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Case Report

GIANT ADRENAL PSEUDOCYST PRESENTING WITH HYPERTENSION: LAPAROSCOPIC MANAGEMENT

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Abstract
Adrenal pseudocyst is a rare clinical entity. When present, they are non-functioning and asymptomatic. Most adrenal cysts are less than 10 cm in diameter and usually discovered incidentally. However, they can grow to a much larger size and cause size-related symptoms. . We report a pseudocyst of the left adrenal gland measuring 21 cm in diameter in a 49-year-old woman. She presented with left flank fullness, hypertension, and back pain over a 6 month period. Abdominal ultrasound and CT imaging revealed a large cystic mass in the left adrenal gland. The patient underwent trans-abdominal laparoscopic excision of the cyst following fluid aspiration. On histopathological examination, the cystic wall was made of pseudocapsule with no endothelial or epithelial lining. Blood pressure normalized 3 months after the surgery.
Keywords: adrenal pseudocyst, laparoscopic excision.

Introduction

Cystic adrenal neoplasms are rare lesions and are usually incidental finding.[1] Adrenal cysts are more common in women and typically diagnosed in late adulthood. They exhibit a varied spectrum of histologic changes and ranging from benign pseudocysts to malignant cystic neoplasms. Adrenal pseudocysts are the most common clinically recognized adrenal cysts. Usually cysts are non-functional and asymptomatic but can grow in size and become symptomatic due to pressure effect on adjacent organs, infection, rupture and haemorrhage.[2]

Case report

A 49-year-oldfemale came with complaints of pain and swelling in upper abdomen since 6 months. Pain was localized to the left side of the upper abdomen, dull aching in nature which aggravated on exertion and relieved with rest and analgesics. Swelling has increased gradually to the present size. Patient also gave history of decreased appetite, weight loss and altered bowel habit since 1 month. On clinical examination, she was found to have palpable solitary mass in the left hypochondrium extending into the left lumbar and epigastric region which was non-tender.

Initial evaluation with CT of the abdomen and pelvis showed large, well defined thin walled, non-enhancing fluid filled lesion measuring 21 cm x 15.4 cm x 16 cm in the left adrenal gland (Fig. 1a and 1b). Laboratory investigations were unremarkable, re-enforcing the diagnosis of a benign non-infective lesion in the left adrenal gland.

Treatment

Patient was placed in supine position. A large supra-renal cyst was seen in close relation with left suprarenal gland (Fig. 2). Cyst was punctured and reddish brown fluid was aspirated (Fig 3a and 3b). The whole capsule of pseudocyst was dissected off and left adrenal gland was preserved. Post procedure the patient recovered well and was discharged on the third post-operative day.

Discussion

In 1960, Griselius described the first case of an adrenal cyst in a 45-year-old man who died due to rupture of the cyst.[3]The incidence of adrenal cysts found in literature is 0.064%-0.18%in the general population. [4]The use of CT and MRI scans has led to an increase in the rates of detection of adrenal cysts. Adrenal cyst accounts for approximately 5.7% of incidentalaloma. A little more than 300 adrenal cysts have been reported in the literature.[5]Adrenal cysts are mostly found in the 3rd to 5th decades with male: female ratio of about 1:3, but may occur at any age, they have been reported also during the neonatal period of life.[4]Cysts are usually single in number, but can be bilateral in about 10% of cases. Cystic lesions of the adrenal gland have a varied histology, ranging from pseudocysts to malignant cystic neoplasms. Aetiologically, adrenal cystic lesions are classified as follows:[6]

1. non-neoplastic cysts (classification by Barron and Emanuel)
    a. Endothelial (vascular) cyst (with an incidence of 45%)
        i. Lymphangiomatous cyst
        ii. angiomatous cyst
    b. Pseudocyst (39%)
    c. Epithelial cyst (9%)
    d. Parasitic cyst 7% (generally echinococcal)
2. neoplastic cysts

Dermoid, mesothelial and lymphangiomatous cysts are also found in adrenal gland with very rare subtypes. Pseudocystsare the most common of cystic lesions, representing about 40% of all non-neoplastic cysts of the adrenal glands.[7]Only about 130 cases of pseudocysts have been reported till date. They may arise in the medulla or in the cortex and are defined as cystic formation, which lack a recognizable endothelial coverage and filled with fresh or altered fluid.

