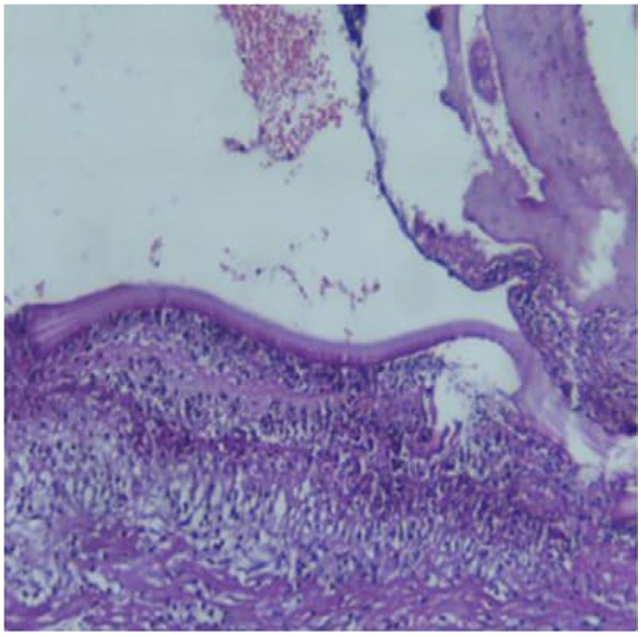

FIGURE 4 Retractile chitinous outer layer of hydatid cyst surrounded by fibro inflammatory necrotizing reaction in ovary. (H&E staining)

albendazole can inhibit furthermore growth of seeded protoscolexes which can release during the operation. In cases that surgery will cause more danger than benefit, it is recommended to do partial cystectomy and drug therapy. Also, imaging is the adequate method to monitor positive responses to medication.[13-16]

5 | CONCLUSIONS

It is important to consider hydatidosis as a differential diagnosis in all suspected cysts in endemic regions. The wide range of hydatidosis presentations makes it difficult to differentiate from similar symptoms. Using imaging techniques and ultrasonography will help to diagnose the causative agent. The adhesive nature of these cysts may cause serious complications like infertility and should be considered in infertile cases.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

All authors, MG, EK, and AS: equally contributed to the design, analysis, and presentation of this study. MG: specialist in infectious disease and involved in study design. EK: involved in study design and writing. AS: Involved in study design, writing, submission, and revision.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Applicable.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

DATA AVAILABILITY STATEMENT

All the data are available without restriction.

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