Coexistence of Primary Adrenal Hydatid Cyst and Arterial Hypertension: Report of a case and review of the literature

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Abstract. Adrenal gland has been considered as an atypical localization of Echinococcus Granulosus and the reported incidence is 0.5%. We report a rare case of coexistence of primary adrenal hydatid cyst and arterial hypertension. The patient underwent surgical excision of the adrenal gland with a slight improvement of blood pressure’s regulation, requiring antihypertensive medication postoperatively. Till today, two cases of coexistence of primary adrenal hydatid cyst and arterial hypertension have been reported in the literature. However there is not clear and acceptable explanation about the relation and the involved pathogenetic mechanism. Resection of the cyst with conservation of the gland remains the optimal procedure. In case of hemorrhage or failure to perform a cystectomy, ablation of the entire adrenal gland including the cyst should be performed.

Introduction

Despite the improved knowledge regarding parasitic biology and the introduction and widespread use of chemotherapeutic agents, hydatid disease is still considered an occupational public health problem for sheep farmers. Human echinococcosis is still endemic in some areas of the world, including Mediterranean countries. Most human infections are due to Echinococcus granulosus transmitted between synanthropic hosts and livestock, while Echinococcus multilocularis infection, is uncommon and appears to be endemic in some specific areas.

The liver is the first and main barrier of parasitic embryos migrating from the intestine. The liver the lung and the spleen are the organs most frequently involved in human echinococcosis (80-95%) (1-3). Adrenal gland has been considered as an exceptional localization of the hydatid cyst. Parasitic cysts involving the adrenals are usually secondary and part of generalized echinococcosis. The reported incidence of adrenal hydatidosis is 0.5% (4). We report a rare case of coexistence of primary adrenal hydatid cyst and arterial hypertension. The patient underwent surgical excision of the adrenal gland without improvement of blood pressure’s regulation, requiring antihypertensive medication postoperatively.

Case report

A 61-year old woman was admitted to the Emergency Department of our hospital with epigastric pain radiating to the back, nausea and vomiting which had occurred 3 hours prior to presentation. In the morning, she began to feel abdominal distension. The patient had a 2-year history of arterial hypertension - P_SYSTOLIC 170-200 mmHg and P_DIASTOLIC 90-110 mmHg - requiring antihypertensive therapy. The hematological examination was characterized by a slight augmentation of white blood cells (11.000/mm³) and by eosinophilia 7%. The biochemical tests were within normal levels. Analysis of the urine proved a microscopic haematuria. As part of the diagnostic procedures a computed tomography scan was performed which showed a solitary cystic lesion 5.5 by 5.8 cm. with curvilinear calcification in the left adrenal gland. No other intraabdominal masses were detected. The presumptive diagnosis was a primary adrenal cyst.

Serological examination revealed a positive titer for immunoglobulin antibodies to Echinococcus Granulosus. The patient underwent an open laparotomy through a midline incision and the cystic lesion of the left adrenal gland was identified. The abdominal cavity and especially the region around the cyst was carefully protected and isolated by pads soaked in hypertonic 20% saline solution. As a plane of cleavage between the cyst and the adrenal gland could not be found the cyst was aspirated and drained. Thereafter, a pericystic excision was attempted but a severe hemorrhage forced us to remove the entire adrenal gland. The macroscopic examination revealed a germinate membrane and daughter cysts and the microscopic examination revealed scolices in the cyst fluid. Histological examination confirmed the diagnosis. The postoperative course was uneventful and the
patient underwent therapy with albendazole (500 mgr/12 hours) for six months. The arterial blood pressure was slightly lowered postoperatively still requiring medical support.

Discussion

The liver is the first and main barrier of parasitic embryos migrating from the intestine and gaining access to the portal circulation. Although most embryos are trapped in the liver, some pass through to the lungs or to other parts of the body. The liver and the lung are the organs most frequently involved in human echinococcosis (80-90%) (1, 2). However any organ can be potentially affected in the human body (3, 5, 6). The adrenal gland has been considered as an atypical localization of the hydatid (1, 7). The diagnosis of an adrenal cyst is usually incidental, and the diagnosis of hydatid cyst is seldom made preoperatively. The clinical presentation is insidious and the most common presenting symptom is abdominal pain.

The reported case presents two important elements. First, primary hydatid disease of the adrenal gland is rare and only few cases have been reported in the literature. Second, the patient underwent surgical excision of the adrenal gland with a slight improvement of blood pressure’s regulation, requiring antihypertensive medication postoperatively. The patient had a 2-year history of arterial hypertension. Till today two cases of coexistence of primary adrenal hydatid cyst and arterial hypertension have been reported in the literature (8, 9). However there is not clear and acceptable explanation about the relation and the involved pathogenetic mechanism. High blood pressure appears to be a multifactorial disorder in which the interaction of several components with each other and with the environment is important.

According to the diagnostic approach computed tomography (C.T.) scan has an increased accuracy in identifying cystic lesions of the adrenal gland. Depending on the condition of the parasite, the host reaction, and therapy, the hydatid cyst will degenerate leaving an area of calcification in the host tissue (10). The most typical imaging feature of adrenal hydatidosis is the characteristic calcification of the cystic wall. However tumors of the adrenal gland may present similar imaging presentation with rounded calcification. The differential diagnosis of an adrenal cyst should include the endothelial cyst, pseudocyst due to infarction or hemorrhage in the adrenal gland, cystic neoplasm and the post-traumatic cyst (7, 11, 12). Serological techniques involve isolating native or recombinant parasite antigens to detect specific serum antibodies in patients with suspected cystic disease (8). A judicious association of usual techniques such as indirect immunofluorescence assay, indirect hemagglutination assay, immunoelectrophoresis, co-electrophoresis, confirms the diagnosis in 65% to 94% of hepatic hydatidosis cases and in 65% of pulmonary hydatidosis cases (9, 11, 13).

Regarding to the surgical techniques, pericystectomy and resection of the entire adrenal gland are the two preferable choices. Resection of the cyst with conservation of the gland remains the optimal procedure. It is preferable an appropriate dissection plan between the cyst and the adrenal gland to be attempted in order a complete cystectomy to be achieved. However it is not always feasible and may cause haemorrhage owing to close adherence of the cystic wall to the adrenal’s gland parenchyma. In case of haemorrhage or failure to perform a cystectomy, ablation of the entire adrenal gland including the cyst should be performed. Partial cystectomy includes a higher risk of dissemination of the parasite due to the dissection’s maneuvers.

References


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