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EDİTÖRE MEKTUP / LETTER TO THE EDITOR

Primer retroperitoneal hydatid cyst

Primer retroperitoneal hidatik kist

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To the Editor,

Cystic echinococcosis (CE) is endemic in the Mediterranean region Central Asia, the Middle East, East Africa, Australia, New Zealand and the southern part of South America[1]. Hydatid disease in humans is caused by the cystic (larval) stage of the tapeworm Echinococcus granulosus[2]. In 85-95% of the cases, the liver and/or the lungs are involved[3]. Primer retroperitoneal hydatid cyst is extremely rare and only occasional case reports have appeared since Lockhard and Sapinza first reported this entity in 1958[4]. Retroperitoneal hydatid cyst case with an atypical location and without a detected premier source is presented along with literature.

A 25 year-old man, who was administered his family physician with the complaint of left flank pain lasting for3 months. He has no past history of any surgery or medical problems. During this period he was apyrexial without any signs of acute infection. Physical examination revealed a non-tender fixed mass about 8x10 cm in the left lumbar area extending down to left iliac fossa. Complete blood count, liver and kidney function tests were normal. Indirect hemagglutination test for hydatid wasnegative. Plain X-ray abdomen did not show any specific diagnostic finding. Ultrasonography of abdomen revealed 11x8 cm cystic lesion in the left iliac fossa. Abdominal contrast enhanced CT scan revealed a large retroperitoneal cystic mass on the left side and pushing the psoas muscle (Figure 1).

Figure 1. Contrast enhanced CT scan abdomen revealed retroperitoneal cystic mass.

There was no evidence of similar cystic lesion in liver, lungs or any other organ. Colonoscopy revealed non extrinsic pressure on the region of the descending colon. Based on the clinical, serological and radiological evidence, a provisional diagnosis of retroperitoneal hydatid cyst could not be made. However, the differential diagnosis of retroperitoneal cystic mass includes retrorectal cyst, teratoma, cystic lymphangioma, abscess and necrotic malignant soft tissue tumours[5]. Exploratory laparotomy revealed a large cystic lesion with

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laminated membrane in the retroperitoneum extending from the lower pole of the left kidney down to pelvis pushing the left psoas muscle. The whole mass was excised intact by performing total cystectomy (Figure 2). Gross pathology of resected material was 11x8x7cm cyst including white fluid and daughter cysts (Figure 3). Histopathological examination of the cyst wall showed an outer multilayer laminated membrane (chitinous layer), an inner germinal layer and protoscolices (Figure 4). Laminated membrane was stained PAS (Periodic Acid Schiff) positive histochemically. Histopathological diagnosis was confirmed as hydatid cyst.

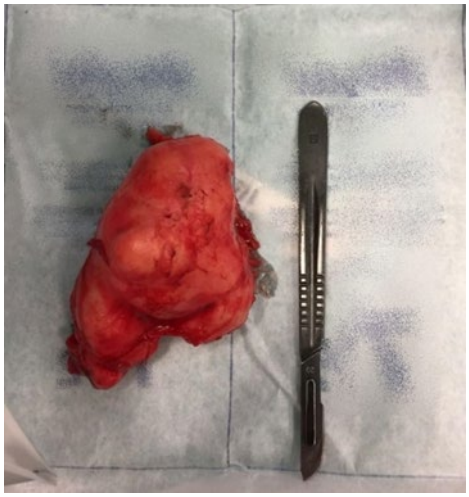

Figure 2. Complete resected retroperitoneal cyst

The patient had an uneventful postoperative recovery and the drain was removed after 2 days. Patient was discharged with antihelmintic drugs. The patient is at follow-up and doing well. Informed consent was obtained. Hydatid disease is an endemic parasitic disease with worldwide distribution. Man is an accidental host in the life cycle of Ecchinococcus granulosus. Human infestation occurs when the ova are swallowed. In the stomach, the outer protective coat of the ovum is digested, and the larvae are liberated. These penetrate the mucosa of the proximal bowel to enter the portal system. About 85-95% of the larvae are trapped in the liver, and the lung and only 5-15% of them escape into the systemic circulation to involve other organs, mainly the muscles, kidney, bone and brain3. So far, retroperitoneal lesions of newly diagnosed hydatid cysts consist only 1,1% of all the reported cases[5]. Retroperitoneal involvement was always thought to be secondary to rupture or spillage during surgery of liver hydatids. A primary retroperitoneal hydatid cyst without other organ involvement was first reported by Lockhard and Sapinza in 1958[4].