The exact pathogenesis of adrenal pseudocysts remains a mystery. They originate within the adrenal cortex or medulla following one of three formation mechanisms: hemorrhage of adrenal veins into the adrenal parenchyma, degeneration of a vascular neoplasm or malformation and cystic degeneration of a primary adrenal malignancy.[8] Sometime birth trauma or sepsis may play a role in formation. The association of adrenal pseudocyst with different pathologic conditions such as acute trauma, crush injuries, burns, shock, toxaemia of pregnancy, syphilis, leukaemia, and incompatible blood transfusions have been reported.[4]A small cavity with a scarred, fibrous lining may develop following above mentioned injury events that slowly grow over time. Alternate theory suggests that a true cyst may lose their endothelial lining because of the inflammation and bleeding within the cyst.

Due to its slow growth, majority of adrenal pseudocysts are non-functioning and asymptomatic and they become

symptomatic only when they attain a large dimension. Usually, patients present with the triad of Abeshouse; a dull back pain, a palpable mass and gastrointestinal complaints[9]. Since these lesions are located in the retroperitoneal space, they are usually not palpable until they reach large dimensions. Gastrointestinal symptoms include intermittent epigastric pain, nausea or vomiting following adhesion and compression and of neighboring abdominal viscera, such as the stomach, the small bowel, colon or kidney.

Adrenal cysts are associated with adrenal hypofunction, Cushing's syndrome or arterial hypertension has been also described. Cysts larger than 5 cm in diameter possess a 7% risk of malignancy.[10] We report a huge left adrenal gland pseudocyst associated with hypertension, which can be explained as Gold blatt phenomenon of hypertension, resulting from partial occlusion of a renal artery by external pressure.[11]

Ultrasonography can be the first choice as it is inexpensive and easily available. However, CT scanning is the gold standard; as it can identify small tumors with 100%sensitivity. In CT scan, most pseudocysts appear as well-demarcated unilocular or multilocular cystic lesions with complicated components such as septae, blood and soft-tissue components. Some of the useful radiological signs are the presence of Phantom organ sign (compressed adrenal gland), the Beak sign (a beak shaped edge of the adrenal gland) and the adrenal Limb-wall sign (the presence of thinned out, stretched limbs of the adrenal gland in the cyst wall)[12]. For better visualization of the intra-cystic components, MRI can be used. Angiographic studies are useful to identifying the parent organ by tracing the vascular supply to the lesion.

Usually full blood count, renal function, liver function tests, serum cortisol, renin, aldosterone, calcium and urinary catecholamines, 5-HIAA and metanephrines can be done to exclude other masses originating from the adrenal gland. Scintigram with the use of I^{131} MIBG also may be required.[2]

Rare association of adrenal gland cysts with a variety of diseases including polycystic renal disease, Beckwith-Wiedemann syndrome, Klippel-Trenaunay-Weber syndrome, an abdominal aortic aneurysm, and pregnancy have been reported. Adrenal pseudocysts with unusual variants also have been reported, including those with intracystic fat and myelolipomatous metaplasia, features of a dermoid cyst, and those with ectopic thyroid tissue.[6]

Observation is indicated for asymptomatic small lesions (less than 4 cm),as most of them are benign. CT scan should be repeated at every 3 months after diagnosis for 18 months. Percutaneous aspiration alone is having high chance of recurrence so it can be used for smaller or recurrent cysts. Surgical excision of adrenal pseudocysts should be offered for larger lesions (i.e. greater than 5 cm), in the presence of symptoms, any suspicion of malignancy or if the adrenal cyst is hormonally active.

Surgical excision with preservation of adrenal gland is curative. Partial or complete adrenalectomy including the cyst should be performed in a difficult situation. Laparoscopic transabdominal aspiration and resection of the adrenal pseudocyst offers an alternative to conventional surgery which is less invasive, has equivalent or low complication rates, rapid functional recovery and cost effective. Laparoscopic approaches vary according to the location of the lesion, its size, patient morphology and most importantly the surgeon's experience. The rate of conversion is less than 3%. Following surgical approaches are described;[4]

- Transperitoneal

- Anterior
- Lateral
- Retroperitoneal
  - Lateral
  - Posterior
- Thoracoscopic transdiaphragmatic

