Primary Hydatid Disease in the Adrenal Gland: A Case Report

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Hydatid cysts may be found in almost any part of the body, but most often they are found in the liver and lungs. Other organs that are occasionally affected include the brain, muscle, kidney, heart, pancreas, and adrenal and thyroid glands. We report a case of hydatid cyst of the adrenal gland.
Key Words: hydatid cyst, hydatid disease, adrenal.

Hydatid cyst of the adrenal gland is rare; this entity is found in only 7% of all adrenal cysts. It occurs in cases of disseminated infection by *Echinococcus granulosus* beyond the hepatic and pulmonary filters. The infection is usually asymptomatic. When symptoms are present, most are related to local visceral compression. Eosinophilia occurs in one-fourth of cases; the sensitivity of serologic tests is 90%. We examined a case of an adrenal mass diagnosed finally as a hydatid cyst.

Case report

A 54-year-old woman with a two-year history of arterial hypertension was referred to the Department of Internal Medicine at the University of Istanbul. As part of the diagnostic procedures, clinical examination was followed by abdominal ultrasonograpy (US) and abdominal magnetic resonance imaging (MRI). US of the abdomen revealed a 5 x 5 cm solitary mass in the right retroperitoneal area.

An MRI scan of the abdomen demonstrated a 4.5 x 3.5 x 3 cm solid adrenal mass. No other intraabdominal masses were found (Figure 1). As a screening procedure, the patient underwent the following endocrine evaluation: baseline serum cortisol, serum baseline 17 hydroxyprogesterone (17-OHP), dehydroepiandosterone sulfate (DHEA-S), and low-dose 2mg dexametasone suppression tests. In all patients, urinary metanephrin, normetanephrin, and vanilylmandelic acid (VMA) excretion and plasma aldosterone to plasma renin activity ratio and serum potassium levels were also determined to evaluate the possibility of pheochromocytoma or an aldosterone-producing adenoma. Serum baseline cortisol was 12 µg/dL, 17-OHP was 0.6 ng/mL, DHEA-S was 65 µg/dL, 24 h urinary excretion of metanephrine was 200 µg/24h, 24 h urinary excretion of normetanephrine was 130 µg/24h, and 24 h urinary excretion VMA was 4.5 mg/24h. The plasma aldosterone/plasma renin activity ratio was 12.

Received on 11 May 2006; revised 15 September 2006.
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The Brazilian Journal of Infectious Diseases 2006;10(5):362-363.

The patient underwent surgery, with a preoperative diagnosis of the incidental adrenal solid mass. In the surgical exploration, a 5 x 4 cm solid mass was defined, adhered to the neighboring structures of the liver and kidney and compressed against the right kidney's upper pole. After blunt and sharp dissection, the entire mass, together with normal adrenal tissue, was removed. In the cross sections of the specimen after removal, daughter cysts and the existence of a germinating membrane revealed a hydatid cyst (Figure 2). Histopathologic examination confirmed hydatid disease in the adrenal gland. In the immediate postoperative period, a complement fixation test was positive. After the operation, albendazole treatment (10/kg/kg) for six months was prescribed.

Discussion

Hydatid cysts of *Echinococcus granulosus* can develop in any part of the body. The thyroid gland is an extremely rare site of hydatid cysts, even in endemic regions. Among hydatid cysts, 60% to 70% are found in the liver, 5% to 15% in the lungs, and only 0.5% in the adrenal gland. Hydatid cysts are included in the group of adrenal cysts, and they account for only 6% -7% of all adrenal cysts. Hydatid cysts in the adrenal gland are usually secondary, and only 15 cases have been reported as primary. The etiological-morphological classification of adrenal cysts is as follows: 1. endothelial cysts (45%), 2. pseudocysts (39%), 3. epithelial cysts (9%), 4. parasitic cysts (7%).

Because most adrenal cysts are asymptomatic, they are usually found as incidental findings in imaging studies or incidentally during surgery performed for other abdominal pathologies [1]. Hydatid disease of the adrenal gland is usually asymptomatic. When symptoms are present, most are related to local visceral compression. The most prominent clinical features consist of pain in the lumbar area, gastrointestinal symptoms (bloating, fullness, nausea, vomiting, constipation, and anorexia), and a palpable mass. Rarely, hydatid cysts cause hypertension. Hydatid disease of the adrenal gland with hormonal function has not been reported. The most common presenting symptom is pain. Anaphylactic shock may be caused by the rupture of a hydatid cyst. Adrenal cysts can be fatal if they hemorrhage and are not rapidly diagnosed. It is thought that hemorrhage occurs secondary to trauma or due to some toxic or infectious process [2,3].

