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Case Report / Olgu Sunumu

Unusual localization and coexistence of primary hydatid cyst: a case report

Birincil kist hidatiğin alışılmadık lokalizasyonu ve birlikteliği: olgu sunumu

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Abstract

Hydatid cysts are zoonotic parasitic infections caused by *Echinococcus granulosus*. Although witnessed in all body parts, the first and most important locations for this parasite are the liver and lungs. Unusually, hydatid cysts are rarely located in the pelvic region. The majority of such cysts generally develop secondary to spontaneous rupture of an hepatic hydatic cyst or due to surgical inoculation. Incidentally diagnosed in a patient admitted with the picture of acute abdomen, a case of primary hydatid cyst located in retrovesical region, an uncommon localization for hydatid cysts, is presented in this report. In patients admitted with symptoms of abdominal pain in endemic regions, and in those suspected of having unidentified cystic lesions, the unusual localization of a primary hydatid cyst should be considered.

Keywords: Acute abdomen, appendicitis, hydatid cyst, retrovesical region

Öz

Kist hidatik *Echinococcus granulosus* tarafından sebep olunan, zoonotik bir paraziter enfeksiyondur. Hemen hemen vücudun bütün bölümlerinde görülmesine rağmen, bu parazitin ilk ve en önemli yerleşim yeri karaciğer ve akciğerdir. Alışılmadık bir şekilde, kist hidatik nadiren pelvik bölgede yerleşebilir. Böyle, pelvik bölgede yerleşmiş kistlerin çoğu önceden karaciğerde bulunan kistlerin kendiliğinden yırtılmasına ya da herhangi bir cerrahi işlem sırasında kistin periton boşluğuna ekimine bağlı olarak gelişir. Bu olguda akut abdomen tablosuyla başvuran bir hastada, tesadüfen teşhis edilen, retrovezikal bölgede lokalize olmuş, alışılmışın dışında bir hidatik kist olgusu sunulmuştur. Endemik bir bölgede karın ağrısıyla başvuran ve tam olarak tanımlanmamış kistik lezyonu olduğundan şüphelenilen olgularda birincil kist hidatik akla gelmelidir.

Anahtar sözcükler: Akut batın, apendisitis, kist hidatik, retrovezikal bölge

Introduction

Hydatid cysts are zoonotic parasitic infections caused by *Echinococcus granulosus*. Although witnessed in all body parts, the first and most important locations for this parasite are the liver and lungs. Unusually, hydatid cysts are rarely located in the pelvic region. The majority of such cysts generally develop secondary to spontaneous rupture of an hepatic hydatic cyst or due to surgical inoculation. Incidentally diagnosed in a patient admitted with the picture of acute abdomen, a case of primary hydatid cyst located in the retrovesical region, an uncommon localization for hydatid cysts, is presented in this report.

Case

A 12-year-old boy was admitted to the pediatric emergency department with symptoms of vomiting and severe abdominal pain. On physical examination, tenderness in the right lower quadrant was also detected, as well as the following vital findings: temperature 37.4°C, pulse

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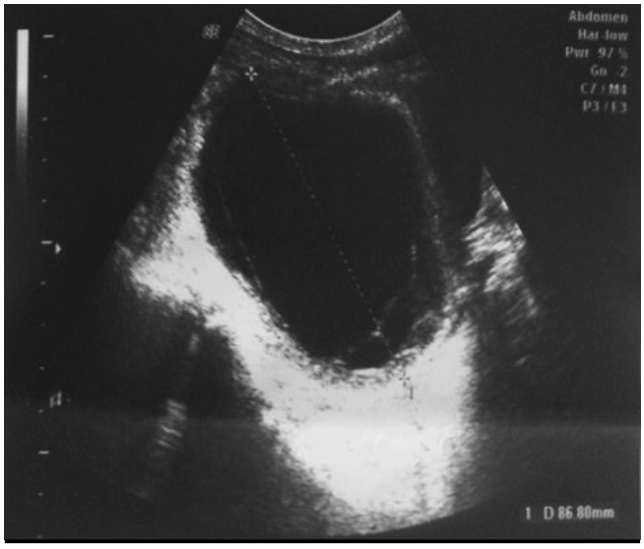

Figure 1. Preoperative ultrasonographic image of the cyst

rate 150 bpm, respiration 40 breaths/min, and blood pressure 105/70 mm Hg.

Laboratory findings revealed that hemoglobin was 15.4 g/dL, and the leucocyte count was $15.6\times10^3/mm^3$ with a differential of 55% neutrophils, 40% lymphocytes, and 5% monocytes. Serum creatinine, electrolytes, amylase, aspartate aminotransferase, alanine aminotransferase, alkaline phosphatase, and gamma-glutamyl transferase concentrations were within normal limits. The C-reactive protein concentration and erythrocyte sedimentation rate were 15.2 mg/dL and 85 mm/h, respectively. Urine sediment microscopy was normal; however, ketone, protein, and blood reactions were positive in urine dipstick test. The urine density was determined as 1.020.

In evaluating the abdomen via ultrasonography (USG), a retrovesical cystic mass with septate and 9 cm in diameter was determined (Fig. 1). In the differential diagnosis, the patient was considered to be hematoma or plastron to appendicitis and operated on. A cystic material of nearly 10 cm that was located in retrovesical region was intraoperatively detected and totally removed (Fig. 2). Then, appendectomy was performed.