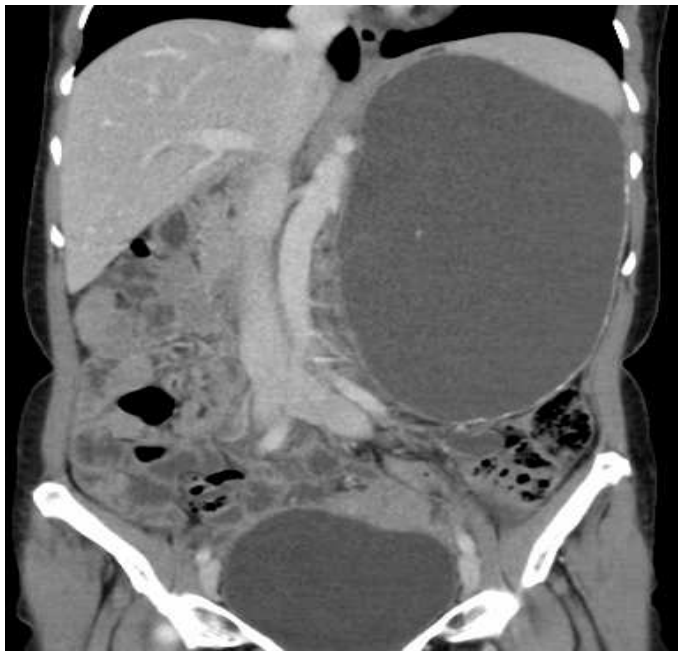

Figure. 1(a) CT scan showing giant left adrenal cystic lesion (Coronal section).

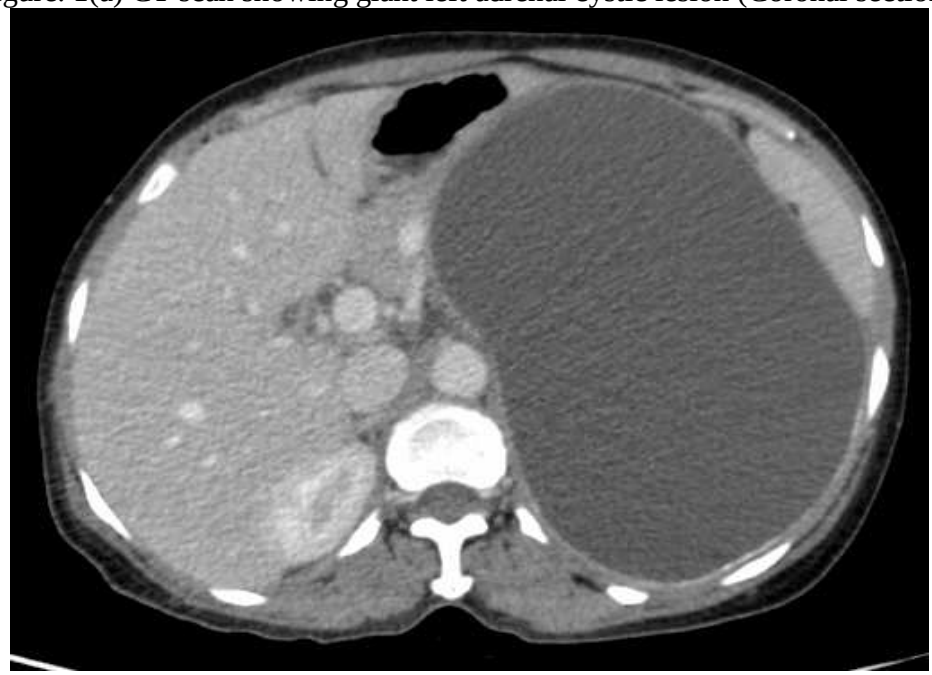

Figure. 1(b) CT scan showing giant left adrenal cystic lesion (Axial section).

