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A giant hemorragic adrenal pseudocyst: contrast-enhanced examination (CEUS) and computed tomography (CT) features

V. CANTISANI, L. PETRAMALA[1], P. RICCI, A. PORFIRI, C. MARINELLI[1], G. PANZIRONI, A. CIARDI[2], G. DE TOMA[2], C. LETIZIA[1]

Department of Radiology, [1]Department of Internal Medicine and Medical Specialities, [2]Department of Surgery "P. Valdoni", School of Medicine, "Sapienza" University of Rome, Rome, Italy

Abstract. – INTRODUCTION: Adrenal pseudocysts are rare cystic masses that arise from the adrenal gland and which are usually non-functional and asymptomatic. We report a rare case of a giant hemorrhagic adrenal pseudocyst presenting with abdominal pain and we discussed the radiological features.

PRESENTATION OF CASE: A 75 year old man was admitted with acute abdominal pain post mild-trauma. Computed tomography (CT) of abdomen revealed a hemorrhagic mass measuring 18 cm located in the right suprarenal region, displacing the right kidney and liver. He subsequently underwent to contrast enhancement ultrasound (CEUS), which showed features suggestive for hemorrhagic adrenal pseudocyst. A complete endocrine working didn't show any hormonal hypersecretion. The patient underwent laparotomy and right adrenal mass was excised. Histological examination revealed giant hemorrhagic adrenal pseudocyst. The abdominal pain resolved after surgery.

CONCLUSIONS: to the best our knowledge, this is the first case studied with CEUS reported in the literature.

Key Words:
Adrenal disease, Contrast-enhanced examination, Radiological features.

Introduction

Adrenal cysts are infrequently observed, since less than 500 cases have been reported in Western literature. Adrenal cysts are histologically classified into four categories: endothelial cysts (45%), pseudocyst (39%) and infectious cysts (7%). True cysts are lined with endothelial or mesothelial cells[1].

Adrenal pseudocysts consist of a fibrous wall without a cellular lining[2]; their etiology remains uncertain, and hypothesis include hemorrhage into adrenal gland parenchyma secondary to trauma, cystic degeneration of a primary adrenal neoplasm and a vascular neoplasm or malformation[3-4]. Epithelial cysts have fluid density and in computed tomography (CT) images they appear as uni-multilocular, well defined round or oval, homogenous mass. On CT scan, cysts are characterized by thin non-enhancing walls and fluid attenuation with water or near density. On magnetic resonance imaging (MRI), cysts have the characteristics homogenous low T1/high T2 signal[5].

The ultrasonography scan (US) evaluation shows an anechoic mass with posterior signs of vascularization. However, occasionally cysts could appear hypo or hyper-echoic according to the content.

Preoperatively, complex adrenal pseudocyst may be different to distinguish from malignant lesions. In particular, it should be suspected in patients with great-size lesion, and mixed echos on US with heterogeneous texture[6-7].

We report a case of patient with a 18 cm right-sided suprarenal cysts mass which was studied with CT and contrast-enhanced ultrasound (CEUS). We specifically focus on the features of the CEUS, which to the best of our knowledge were not previously reported in the literature.

Case Report

A 75 years-old man was admitted to our Hospital, complaining acute abdominal pain that did not responded to analgesic therapy. He reported a previous mild trauma occurred two days before accompanied by dyspepsia and postural dizziness. The patient was receiving aspirin, beta-blocker and ACE-inhibitor medications for arterial hypertension and atrial fibrillation. The clinical examination didn't reveal abdominal tumefaction or tenderness. The patient had a pulse of 102 b/min, blood arterial pressure 140/90 mmHg,