An indirect hemagglutination test was negative for hydatid disease. Histo-pathologic examinations showed the features of hydatid cyst and appendicitis (Fig. 3). Based on these findings, the diagnosis of hydatid cyst and appendicitis was verified, and treatment of albendazole was initiated. The patient was given a 4-week courses of albendazole of 15 mg/kg per day, which was followed by a 14-day free-interval for a total of 3 cycles. Investigations related to the primary focus showed no other involvement sites of the hydatic cyst on chest radiography and abdominal

Figure 2. The excised cyst

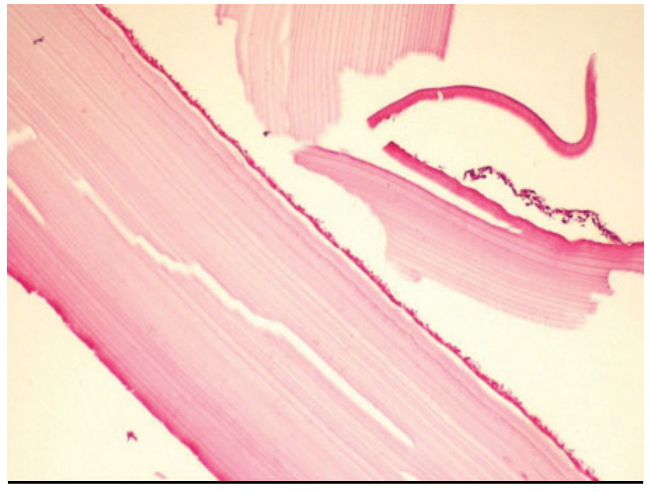

Figure 3. Histologic view of the cyst showing cuticula and germinative membrane (HEx400)

computed tomography (CT). The patient was assessed as having a primary retrovesical hydatid cyst. No recurrence of hydatid cysts was seen for one year within postoperative period. Written informed consent was obtained from the patient's parents for publication of the case report and accompanying images.

Discussion

Hydatid cysts are seen as a common condition in endemic regions, generally involving the liver or the lungs (1). Intraperitoneal or pelvic hydatid cysts usually develop secondary to spontaneous or traumatic perforation of hydatid cysts located in another organ. Retrovesical hydatid cysts constitute 0.1–0.5% of all cases of hydatid cysts (2).

Among these, primary cases are quite rare, however. Primary formation of hydatid cysts is uncommon without any focuses in other organs. The physiopathologic mechanism of primary retrovesical hydatid cyst formation has yet to be clearly enlightened, but different assumptions related to their formation have been proposed.

The classic theory is that primary intraperitoneal hydatid cysts with resultant seeding of their contents are formed in the pouch of Douglas due to blunt trauma. Primary cysts may, then, heal leaving behind an unremarkable scar. In addition, hematogenous seeding or a direct access of the embryos via the rectosigmoid mucosa to the pelvic venous plexus and perivesical tissue are hypothesized as possible mechanisms leading to the formation of hydatid cysts (3).

A hydatid cyst located in the retrovesical region can cause symptoms related to the mass effect on adjacent organs including the bladder, ureters, rectum, seminal vesicles, and vas deferens. While the presenting symptoms can be palpable mass, flank pain, urinary retention, obstruction and urgency in children, they are hematospermia and obstructive azospermia in adult males and adolescents, and obstructed labor in women (4–6). In our patient, however, the main symptoms were detected as vomiting and severe abdominal pain. We consider that the symptoms in our patient developed due to acute appendicitis, and that our patient's hydatid cyst was incidentally diagnosed.

The sensitivity of serologic tests is low in the diagnosis of hydatid cysts. In the study performed by Akbulut et al. (7), serologic tests determined that 67.8% of patients were indirect hemagglutination positive, and 71.4% were positive on enzyme-linked immunosorbent assay. The serologic test result was also negative in our case. Ultrasonography and CT are assistant imaging techniques used to diagnose hydatid cysts, especially in atypical localized cysts (8). However, it is difficult to diagnose, especially with intraabdominal hydatid cysts in the preoperative period via radiologic instruments. In the differential diagnosis, disorders such as congenital mesenteric cysts, retroperitoneal mesenchymal tumors, and pancreatic pseudocysts should be considered. The diagnosis of preoperative acute abdomen (plastron to appendicitis) was initially considered for our case, and then the definitive diagnosis was established after surgical intervention.

In a study by Mottahian et al. (9), nearly 10% of cases were reported to recur due to surgical contamination in hydatid cyst disease. While treating, the whole cyst is aimed to be excised without rupture, and so spreading of echinococcosis to surrounding tissues and anaphylactoid reactions are prevented. There are also studies reporting that treatment with albendazole decreases recurrence rates (10). In our case, we also started treatment with albendazole in the postoperative period. If the hydatid cyst was diagnosed in the preoperative period, our patient could have been started on albendazole preoperatively. Fortunately, the hydatid cyst was removed successfully without rupture.

Conclusion

In patients admitted with the symptoms of abdominal pain in endemic regions, and in those suspected of having unidentified cystic lesions, unusual localization of primary hydatid cyst should be considered. If necessary, further imaging can be performed using pelvic CT. Therefore, the risk of anaphylaxis and spillage of hydatid cysts into the abdomen may be reduced with preoperative albendazole therapy.

Informed Consent: Written informed consent was obtained from the patient's parents for publication of the case report and accompanying images.

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