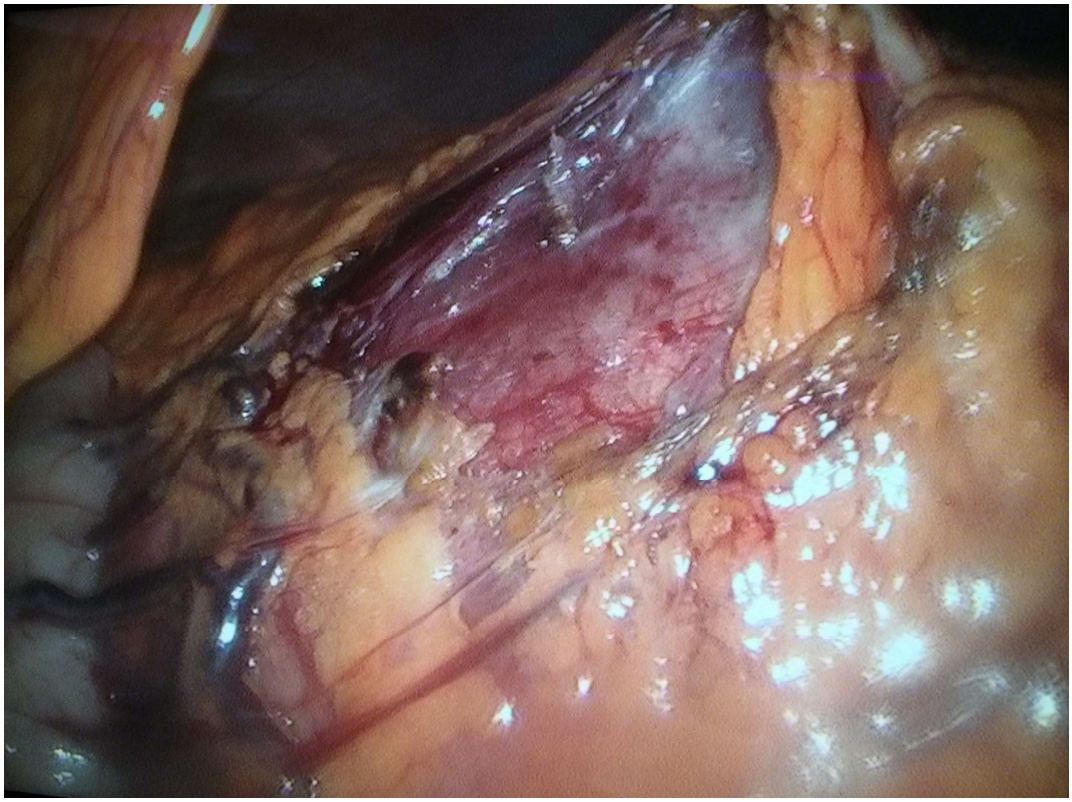

Figure. 2 Laparoscopic demonstration of left adrenal cystic lesion.