Corresponding Author: Claudio Letizia, MD; e-mail: claudio.letizia@uniroma1.it

respirations 14/min. Electrocardiography demonstrated atrial fibrillation with no signs of ischemia. Chest X-ray did not demonstrate pulmonary congestion. Arterial blood gases were as follows: pH 7.41, PO_2 90, PCO_2 20.1 and HCO_3^- 19.2. Laboratory data were notable for anemia (Hb 9.1 g/dl) and high ESR (105 mm/h) (Table I). Gastrointestinal treat bleeding had been excluded by negative of occult blood stool, and secondary by pancolonscopy. The patients underwent to abdominal CT, which showed a right suprarenal mass (18 × 14 cm), appearing hypodense, mild peripheral enhancement of the thickened capsule while the most part of it was inhomogenously hypodense with some peripheral calcification (Figure 1). The day after, since the patient accused a worsening of the lumbar pain he was then submitted to a CEUS. The baseline ultrasonography evaluation showed a 18 cm of maximum diameter mass with regular margins. This lesion was locate in the right kidney, without infiltrative effect. The mass showed inhomogenous iso-hypoechogenecity, within sclerotic and fluid-particle areas. After contrast injection CEUS evaluation showed that the lesion did not enhance during the whole examination but only some tiny vessels disposed peripherally were encountered. CEUS showed features suggestive for "hemorrhagic adrenal pseudocyst" (Figure 2). Hormonal levels, including 24-hours urinary metanephrines, aldosterone, free cortisol excretion and plasma levels of cortisol, ACTH, DEA-S, Δ4-androstenedione, plasma renin activity (PRA) and aldosterone were all within usual limits (Table I). Based on these findings, we diagnosed a non-functional right adrenal hemorrhagic mass. Elective surgery was planned to prevent further pain and for the high size of the adrenal lesion (an adrenal mass ≥ 6 cm carries an increased risk of adrenal malignancy). Based on a multidisciplinary approach, we decided on open surgery to remove the right adrenal mass. Our surgery colleagues performed exploratory laparatomy via a midline celiotomy under general anesthesia in elective conditions with appropriate therapy. Exploration of the abdomen revealed a 18 × 14 cm dark lesion originating from the right retro-peritoneal region. The lesion was adherent to the right adrenal gland and received its blood supply from all adrenal arteries. The adrenal mass was completely excised with a right adrenalectomy (Figure 3). Histopathological examination of the mass showed an oval massive hemorrhagic adrenal lesion, measuring 19×14×12 cm of diameter, containing mostly coagulated blood with fibrosclerotic wall. A diagnosis of an hemorrhagic adrenal pseudocyst was made. The patient's post-operative course was uneventful and he was discharged 9 days after the operation. Follow-up demonstrated that the patient remains well and asymptomatic 3 months after surgery, with good laboratory data (Table I).

Table I. Laboratory analysis before and after adrenalectomy.

	At diagnosis	After surgery (3 months)	Normal value
Urinary free cortisol	50	47.7	26-134 (μg/24h)
Plasma cortisol (8:00 am)	24	20	5.25 (μg/dl)
Plasma ACTH (8:00 am)	44	31	10-50 (pg/ml)
Plasma cortisol 8:00 am after DXM (1 mg/dl)	2	_	< 3 (μg/dl)
Serum DEA-S	117	108	33-249 (μg/dl)
Serum testosterone	3.89	4.1	2.8-8 (ng/ml)
Urinary metanephrine	48	40	20-320 (μg/24h)
Urinary aldosterone	19.5	13.60	2.8-30 (μg/24h)
Serum Δ4 Androstenedione	4.5	3.6	1.2-3.1 ng/ml
Plasma renin activity	0.8	0.6	0.2-2.7 (ng/ml/h)
Plasma aldosterone	172.3	163.8	7.5-300 (pg/ml)
Blood fasting glucose	98	76	73-109 (mg/dl)
Serum creatinemia	0.92	0.93	0.70-1.2 (mg/dl)
Serum ferritinemia	863	278	22-322 (mg(dl)
Serum fibrinogen	5.84	3.74	1.5-4 (g/L)
Red blood cells (RBC)	3.79 × 106	4.90	4.30-5.9 (μL)
White Blood cells (WBC)	10.1 × 103	6.92	4.40-11.30 (μL)
Hemoglobin (Hb)	9.1	11.3	13.50-16.50 (g/dl)
Platelets (PLT)	95 × 103	85	150-450 (μL)
Erythrocyte Sedimentation Rate (ESR)	105	48	< 20 (mm/H)

