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Case Report

An Atypical Presentation of Hydatid Disease, Accompanied by a Pancreatic Pseudocyst: A Case Report

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Abstract

Hydatid disease (HD) is a zoonotic disease, which typically affects the liver and the lungs. Primary retroperitoneal localization of the disease is infrequent. The most common symptom is abdominal pain; however, the clinical features of HD may be generally dependent on the location of the cyst.

A 53-year-old female patient presented to Kocaeli University General Surgery Clinic, Turkey in September 2019 with complaints of flank pain, constipation and vomiting. The MRI revealed cystic lesions in the pancreas and psoas muscle. Both cysts were excised totally during laparotomy. Histopathology reports showed that the lesion in psoas muscle was hydatid cyst whereas the lesion in pancreas was pancreatic pseudocyst. Postoperative period was uneventful and the patient was discharged with albendazole treatment. No recurrence occurred during follow-up period. Atypical localization for hydatid disease is a diagnostic dilemma for surgeons, especially in endemic countries. Hydatid disease should always be considered in the differential diagnosis of retroperitoneal lesions, inadequate management can lead to complications and fatalities. Surgery combined with antiparasitic therapy, may be an option in the presence of obstruction of gastrointestinal and/or urinary tract.

Introduction

Hydatid disease (HD) is a zoonotic disease that is known for more than 2000 years. It is mainly caused by the larval form of the genus *Echinococcus*, a cestode living in the intestines of animals, especially

canines. In most cases, the liver and/or the lungs are involved (1).

The occurrence of HD in retroperitoneal space without concomitant lesions in other organs is very rare (2). Symptoms of the unusual localization depends on the affected organ, size, adjacent structures, secondary infections and presence of rupture. On some occasions, this benign disease can result in unexpected fatalities, due to major complications (3). Urinary and gastrointestinal tract obstructions may be concerning especially in the retroperitoneal localization of hydatid disease. Duodenum, pancreas, kidneys and ureters may be involved, which can complicate the clinical management (4, 5).

Here, in this case report, we aimed to present an unusual localization of hydatid disease and its manifestations in a female patient. Primary hydatid disease should be kept in mind in the differential diagnosis of retroperitoneal lesions, especially in endemic countries. Surgical treatment of the lesions may be beneficial in selected cases.

CASE

A 53-year-old female patient, presented to Kocaeli University General Surgery Clinic, Turkey in September 2019 with a persistent left flank pain.

A detailed history revealed that she had been having abdominal discomfort, loss of appetite, intermittent fever with shivering, nausea and vomiting symptoms for 6 months. Physical examination revealed tenderness in the left lower quadrant. Routine laboratory results were within normal limits. Chest and abdominal radiography were nonspecific. Abdominal USG revealed a bilobular cystic mass of about 55x35 mm in the anteromedial area of the left kidney. Abdominal MRI scan showed two cystic masses, first measured 5.5 cm adjacent to the psoas muscle in the lower zone of the left kidney and second measured 3 cm in the corpus of pancreas (Fig. 1).

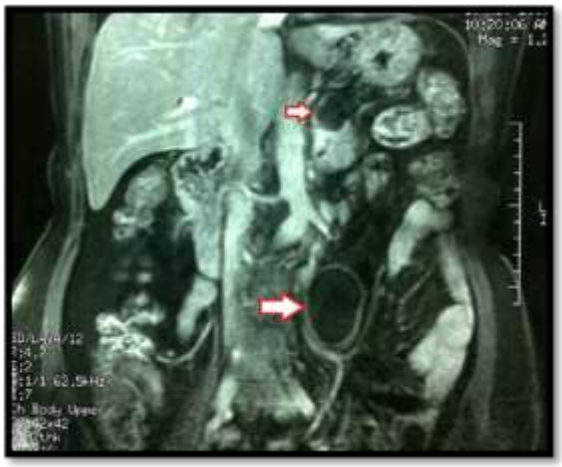

Fig. 1: Abdominal MR scan revealed a large retroperitoneal cystic mass on the left side localized between psoas and kidney and a cystic lesion on pancreas

The patient was discussed in a multidisciplinary team meeting consisting of general surgery, pathology, radiology and oncology teams. Excision of the lesions with intraoperative frozen section procedure were decided after discussing the case. Exploratory laparotomy revealed a 4 cm cystic lesion in the corpus of pancreas and a 8 cm cystic lesion in the left psoas muscle which is adjacent to the left ureter and kidney (Fig. 2). Masses and the cyst walls were dissected from the surrounding structures and totally excised carefully, preventing spillage and organ injury (Fig. 3).

Fig. 2: A large cystic mass that is attached to the ureter and psoas muscle

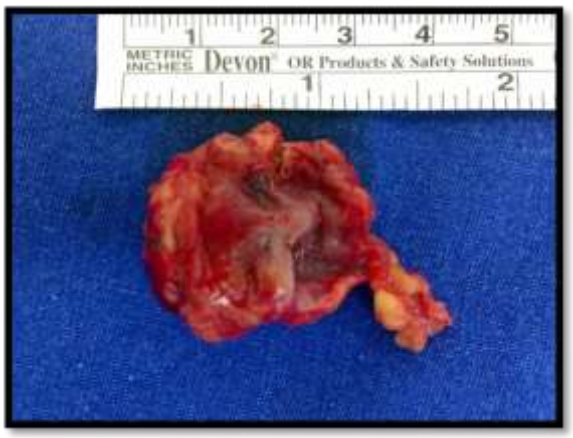

Fig. 3: Gross specimen showing cyst along with the capsule, excised from the pancreas

A frozen section was performed and according to evaluation, the cystic lesion in the pancreas was pancreatic pseudocyst and the other cystic lesion from psoas was hydatid cyst. Postoperative final pathology analysis supported the frozen section results. There were no postoperative complications after surgery. 800 mg/day albendazole treatment was started on postoperative day 1. The drains were removed on the postoperative day 3. The patient was discharged on the postoperative day 4. Albendazole was continued for 6 months. Clinical improvement was seen in the postoperative 6th month and 1st year follow-up visits. There were no radiological and clinical recurrence, therefore it was decided to follow up the patient with USG every year, without giving any treatment.

