

Percutaneous Treatment of Primary Retroperitoneal Hydatid Disease

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Abstract

Primary retroperitoneal hydatid disease (HD) is extremely rare. The conventional treatment of primary retroperitoneal HD is surgery. There is no report in literature for percutaneous treatment of primary retroperitoneal HD. We report on a patient with primary retroperitoneal HD, who was treated successfully with percutaneous treatment using the PAIR (percutaneous puncture-aspiration-injection-reaspiration) method with drainage technique. After the procedure, the patient was asymptomatic at 5 months of follow-up. In conclusion, percutaneous treatment can be used as an alternative to surgery for the treatment of primary retroperitoneal HD. However, further studies are needed.

Keywords; Retroperitoneal, hydatid disease, percutaneous treatment

Introduction

Hydatid disease (HD) is an endemic disease caused by larval forms of the tapeworm *Echinococcus granulosus*. HD may develop in any organ, most frequently in the liver (60-70%) and the lungs (20–30%) (1). In 10% of the cases, HD unusually develops in viscera such as spleen, pancreas, kidney, muscles, heart, bones, and brain. Other rare locations have been reported in less than 1% of all cases of hydatid cysts (1, 2). Primary retroperitoneal HD is extremely rare.

The conventional treatment of primary retroperitoneal HD is surgery and total excision (55.8%), partial cystectomy (39.5%) and unroofing (4.6%) are the surgical approaches used (3). However, surgery is associated with seeding, significant morbidity and prolonged hospital stay (4). Percutaneous treatment (PT) of abdominal HD has been used as a minimally invasive, safe and effective treatment in the past 20 years (5). It is a potential alternative to surgery. PT is performed either by puncture, aspiration of cyst content, injection of hypertonic saline solution, and re-aspiration of all fluid (PAIR) or evacuation of the entire endocyst (modified catheterization technique) (5, 6). PT of HD in retroperitoneal organs such as pancreas and kidney has been reported successful, but there is no report in literature of PT of primary retroperitoneal HD (7, 8). We here report a patient with primary retroperitoneal HD, who was treated successfully with PT.

Case Report

A 62-year-old male presented with backache for the previous 6 months. He had no other complaints. Physical examination showed no abnormality. Laboratory results showed an erythrocyte sedimentation rate of 9 mm/h (Westergren) and C-reactive protein, 0.4 mg/dL (normal range 0-5 mg/dL). Biochemical and hematological tests, which include renal and liver function tests, hemoglobin level, and total leukocyte count, were found to be within the normal ranges. No liver or lung manifestations were present. Anti-echinococcal IgG was positive by enzyme-linked immunosorbent assay (titer 6.9; positive if > 1.1). Direct abdominal plain radiography did not show any abnormality. Abdominal computed tomography (CT) revealed a well-defined low attenuation cystic lesion with calcified cyst wall in some places, measuring 9 and 16 cm in diameter, filling the left iliac fossa (Figure-1). Abdominal magnetic resonance imaging (MRI) documented hyper intense mass within the hypointense septations lesion in the retroperitoneum (Figure-2). Chest CT examination was normal. He was treated with albendazole for 3 weeks before PT. All measures were taken for anaphylaxis and medical personnel were kept present at the intensive care unit throughout the interventional procedure. The procedure was performed following an 8 hour-fasting, under local anesthesia applied to the point of puncture and aseptic conditions. The cyst was punctured with a 22 cm 18 gauge needle via translomber route with the patient in the prone position; all cyst content was rapidly aspirated and hypertonic saline (20%) was injected into the cavity under ultrasonography (USG) guidance. Following re-aspiration of the cyst cavity. There were no complications during and after the procedure. Direct microscopic examination of the aspirated fluid showed fragments of the laminated membrane and cytological

examination was consistent with a hydatid cyst. The catheter in the cyst cavity was withdrawn after 24 hours. The patient was discharged from the hospital on the third day after the procedure. Albendazole was continued for 3 months. The patient was asymptomatic at the 5-month follow-up. Pseudo-solid appearance of cysts was observed in abdominal USG on the 5th month, which measured 6x9 cm in diameter (Figure-3).



Figure-1: Coronal CT showing low attenuation cystic lesion with calcified cyst wall in some places, filling the left iliac fossa (measured 16.5x8.5 cm diameter).