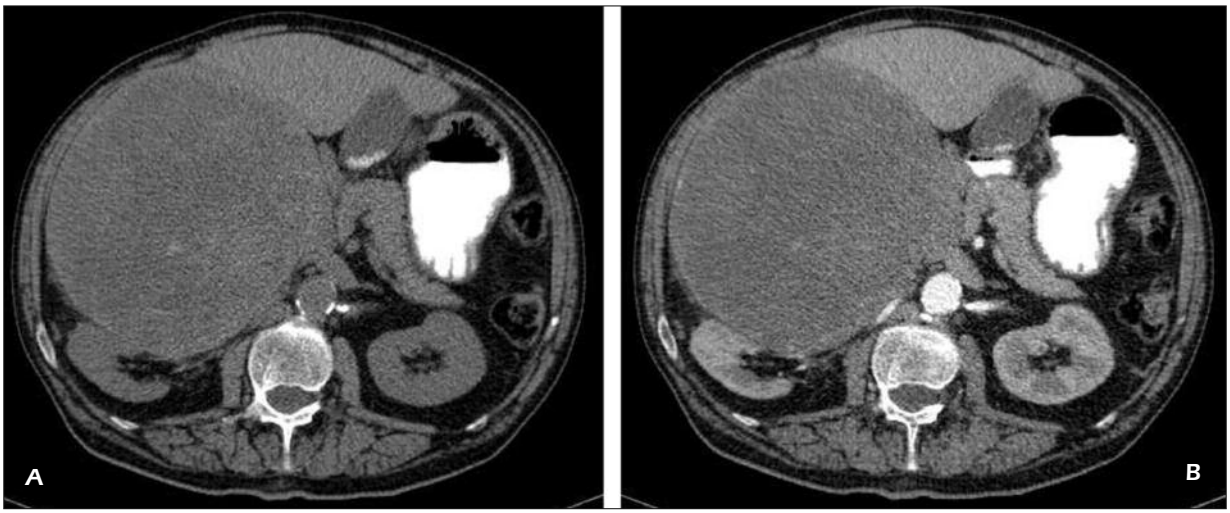

Figure 1. Computer tomography scan showing a large adrenal hemorraghic lesion ***(A)***, which after contrast administration shows only mild peripheral enhancement ***(B)***.

Discussion

Adrenal pseudocysts are rare and account for 32% to 80% of all adrenal cysts[8-9]. The majority of adrenal pseudocysts are found because of their size-related symptoms[10-11].

Patients can present with acute abdominal findings if intracystic hemorrhage or rupture occurs[12]. Large adrenal pseudocysts are prone to complications such as intracystic hemorrhage, infection and rupture. In particular, the incidence of adrenal hemorrhagic pseudocyst is very low. Less than 100 hemorrhagic pseudocysts have been reported, but only few of these had such giant proportions[13]. The true origin of adrenal pseudocyst remains a mystery. One theory suggests that these lesions result from an intra-adrenal hemorrhage caused by trauma, a sepsis event or some other form of shock. Another theory suggests that these lesions are true cysts that have lost their cellular living because of the inflammation and bleeding within the cyst. The etiology of our patient's pseudocyst seem to be similar to first theory. In fact, the patient was admitted in our hospital with post-trauma abdominal pain. The adrenal glands are a frequently site of pathologic processes, and the majority of adrenal lesion are incidentally detected at imaging[14]. Occasionally, they are found because of their size-related symptoms. US, CT, MRI and nuclear medicine are the imaging modalities usually

