https://doi.org/10.4038/sjdem.v14i1.7491

A case of Adrenal Endothelial Cyst in a young female

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Abstract

Adrenal cysts are rare, often asymptomatic and non-functional. Here, we discuss the case of a 24-year-old young female who had been experiencing unexplained abdominal heaviness and pain for 6 months' duration. An almost 5 cm anechoic lesion without a septum was detected by abdominal sonography across the left abdominal region. A 4.8x4.2x4.8cm suprarenal cyst was discovered on abdominal computed tomography (CT) scan displacing the left kidney inferiorly and anteriorly. There was no evidence of hormonal hypersecretion clinically or biochemically. She underwent successful laparoscopic left adrenalectomy, revealing a histopathological diagnosis of an endothelial cyst without any features of malignancy. The postoperative course was uneventful without recurrence during the period of follow-up. Current literature on the management of adrenal cysts is sparse although, personalized management with multidisciplinary team involvement is paramount important.

Keywords: Adrenal, Epithelial cyst, Benign, Adrenalectomy

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Introduction

Cystic adrenal lesions (CALs), which first described by Greiselius in1670 [1], represent a rare entity with heterogeneity in etiology and clinical manifestations. Adrenal cysts are asymptomatic and rather infrequent. Their frequency ranges from 0.064 to 0.18% in autopsy studies [1] and from 5 to 6% in clinical data [3, 4]. Smaller CALs are often detected incidentally during US, CT scan or MRI evaluations, whereas larger cysts may present with pain, gastrointestinal symptoms or palpable masses and account for 4-22% of all adrenal incidentalomas (3, 16). CALs are often unilateral, even if bilateral lesions have been found in 8 to 15% of cases [3]; they are more common from the 3rd to the 5th decade in which more common in females, with a ratio of 2:1 to 3:1, and may range from few millimeters to 50 or more cm in diameter [2,4,11,12]. Neri and Nance examined 613 adrenal cysts in 1999 and found that the majority of them ranged in size from 5.1 to 10 cma mean size [2] of around 9.6 cm. Esquivel and Grabstald reported the 50 cm-diameter cyst that was the largest one reported to date [6]. Bilateral lesions are found in 8-15% of cases [3], whereas CALs are usually unilateral [2,4,11,12].

Only a small number of case-series have been documented in the literature due to the rarity of these lesions [2 -7]. After the first histological classification scheme (based on autoptic examinations) proposed in 1959 by Abeshouse [8] and revised by Foster in 1966 [9], CALs have been divided into four categories: endothelial cysts, epithelial cysts, pseudocysts, and parasitic cysts. In 1999, Neri and Nance identified pseudocysts as the most common subtype in their extensive review on surgically treated cases Endothelial cysts can be further subdivided into lymphangiomatous and angiomatous varieties.

For this heterogeneity in clinical aspects, many controversial still exist about their management [2]. The treatment of adrenal cysts is based on their size, symptoms, and functional status. Surgical intervention

is advised when adrenal cysts are greater than 5 cm, symptomatic or hormonally active, or when malignancy is suspected. Laparoscopic Adrenalectomy is recognized as a safe and minimally invasive therapeutic strategy for CALs requiring intervention. Here we describe a case of a young female with a cystic adrenal lesion with a brief review of available literature.

Case Description

A 24-year-old female presented with a history of abdominal heaviness and pain for six months duration with an unaltered appetite, bowel habits, and a stable weight. Her systemic examination was unremarkable including the hemodynamic parameters. Initial evaluation with a chest radiograph revealed a left-sided diaphragmatic elevation. An anechoic lesion covering the left abdominal region was detected in abdominal sonography which was confirmed by abdominal CT, revealing a well-defined thick-walled cystic lesion measuring 4.8cm (AP) x 4.2cm (Lat) x 4.8cm with minimal calcifications and without contrast enhancement originating from the left adrenal gland suggestive of an adrenal cyst (Figure 1A and 1B). The biochemical evaluation was unremarkable with no evidence of hormonal hypersecretion (Table 1). Following a multi-disciplinary team discussion, she underwent laparoscopic left adrenalectomy successfully through a retroperitoneal approach. The post-operative period was uneventful and she did not show any recurrence to date. The lesion was devoid of any malignant features and measured 15 x 13 x 10 mm in size (Figure 2). The fibrous wall of the lesion which devoid of any epithelial lining, and has a 3mm thickness and calcifications suggestive of an endothelial cyst of the adrenal gland according to the histological classification (Figure 3).

