

Hemorrhagic adrenal cyst

Cisto hemorrágico da glândula adrenal

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ABSTRACT

The authors present a case of a hemorrhagic adrenal cyst, one of the tumors known in literature as incidentalomas, emphasizing the clinical characteristics, since adrenal cysts or pseudocysts are generally rare and observed by chance during imaging procedures. Traditionally they are classified as pseudocysts, endothelial, epithelial or parasitic cysts. Laparoscopic adrenalectomy has been considered the treatment of choice for benign, functioning or non-functioning adrenal lesions. Small cystic adrenal tumors can be managed conservatively by laparoscopic decortication or marsupialization, but larger cysts should be treated by total or partial adrenalectomy.

Keywords: Adrenal gland diseases/diagnosis; Adrenal gland diseases/surgery; Cysts/diagnosis; Adrenalectomy; Laparoscopy/methods; Case reports

RESUMO

Os autores apresentam um caso clínico de cisto hemorrágico da glândula adrenal, conhecido na literatura como uma das doenças do grupo dos incidentalomas, enfatizando características clínicas, como cistos e pseudocistos da adrenal, que são raros e geralmente diagnosticados incidentalmente por métodos de imagem. Tradicionalmente, essas lesões são classificadas em: pseudocistos, cistos endoteliais, epiteliais e parasitários. A adrenalectomia laparoscópica vem sendo considerada a conduta cirúrgica de escolha para o tratamento das lesões benignas da adrenal (funcionantes ou não funcionantes). As pequenas lesões císticas da adrenal podem ser manuseadas conservadoramente pela decorticação ou marsupialização laparoscópica; no entanto, os

cistos maiores devem ser tratados por meio de adrenalectomia total ou parcial.

Descritores: Doenças das glândulas supra-renais/diagnóstico; Doenças das glândulas supra-renais/cirurgia; Cistos/diagnóstico; Adrenalectomia; Laparoscopia/métodos; Relatos de casos

INTRODUCTION

Adrenal cysts and pseudocysts are a rare condition usually incidentally diagnosed by imaging methods^(1,2). Patients are asymptomatic until the tumor becomes large enough to produce mass effect on adjacent organs. Traditionally, these lesions are classified as pseudocysts or endothelial, epithelial, and parasitic cysts. Pseudocysts are the most common and comprise a heterogeneous group, in which a specific type of apparent vascular origin, characterized by well-defined histological and immunohistochemical findings⁽³⁾ is the so-called vascular hemorrhagic adrenal pseudocyst.

These pseudocysts show unique microscopic features, as they are composed of a fibrous hyalinized capsule containing clotted blood, residual adrenocortical tissue, and thin-walled vessels without identifiable cystic membranes. This lack of epithelial layer characterizes it as a pseudocyst. They respond strongly to type IV

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collagen stain, to VIII-AR (related antigen), CD34, and laminine. These immunohistochemical reactions strongly suggest the vascular origin of the lesions. However, there are other hemorrhagic pseudocysts occurring due to trauma and non-traumatic adrenal bleeding, such as stress, tumors, bleeding diathesis or coagulopathies. Those of unknown cause are called idiopathic. However, these pseudocysts are very seldom found in association with cortical neoplasm (carcinoma or adenoma) and pheochromocytoma^(1,3).

Laparoscopic adrenalectomy is considered the surgical method of choice to treat benign functioning or non-functioning adrenal lesions. Several papers show the advantages of the laparoscopic approach, when compared to open surgery. Besides the improved cosmetics, the technique leads to less bleeding in the intraoperative period, decreased pain in the postoperative period (less use of painkillers), decreased length of hospital stay and lower morbidity rates. Although these papers only comment on the use of laparoscopy for limited-size benign solid tumors, videolaparoscopy increasingly has gradually been used to treat larger lesions including malignant tumors^(1,3,4).

Small cystic adrenal lesions can be managed conservatively by laparoscopic decortication and marsupialization, but larger cysts should be treated by partial or total adrenalectomy. A recent study described the removal of a giant adrenal cyst by laparoscopic surgery⁽⁵⁻⁸⁾.

CASE REPORT

A 22-year-old black, female patient with a history of trauma (fall from standing height) five days before. After falling she felt abdominal pain, more intense in the right hypochondrium and flank. She reported no nausea, fever and/or mucous and sanguinolent diarrhea. One day before coming to the *Girassol* Clinic Emergency Room, she still suffered pain and new signs or symptoms were jaundice, postprandial vomiting, anorexia, and inability to pass stool or gas. Upon examination she was conscious, Glasgow 15/15, calm, collaborative, and afebrile. The vital signs were heart rate of 110 bpm, thready pulse; blood pressure of 122/90 mmHg; temperature of 36°C; respiratory rate of 22 rpm. The regional physical exam showed bulging in the right hypochondrium; pain on deep palpation and hepatomegaly (3 cm below the lower edge of the ribs) with smooth, regular edges, and little mobility; positive Murphy sign. Laboratory tests upon arrival (blood tests, biochemistry and echography): glucose 129 mg/dL; creatinine 0.6; urea 22 mg/dL; GPT

17; GOT 35; direct bilirubin 0.1; platelets – 241 x 10³; red blood cells 3.90; Hb 8.7 g/dL; hematocrit 29%; leukocytes 9.89 x 10³ (neutrophils - 84.1%; lymphocytes – 9.1%; monocytes – 6.8%); Widal test (H: 1/320, O: 1/80); amylase 220 IU/L.