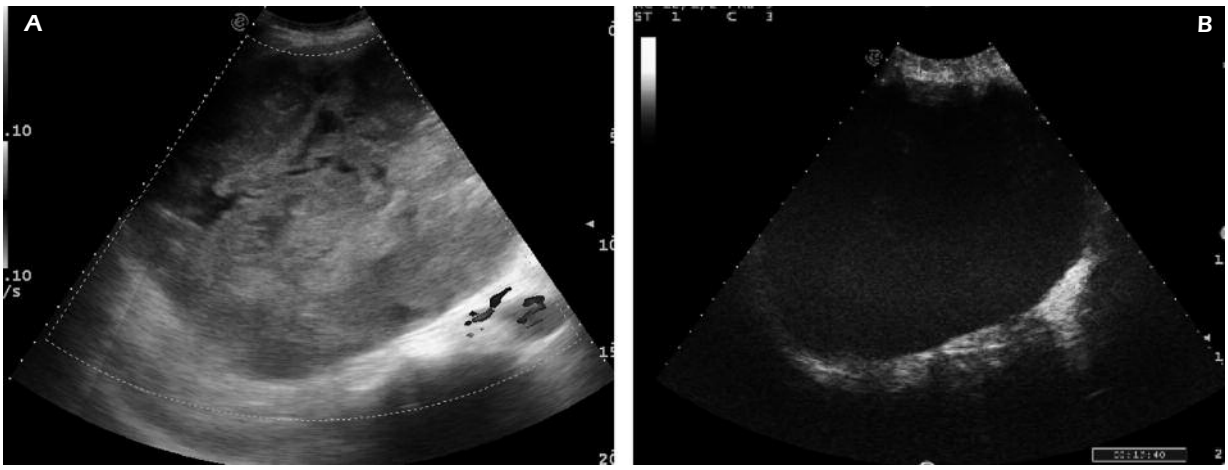

Figure 2. ***A,*** Color-Doppler ultrasonography (US) shows the presence of large inhomogenous adrenal lesion without any clear vascular signs. ***B,*** At CEUS the lesion does not show any signs of enhancement.