Discussion

Primary retroperitoneal hydatid cyst with no hepatic or pulmonary involvement is rare (6). More than 80-90% of hydatid cysts localize in the liver, lungs, or both. In a retrospective study, unusual localization of hydatid cysts were identified in just 8 of the 79 cases (7). The presence of hydatic cysts has been reported in the spleen, kidney, peritoneal cavity, skin and muscles, brain, vertebral column, ovaries, pancreas, gallbladder, thyroid gland, breast and bones (7, 8). Lockhart and Sapinja (9) were the first to report the presence of primary retroperitoneal hydatid cyst in 1958. Exact pathogenesis resulting in retroperitoneal localization remains unknown but it could possibly happen as a result of protoscoleces transition from the gastrointestinal tract into the retroperitoneum through the hematogenous/lymphatic drainage routes (10).

Clinical history, radiology and serological test results may help with diagnosis (1). Although most cases with primary retroperitoneal HD are asymptomatic, abdominal mass, nausea, vomiting, flank pain are frequently seen. In addition, upper urinary and gastrointestinal system obstruction can be detected at the time of diagnosis (4, 5). Our patient also had flank pain, nausea, vomiting, and constipation for 6 months without any known causes which may be associated with the obstruction due to masses.

Serological methods such as ELISA, Western-Blot and (IHA) can be used in the diagnosis and follow-up of the disease. Immunoglobulin G antibody detection by ELISA has a sensitivity of 95% and a specificity of 94% (11). IHA sensitivity was 87.5% (11, 12). These methods are generally easy to apply, but it should be kept in mind that the diagnostic significance of such tests will decrease in cases where the cyst wall is intact and calcified, which will create a diagnostic dilemma for the surgeons. Any increase in titers following a decrease after resection may assist confirming of recurrence. In our case, we did not perform an IHA test owing to unusual presentation.

Modern radiological modalities lead the way to the diagnosis of HD. Calcifications of the cystic wall can be seen on abdominal radiographies (13). Ultrasonography (USG) can assist the physician about intraabdominal cystic lesions, but it may sometimes be insufficient for diagnosis due to rare localization and atypical appearance (14). CT can confirm the diagnosis by revealing the presence of daughter

cysts and plaque-like calcifications in the cystic wall. CT can also provide information about the exact localization of the cyst and reveals possible anatomical challenges. CT sensitivity ranges from 90% to 97% (13). MRI can provide more comprehensive visual information about lesions localized in hepatopancreaticobiliary region, muscles and kidneys, compared to USG. In our case, the presence of cystic lesion was demonstrated by USG and due to its atypical localization further examinations were performed with MRI.

Preoperative differential diagnosis is important. Pancreatic pseudocysts, pancreatic neoplasms, retroperitoneal sarcomas, extragonadal germ cell tumors, cystic lymphangiomas, renal cystic lesions, retroperitoneal abscess and embryonal cysts should all be considered in the differential diagnosis of retroperitoneal hydatid cyst (5). The final diagnosis is confirmed by surgical and histopathological examination (13). In our case, histopathological evaluation after excision showed that the lesion in the psoas was hydatid cyst, and the lesion in the pancreas was a pseudocyst.

Surgery had been acknowledged as the first-line of treatment of hydatid cyst in the history. However, conservative methods like "watch and wait" are among the treatment methods recently. If asymptomatic, watchful waiting (with or without albendazole treatment) of hydatid cysts can be performed (14). In cases where medical treatment is not successful or complications occur, surgery may be a choice. In surgery, it is essential to take the entire cyst - together with the germinative membrane - out of abdomen, in a controlled manner to avoid spillage into abdomen, which can trigger anaphylaxis. However, retroperitoneal cysts may be adjacent to kidney, ureter and large vascular structures, as in our case. Depending on the localization, it may not be possible to remove the lesion completely. If such situations are observed pre-operatively, the surgical procedure can be planned at a multidisciplinary (urology, cardiovascular surgery) level. Partial excision of the cyst is another surgical method that can be chosen in such situations to prevent organ damage. In selected cases, the cyst content can be drained with preparing an omentum patch for the cavity, which can help absorption of the liquid content and reducing the risk of secondary infections (15). After surgery, antiparasitic treatment (albendazole, mebendazole) can be given to reduce recurrence (3)

Primary hydatid disease in retroperitoneum is an extraordinary scenario, which should always be kept in mind and included in differential diagnosis by surgeons, especially in endemic regions. Inadequate management of a retroperitoneal hydatid disease may lead to serious complications such as rupture, secondary infections and intestinal obstruction.

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Conflict of interest

The authors declare that they have no conflict of interest.

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