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Case Report

Primary hydatid cyst of adrenal gland: Case report^{☆,☆☆,★}

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ABSTRACT

The hydatid disease is an anthropozoonose caused in humans by a larval form of *Echinococcus granulosus*. Adrenal gland is a rare and unsymptomatic site. We illustrate the case of a 46-year-old patient, admitted for left hypochondrial pain with back radiation. The CT reveals a multiloculated, cystic mass on the left adrenal gland, which contains septae and a calcified wall.

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Introduction

Hydatid cysts can affect all organs and tissues [1,2]; however, in the human body, liver (45%-77%) and lungs (10%-50%) are the two most commonly affected organs [3,4,7,9,10]. Hydatid cyst disease is rarely seen in the adrenal glands. Hydatid cysts can occur in the adrenal glands as primary (isolated) or secondary (associated with other organs) disease. In this article, we try to remind the clinical features and to clarify the diagnostic input of imaging.

Case report

A 46-year-old patient, without any particular history, consulted for left hypochondrial pain with back radiation without any other signs. Physical evaluation was normal. Abdominal ultrasound revealed a cystic mass over the kidney, containing vesicles and calcifications causing a posterior shadow cone. It measured 6 cm in length. A hypersecretion test was performed (methoxyldinated derivatives, normotadrenaline, metadrenaline, cortisol) and was negative. Abdominal CT

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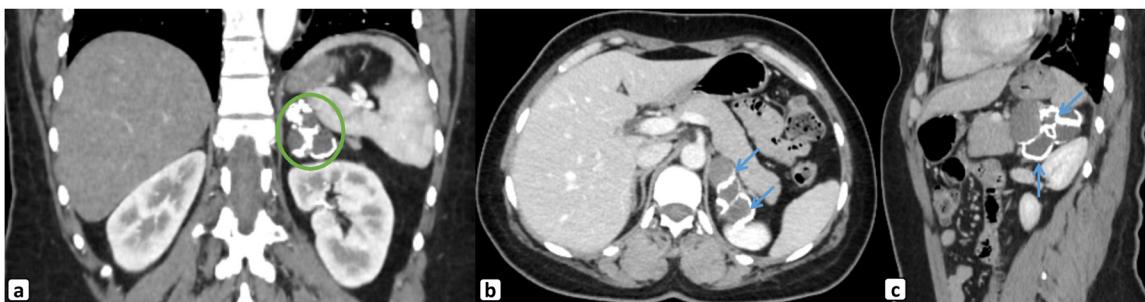


Fig. 1 – Abdominal CT scan after injection of contrast product: a) coronal section, b) axial section, c) sagittal section: Retroperitoneal mass at the left adrenal gland, well limited, multiloculated, homogeneous, hypodense, contain calcified membranes, not modified after contrast at the different injection times and measuring 60 × 30 mm, in relation with an adrenal cyst.

revealed a multiloculated cystic mass, developed on the left adrenal gland, containing septae and a calcified wall (Fig. 1). The diagnosis of adrenal hydatid cyst was suggested. Hydatid serology (Elisa) was performed and was positive. The diagnosis of left adrenal hydatid cyst was maintained. Chest X-ray and abdominal ultrasound did not reveal any hepato pulmonary hydatid localization. The patient benefited from total excision of the lesion. No recurrence was noted during the one-year follow-up.

Discussion

Adrenal location is an extremely rare location of hydatid disease [11]. All age ranges can be affected, but this site seems to be more common among 50–60 year olds, and slightly more common in women [6,12]. Adrenal hydatid cysts (AHC) are usually asymptomatic lesions, and are frequently found incidentally, on radiologic investigations made for other reasons. They are rarely complicated and become symptomatic most of the time due to symptoms that develop as a consequence of pressure. The most common symptoms are pain existing due to inflammation of adjacent tissues caused by the cyst [6]. The most serious complication of CAH disease is cyst rupture, which causes anaphylaxis and hemorrhage [5]. AHC can apply pressure on the renal artery. This condition is called the Goldblatt phenomenon, and it can lead to hypertension [5,6,8,10]. AHC can exert pressure on the adrenal medulla, causing pheochromocytoma-like symptoms, such as headaches, palpitations, and hypertension. Another serious complication is fistulation of the CAH to adjacent intestinal structures [7].

Identifying hydatid cyst in adrenal gland is mainly based on ultrasonography and CT [13,14], ultrasound has 93%–98% sensitivity for diagnosis, while CT is about 97% [15]. In radiology, hydatid cysts depend on the stage of evolution of the disease and can be described according to the classification of Gharbi et al. [16], which is based on ultrasound characteristics and includes five types. Type I is a well-defined, anechoic lesion. Type II shows membrane separation; the lily pad sign is

formed by the wavy membrane. Type III is characterized by intraluminal septa and daughter cysts. Type IV is a nonspecific solid mass. Type V is a solid mass with a calcified capsule that can be identified by abdominal radiography as calcification of the cystic wall.

The diagnostic differential of adrenal hydatid cyst is with other adrenal cystic masses: cystic lymphangioma, epithelial wall cyst, calcified hemorrhagic cyst, and extra-adrenal cystic masses.

Treatment of HD of the adrenal glands is mostly surgical. Pericystectomy and resection of the entire adrenal gland are the 2 preferred choices. Resection of the cyst with conservation of the gland remains the optimal procedure. Puncture, aspiration, injection, and re-aspiration (PAIR) treatment of hydatidosis is considered minimally invasive, confirms the diagnosis and improves the efficacy of chemotherapy given before and after puncture. PAIR is usually avoided in patients with adrenal hydatidosis, but can become an alternative method in inoperable cases [17]; however, many authors contraindicate its use because of potential complications, such as anaphylactic shock and the spread of daughter cysts [18]. Antihelmintic agents have been used in the treatment of systemic echinococcosis in endemic areas [18]. There are reports that antihelmintic agents can reduce the size of cysts in some cases, however the results are not satisfactory and this treatment should be limited for disseminated and recurrent cysts or in cases where surgery is contraindicated [18]. Prevention of hydatid contamination (by interrupting the parasite cycle) remains an essential measure to avoid hydatid disease, whatever its location.

Conclusion

Hydatid cyst of the adrenal gland is an uncommon pathology, which should be suspected in case of any cystic tumor of the adrenal gland, especially in endemic countries. The clinical aspects of this disease are variable and nonspecific. Preoperative diagnosis based on imaging (ultrasound and CT scan) coupled to hydatid immunology can be difficult especially when

the cyst is univesicular, with a noncalcified wall and negative immunology. Surgery is the treatment of first choice.

Ethics approval and consent to participate

Oral and signed consent was obtained from the patient concerned. The study was conducted anonymously.

Patient consent

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