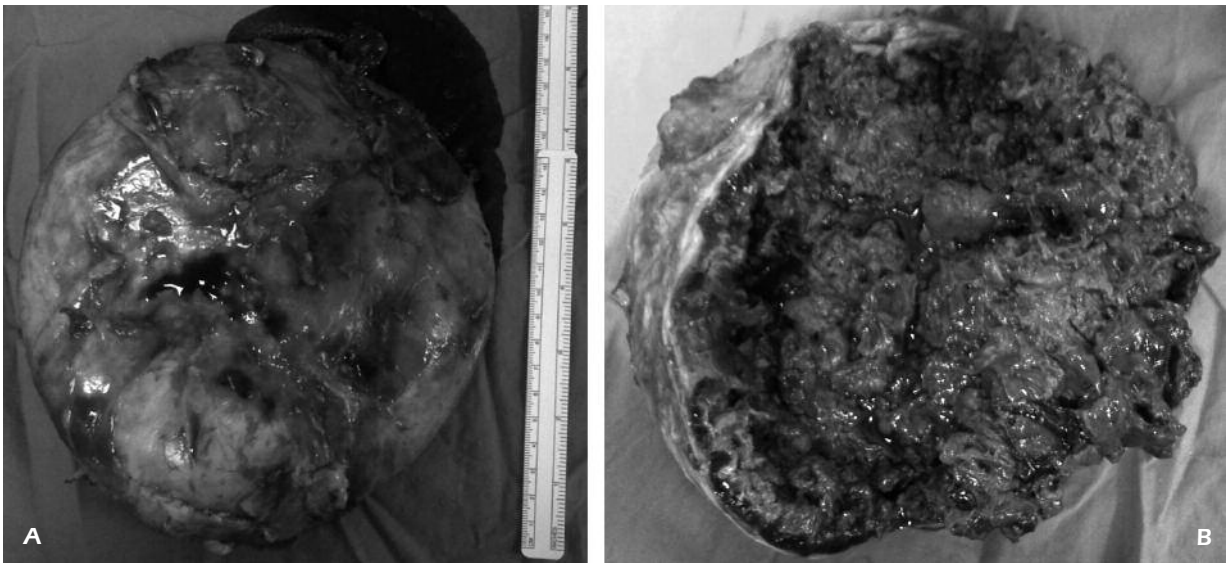

Figure 3. A, A 19X14X12 cm adrenal mass with massive hemorrhage. ***B,*** Containing centrally coagulated blood with fibrosclerotic wall.

encountered in the daily work-up of the patients in order to obtain a better characterization of lesions to start soon the prompt treatment. Different studies have shown that benign and malignant adrenal masses can be differentiated at CT with a sensitivity of 85-100% and a specify of 95-100%[15].

MRI is also an accurate method for studying adrenal mass and in particular is important in the case of indeterminate adrenal mass after CT[15]. These techniques, although represent gold standard in the adrenal lesion diagnosis, present some limits: high cost, patients with claustrophobia or refusing to the examination, possible adverse reaction of contrast medium , still limited availability on the territory for MRI, while adverse reaction, scan radiation exposure for CT scan.

As a consequence of these limitations, US plays an increasingly important role as first-line examination for the adrenal masses evaluation. However, the sensitivity of US for the detection of adrenal lesions varies from 66.7%[10] to more than 90% of all-even small-adrenal gland lesion[16]. Recently, CEUS which is nowadays a well-established imaging modality indifferent fields such as liver[17], vascular imaging[18], so on was also used in adrenal pathology evaluation[15]. However, the results showed by the two studies present in literature were contradictory. In fact, Friedrich- Rust et al[15] reported that CEUS was able to differentiate adenomas and non-adenomas lesions with a sensitivity comparable to CT and MRI; contrast-enhanced sonography, however, is not reliable for differentiating the various histopathologic non-adenomatous lesions from one another. Conversely, Dietrich et al[19], recently showed that CEUS may allow to visualize the vascularization and perfusion even in small adrenal masses, but it cannot be applied to differentiate reliably between malignant and benign lesions. However, they did studied only solid lesions. We report the CEUS features of a non-completely determinate case at CT of hemorrhagic adrenal pseudocyst. A complex pseudocyst may be difficult to differentiate from metastasis and other necrotic tumor or abscess[5]. Underlying, carcinoma should be suspected in patients with a high, erythrocyte sedimentation rate (ESR), mixed echoes, an US and stippled calcification[20]. In fact, in our patient CT scan showed an heterogeneous, mildly hyperdense extensive lesion before contrast agent administration. The lesion after contrast medium administration showed enhancement of the capsule, with increase of the Hounsfield Unit (HU). The features were interpreted as suggestive of probably aggressive adrenal hemorrhagic lesion; because of the lack of any story of trauma when the examination was performed, and because of the size of the lesion, the patient was submitted then to surgery. It should be taken in account that an exact diagnosis in clinically important because an adrenal cyst ≥ 6 cm carries an increased risk of malignancy. In fact, the incidence of malignancy in adrenal cystic lesions is approximately 7%.

Conclusions

Taking in account the revealed previous mild traumatic event, and combined with the CT features, our patient was studied with CEUS. CEUS revealed that the lesion was not characterized by any enhancement during the whole examination. Therefore, the suspicion of hemorrhagic adrenal pseudocyst arised and the patient was operated. This diagnosis was confirmed at histological examination. Radiologist should be aware of the US and CT feature of large adrenal lesions with not characteristic behavior of malignant ones. In this respect, CEUS may play an important and useful role to better characterize this kind of lesions.

Conflict of Interest

The Authors declare that they have no conflict of interests.

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