Figure 1. A 4.5 x 3.5 x 3 cm homogenous mass is seen neighboring the right adrenal gland. Moderate hyperintensity in T2W images is present, and peripheral rim-like contrast enhancement was obtained after contrast administration.

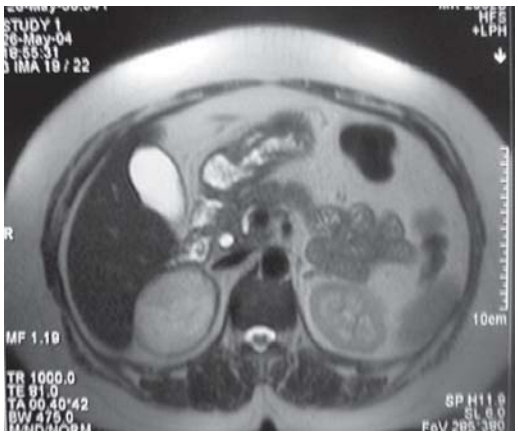
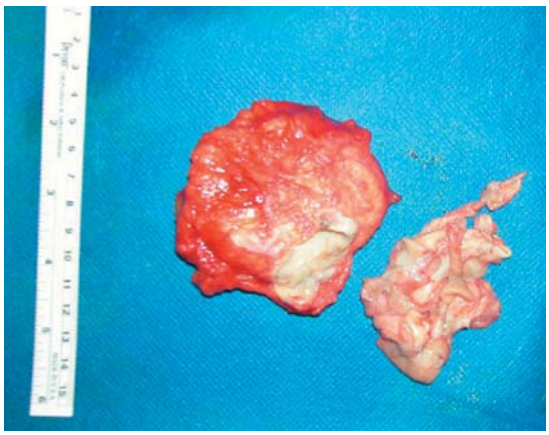


Figure 2. Overall view of the adrenal cyst mass and daughter cysts after removal of the lesion.



Immunodiagnostic tests are helpful, but the actual diagnosis is usually surprising. Eosinophilia occurs in one-fourth of cases; the sensitivity of serologic tests is 90%. There are many new sensitive and specific serological tests available, such as complement fixation and specific hydatid IgE tests [2,3]. We used the complement fixation method for our patient, and it was positive.

Radiology has an important role in the detection of adrenal masses. Differentiation of benign *versus* malignant lesions of adrenal gland also mostly depends on radiological examinations. The detection of adrenal masses has increased over the last decade with the help of cross-sectional imaging modalities, precisely tailored contrast examinations and reconstruction techniques. Computed tomography is usually the primary imaging modality for both detection and differentiation of adrenal masses; dedicated thin-sliced images with coronal reconstruction depict the anatomical relationships clearly. Chemical shift magnetic resonance imaging is useful for masses that can not be clearly defined by CT. Adrenal cysts have been mostly diagnosed incidentally, without any specific symptoms, and most patients require further investigation with CT or MR, due to suspicion of malignancy with nodular thickening, septations, solid components or irregular thick walls. To identify a hydatid cyst in the adrenal gland, US, CT, and magnetic resonance imaging (MRI) can demonstrate cystic lesions and reveal daughter cysts. The imaging features depend on the stage of evolution of the disease. On CT scans, concentric areas of septation and calcification indicate that a cyst is of parasitic origin. US and CT may be able to successfully image all cysts, but the definitive diagnosis is made by macroscopic and microscopic examination of the cyst's content [4]. Early lesions appear purely cystic; after modifications of the germinal layer and reduction of intraluminal pressure, the capsule becomes fibrotic and sometimes appears calcified, while the daughter cysts discharge from the wall and float in the lumen. Stage 4 and 5 hydatid cysts are seen as solid masses in CT and US. Since our case had a stage 4 lesion, all preoperative radiological imaging methods defined it as a solid mass [4].

Indications for surgery for cystic lesions of the adrenal gland include large and complicated cysts, as well as parasitic, functioning, and malignant cysts. Percutaneous drainage should be avoided, especially in areas endemic for hydatid disease. The definitive treatment method for hydatid cysts of the adrenal gland is surgical excision. Chemotherapy is necessary to avoid recurrence.

In conclusion, although hydatid cysts in the adrenal gland are rare, they should be included in the differential diagnosis of cystic lesions, especially in endemic regions.

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