CASE REPORT

A giant adrenal cyst with an uncertain preoperative diagnosis causing a dilemma in management

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Introduction

Adrenal cysts are rare, and as at 2010, only about 600 cases had been documented in literature. Often asymptomatic, adrenal cysts may attain huge sizes and present challenges at diagnosis and dilemma in management. Only four previously documented adrenal cysts greater than 20-cm diameter were correctly diagnosed preoperatively. A 63-year-old man with an asymptomatic huge left sided retroperitoneal cyst originating from the left adrenal gland presented a dilemma in management from an uncertainty in diagnosis after ultrasonography and computer tomography (CT) imaging.

Case Report

Mr AA, a 63-year-old man, presented 10 days after suprapubic cystostomy for bladder outlet obstruction and hematuria. He gave an indefinite history of a left sided abdominal mass. He had no history of trauma or abdominal pains and no gastrointestinal symptoms. On examination, he had features of sepsis and the cystostomy wound was infected and discharging pus. His blood pressure was 140/90. He also had a huge nontender cystic mass in the left side of his abdomen extending from the left coastal margin into the pelvis. It was dull to percussion. Abdominal ultrasonography revealed a huge unilocular cyst and suggested a pancreatic pseudocyst. An abdominal CT showed a huge nonseptated cyst of 28 cm by 24.5 cm by 21 cm occupying the left side of the retroperitoneum extending from just below the left hemidiaphragm to the left pelvic brim. It extended beyond the midline pushing the left kidney and the intestine into the right half and the spleen superiorly (Figs 1 and 2). The origin of the cyst and the diagnosis was not certain. His Full blood count showed PCV of 23% and leucocytosis with neutrophilia. The liver function test and the serum amylase were normal. Adrenal secretory tests were omitted. Differentials included urinoma, huge renal cyst, cystic lymphangioma, and pancreatic pseudocyst. Adrenal cyst was not included in our initial differentials.

The associated septicaemia was controlled with parenteral antibiotics. We relied on the history of a poorly managed urinary diversion to choose urinoma as our initial working diagnosis and attempted an extraperitoneal drainage of the cyst with a lumbar incision. Intra-operatively, we found a well capsulated cyst that easily stripped off the parietes rendering our initial diagnosis of urinoma
improbable. We decided to convert to a laparotomy. Laparotomy revealed a huge cyst that occupied the entire left side of the retroperitoneum displacing the left colon anterio-medially and pushing the small bowel and the left kidney into the right half of the abdomen (Fig. 3). It was not tense and was easily freed from adjacent parieties and the left colon. It lightly adhered to the superior pole of the left kidney but an adrenal origin was still not certain. It was unilocular and had a thin collapsible capsule. Some of the content was aspirated before cyst could be delivered out of the abdomen. It contained a uniformly straw colored fluid with floating crystal like substances and without debris. The diagnosis remained indefinite till the histology revealed an epithelial cyst of the adrenal cortex. He made an unremarkable recovery and has been followed up for more than a year without a recurrence.

**Discussion**

Adrenal cysts were first reported by Greaseless in 1670 [1]. They are rare and occur in 0.06–0.18% in autopsy series [2]. The incidence has increased with increasing use of abdominal imaging. They are more common in women and between 30 and 60 years of age [3]. Most small cysts are asymptomatic. Bigger cysts may become palpable as occurred in our patient or cause symptoms from pressure on adjoining structures. Some cysts present acutely after hemorrhage, infection, or rupture. Hormonal secretion, more common in solid adrenal tumors, has been documented in adrenal cysts. However, no documented giant adrenal cysts with sizes greater than 20 cm was secretory [4, 5]. About 7% of adrenal cysts are malignant with malignancy seen more in cysts bigger than 5 cm [6].

Although the guidelines to management of adrenal cysts may be straight forward, diagnosis may be complicated in giant cyst. Adrenal secretory tests are mandatory in solid adrenal tumors but may prove important in
adrenal cysts. This includes 24 h urinary metanephrines, 17-hydroxycorticosteroids, and 17-ketosteroids. This was omitted in the management of our patient because adrenal origin was not suspected. Ultrasonography, CT and MRI imaging are diagnostic investigations. The sensitivity of CT and MRI in adrenal lesions is between 85% and 95% and specificity between 95% and 100% [7–9]. This reduces significantly in adrenal cysts with diameters greater than 10 cm causing dilemma in diagnosis and management [10]. The pathognomonic findings on CT include thin nonenhancing wall and fluid density content. Intracystic hemorrhages may produce higher density values [11]. Cyst calcification may be present in 15% of patients. Because of size, the diagnosis in our patient was uncertain.

Retroperitoneal cysts not arising from major solid organs are rare and seen in 1/5750 to 1/250,000 of the population. They commonly pose diagnostic dilemma but adrenal origin is seldom included in the differentials [12, 13]. M Furihata et al. reviewed 27 adrenal cysts with sizes above 20 cm, and a correct preoperative diagnosis was made in only 14.8%. The others were diagnosed at laparotomy or after histology. Their preoperative diagnosis varied from hepatic cysts on the right side to pancreatic cysts on the left and ovarian cysts in females [14]. Our patient only had a diagnosis after histology.

Asymptomatic adrenal cysts less than 4 cm in size are usually managed conservatively when not secretory and not rapidly expanding. Surgical excision is recommended for cysts >5-cm diameter because of the risk of malignancy [15]. Access to excision includes laparotomy, posterior retroperitoneal approach, laparoscopic, and endoscopic retroperitoneal approach. Adrenalectomy should be considered when malignancy is suspected. Palliative drainage is recommended only in tense giant cysts. Adrenal cysts are classified as pseudocysts, endothelial cysts, parasitic cysts, or epithelial cysts. Of reported cysts above 20-cm diameter, 70.3% were pseudocysts and only one was epithelial.

In conclusion, huge retroperitoneal cysts that are not related to major organs may originate from the adrenal gland. When cross-sectional imaging is inconclusive, such cysts are best managed with exploration and complete excision that allows for histology.

Conflicts of Interest
None declared.

Authorship
IOO: contributed to patient management, literature search, and writing of manuscript. MDA: contributed to patient management and writing of manuscript. EAA: contributed to patient management and writing of manuscript.

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