Echography: hepatomegaly with large semiliquid hyperechoic lesion in the right hepatic lobe pressing the right kidney and hepatic portal vessels, measuring 143.8 x 122.6 mm, with septa inside (Figure 1).

Initially, according to tests available, our presumptive diagnosis was an amoebic liver abscess and the patient was hospitalized for workup and treatment. A gradual drop in hemoglobin levels was observed, an average of 1 g/day. Therefore, an ultrasound-guided fine needle aspiration was performed at the Department of Gastroenterology, obtaining abundant bloody fluid. As this indicated acute abdominal bleeding, we performed an axial computerized tomography (CT) that revealed a large retroperitoneal cystic mass pressing the right kidney and the portal system vessels due to mass effect (Figure 2).

After discussing in the group, the patient was taken to the operating room with a diagnosis of right adrenal hemorrhagic cyst.

Since the patient was hemodynamically unstable, the surgical team decided to perform open adrenalectomy right after the axial CT (Figures 3 to 5).

Figure 6 shows the microscopic image, indicating the capsule and histological tissue.

A right lumbar incision was the best alternative, since this large retroperitoneal mass explained hemodynamic changes mentioned. The patient had an evident and established picture of abdominal compartment syndrome. It is noteworthy that due to the mass size and the compartment effect on the other retroperitoneal structures, the surgeons decided to drain the cyst content

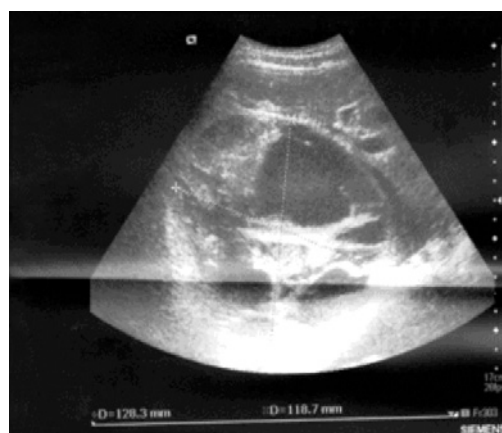


Figure 1. Abdominal echography showing cystic, septated mass



Figure 2. Abdominal axial computerized tomography showing a large mass over the right kidney causing extrinsic compression already causing some mass effect, medically expressed as intra-abdominal hypertension



Figure 3. Identification of the retroperitoneal cystic mass and content aspiration



Figure 4. Surgery the cyst capsule is open and the content is aspirated

and to remove it completely. A Malecot drain was left in the retroperitoneal cavity for 48 hours after surgery. Because there was little drainage and no suspicious exudate that could be sent for microbiological evaluation and culture, the drain catheter was removed and the patient discharged 96 hours after surgery.



Figure 5. Cystic capsule after exeresis. Note the large lesion

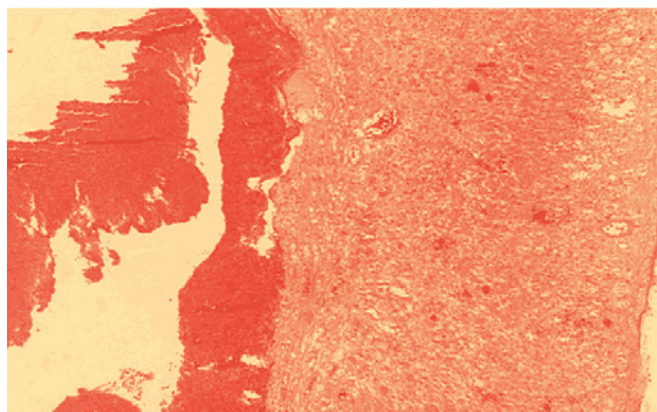


Figure 6. Microscopic image showing the capsule and cystic tissue

DISCUSSION

The case presented is in accordance with the literature on incidental findings of adrenal cysts and pseudocysts, that is, most diagnoses are made by chance during routine abdominal imaging examinations or resulting from pelvic or abdominal trauma⁽⁴⁾. It is not easy to make diagnosis of adrenal hemorrhage, especially due to its unspecific presentation, generally associated to other clinical complications⁽⁹⁾.

Initially, the echographic parameter made us thought of an amebic hepatic abscess, since Subsaharian Africa is an edemic region of amebiasis⁽¹⁰⁾. However, repeated and progressive blood loss verified by hemoglobin drop led the surgical team to choose an invasive procedure, the ultrasound-guided percutaneous biopsy revealing a serous and sanguinolent exudate. Considering the dubious interpretation, clinical worsening, abdominal distention and pain, an axial CT of the abdomen was decisive for diagnosing the right adrenal hemorrhagic cyst⁽¹¹⁾.

Although the best references in the literature state that video-guided surgery is the best treatment available

for adrenal cysts^(12,13), and we fully agree, the learning curve in trauma situations and the intra-abdominal mass effect, the retroperitoneal localization and the fact that our results are better with open surgery had to be taken in account. Thus, one of the aims of this case report is the exchange of ideas about the best approach.

CONCLUSION

An adrenal hemorrhagic cyst case in which surgical resection was a safe option leading to a good outcome was reported.

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