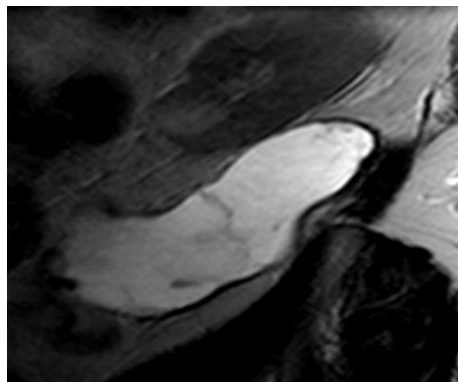


Figure-2: Saggital MRI section of the lesion in the retroperitoneum showing hyperintense mass within the hypointense septations.

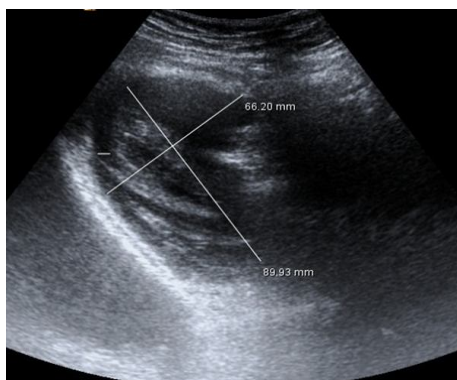


Figure-3: Five-month USG image showing pseudo-solid appearance of the cyst, tension loss in the cyst and reduction in the size (measured 9x6.5 cm diameter).

Discussion

Primary retroperitoneal HD is defined as cysts presented in retroperitoneum independent from any peritoneal viscera, and without concurrent or prior hydatid disease of other organs (9). Even in endemic areas, primary retroperitoneal location has been reported as very rare, because most of the parasites are trapped by hepatic and pulmonary filters (10, 11). Once the oncospheres reach systemic circulation by passing through the liver and the lungs, they develop an implant in the retroperitoneum. Another theory is that the oncospheres disseminate through the intestinal lymph vessel and the thoracic duct (12). The usual presentation of retroperitoneal hydatid cysts is a palpable mass (65.1%) or abdominal or back pain (72%), as in the case reported (3).

Several diseases may present as a retroperitoneal cystic mass such as cystic lymphangioma, retroperitoneal abscess, pseudocyst, ovarian neoplasm, cystic or necrotic solid tumors and embryonal cyst (13). USG, CT, and MRI imaging are frequently used for diagnosis in clinical practice and help in differential diagnosis. The presence of daughter cysts within a large cyst

and increased density of the hydatid membrane are diagnostic signs, but findings at imaging studies are usually suggestive of HD. Percutaneous aspiration can also be used to diagnose doubtful cases of retroperitoneal HD (14). Serological tests including immunoelectrophoresis, indirect hemagglutination test, complement fixation test, and ELISA help for diagnosing HD but have low sensitivity and specificity (5). The preliminary diagnosis of primary retroperitoneal HD based on positive radiological and serological findings in our case was confirmed with percutaneous aspiration findings.

The conventional treatment for retroperitoneal HD is surgery. Khan M. reported two cases treated with laparoscopic surgery that offers minimal invasive surgery (15). However, as mentioned before, surgery is associated with significant morbidity and prolonged hospital stay (4). In some patients, such as elderly and those with high surgical risk, surgical treatment is not preferred. Retroperitoneal hydatid seeding is an important problem in surgical treatment. In two studies, carried out in large numbers of patients, retroperitoneal hydatid seeding has been reported at the rates of 5.3% and 6% (16, 17). Utilization of fine needles and catheters in PT, advances in USG and CT techniques, and medical chemoprophylaxis using praziquantel or albendazole before and after PT reduce hydatid seeding. In three recent studies, no hydatid seeding has been reported (17-20). In our case, we started albendazole treatment 3 weeks before the intervention and continued for 3 months in order to prevent hydatid seeding.

PT is a good alternative to surgery. The catheter technique is described for the treatment of cyst hydatid in the liver. Karaman B and et al. (8) reported a patient with primary pancreatic hydatid cyst treated with PT using the catheter technique. Rapid volume reduction leaves less amount of volume in the cyst and provides good drainage of large particulate structures such as germinative membrane layers, which are the advantages of the technique of catheterization

compared to the simple PAIR technique. The catheter is to remain in the cyst for at least 24 hours to provide complete drainage of the cyst content. We withdrew the catheter from the cyst cavity after 24 hours as well. Complications can occur during and after PT. Minor complications such as fever, pain, urticaria were reported in 10-30% of the cases (17-20). Major complications such as anaphylaxis, infection of the cyst cavity occur extremely rarely. Our case had no complication during or after the procedure.

In conclusion, PT can be used as an alternative to surgery for primary retroperitoneal HD treatment. Further case reports and studies are needed to confirm the safety and efficacy of PT in primary retroperitoneal HD management.

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