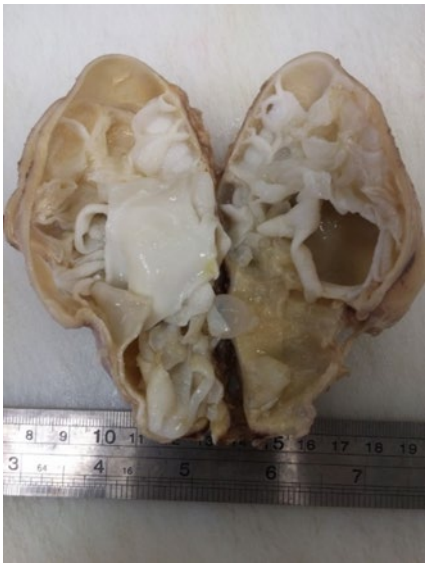

Figure 3. Gross pathology of resected cyst with visible jelly-like membrane consistent with daughter cyst.

Various modes of spread have been suggested to explain the escape of liver and lung involvement via lymphatics or via venovenous shunts within the liver and in the space of Retzius[6]. Waddel had favored air bone transmission and direct implantation of the embryo in the bronchial mucosa as another possible mode of entry[7]. This raises the possibility of an embryo of the parasite entering a venule after penetrating the bronchial mucosa and reaching the left side of the heart to involve other sites and thus bypassing the lung. But this remains largely theoretical and needs to be proved.

In our case retroperitoneal hydatid cyst was detected. The radiological examination revealed no other source of infestation and organ involvement other than retroperitoneal cyst. Thus the recent case was considered as primary retroperitoneal hydatid cyst. Retroperitoneal hydatid cyst disease grows slowly over years and the patient presents with lump in the abdomen and symptoms due to compression of the adjacent structures. Our case presented with lump in the abdomen and left flank pain for 3 months. Preoperative diagnosis of retroperitoneal hydatid disease is difficult. The differential diagnosis

of retroperitoneal cystic mass includes retrorectal cyst, teratoma, cystic lymphangioma, abscesses and necrotic malignant soft tissue tumours[5]. Diagnosis of hydatid cyst disease is made by imaging modalities and serological tests. Radiography, USG and CT studies are important for the diagnosis of disease. Plain abdominal X-rays may show calcifications of the cystic wall[8]. Ultrasonography is the method of choise for the detection of hepatic and extra hepatic echinococal cyst. CT confirmsthe diagnosis by revealing the presence of daughter cysts and plaque-like calcifications in the cyst wall. It is important as it provides information regarding the exact location of extrahepatic cysts in relation to neighbouring structures. CT sensitivity ranges from 90 to 97%[9]. Serological tests contribute the diagnosis. Immunoglobulin G antibody detection by ELISA has a sensitivity of 95% and specificity of 94%[10]. The sensitivity of indirect hemagglutination test has been found to be 87.6%10. In our case, ultrasonography and CT scanning showed multiple large cysts in the retroperitoneum. The exact diagnosis is made by means of surgery and histopathological examination[9].

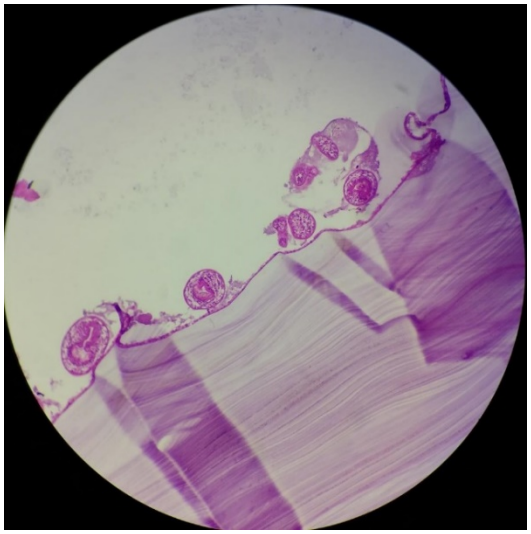

Figure 4. Microscopic examination of the pathological specimen demonstrating the outer multilayer laminated membrane, inner germinal layer and protoscolicsof the hydatid cyst wall (H&E 200×).

Therapy for extra hepatic echinococcal disease is based on considerations regarding the size, location and manifestations of the cyst, and the overall health status of the patient. Asymptomatic small cysts once diagnosed can be treated with antihelmintic drugs, administered for 28 days in one to eight repeating cycles, separated by drug-free intervals of 2-3 weeks. For symptomatic or large hydatid peritoneal cysts, surgery, when feasible, is the principal method of treatment. Surgical treatment can be either radical or conservative. Total cystectomy, whenever possible, is the gold standard. In our case, the whole mass was excised intact by performing total cystectomy.

Although the retroperitoneal location of hydatid cyst is rare, the differential diagnosis of retroperitoneal masses should be taken into consideration in particular endemic areas. Diagnosis of hydatid cyst disease may be made by imaging modalities, serological tests and the clinical history. The exact diagnosis is made by means of surgery or/and histopathological examination.

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