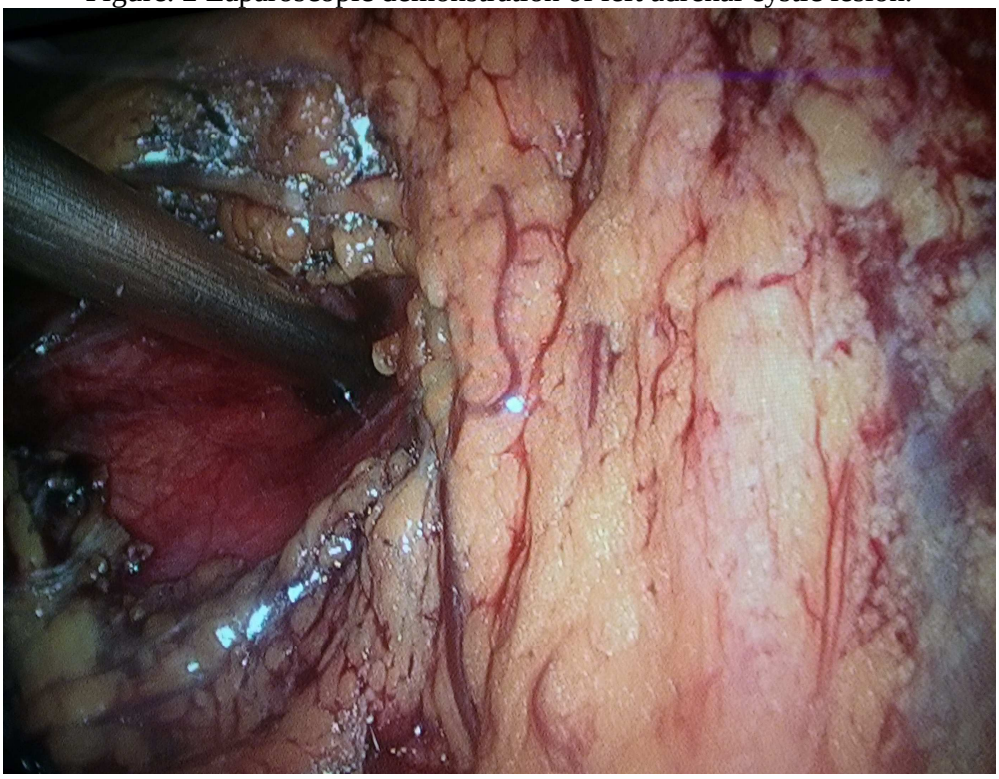

Figure. 3(a) Laparoscopic aspiration of the cyst.

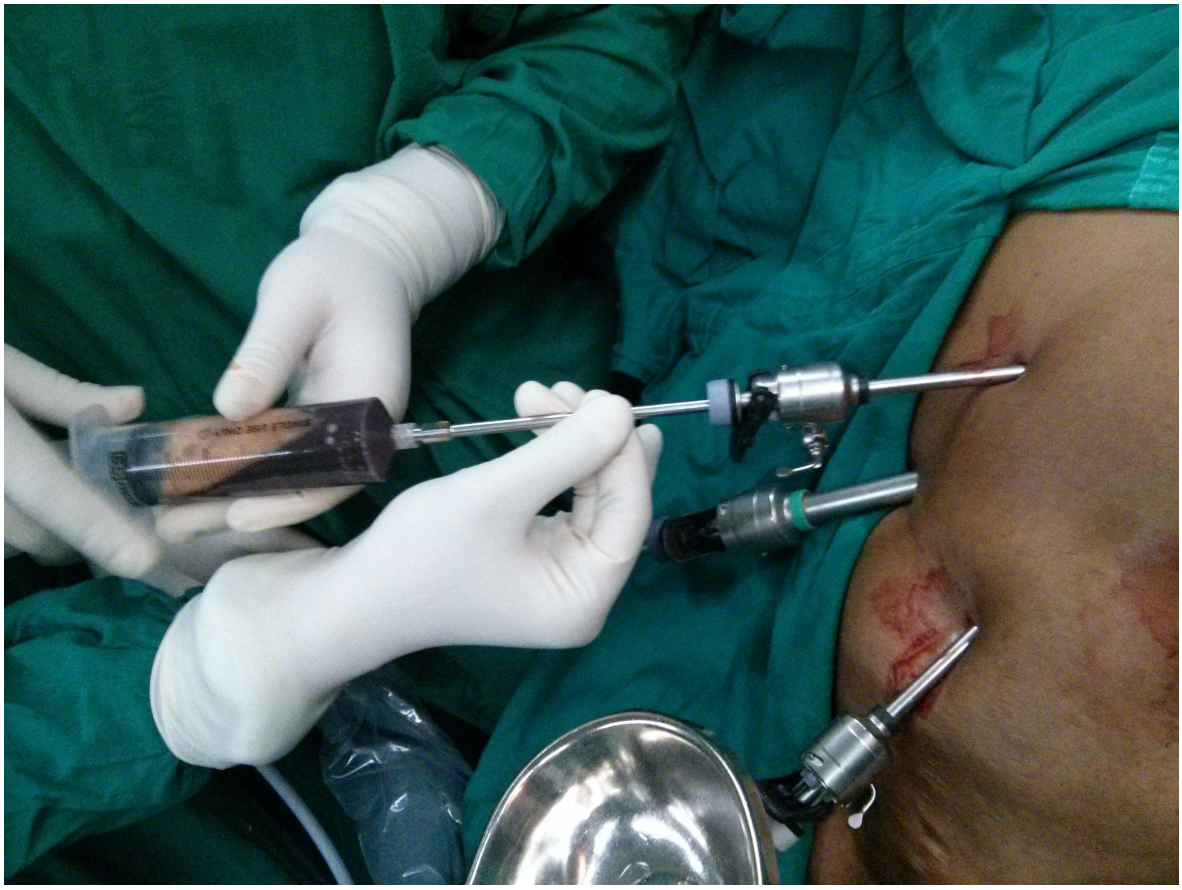

Figure. 3(b) Reddish brown fluid being aspirated from cyst.

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