Discussion

CALs are uncommon tumours of the adrenal gland [2-6]. Due to the advancements in and increased use of imaging studies, their prevalence appears to be rising over the past several decades [7-11]. Adrenal cysts often have no symptoms. The three most noticeable clinical characteristics are a palpable mass, gastrointestinal complaints, and a dull ache in the retroperitoneum. Additionally, intra-cystic bleeding, hypertension, and infections have all been documented [7–9]. The pressure that the cyst places on the renal tissue and its vascular system is most likely the mechanism through which adrenal cyst-associated hypertension develops, which resolves following the surgical excision [9].

Table 1: Biochemical test

Test type	Results
Serum electrolites	Na+ - 137 mEq/L K+ - 4.2 mEq/L
1mg Oral dexamethasone test	Less than 27.6 nmol/liter
24 hours urine metanephrines	0.49 mg/24 hours

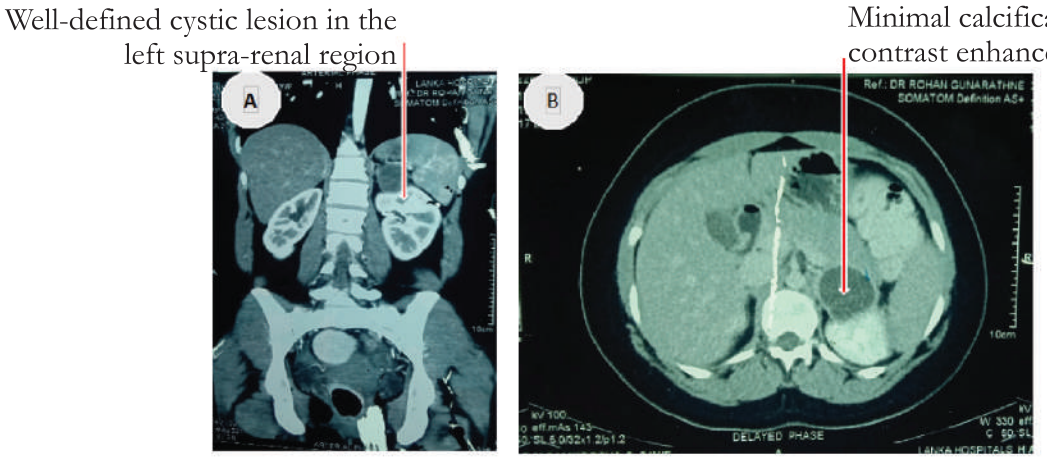

Figure 1: CT scan of the abdomen

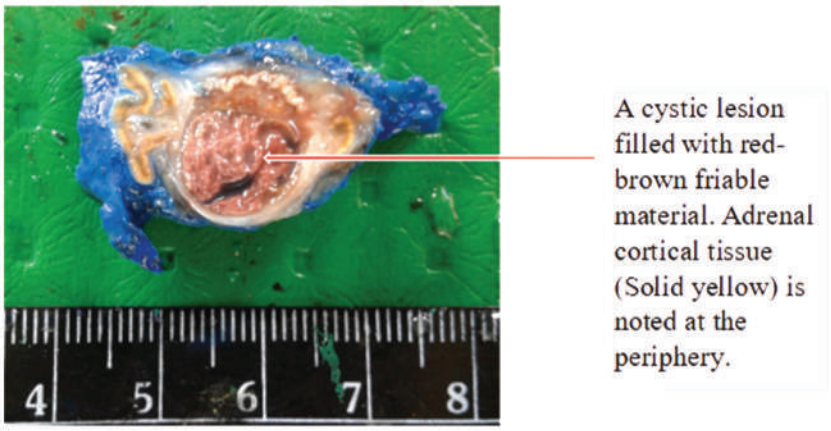

Figure 2: Macroscopy

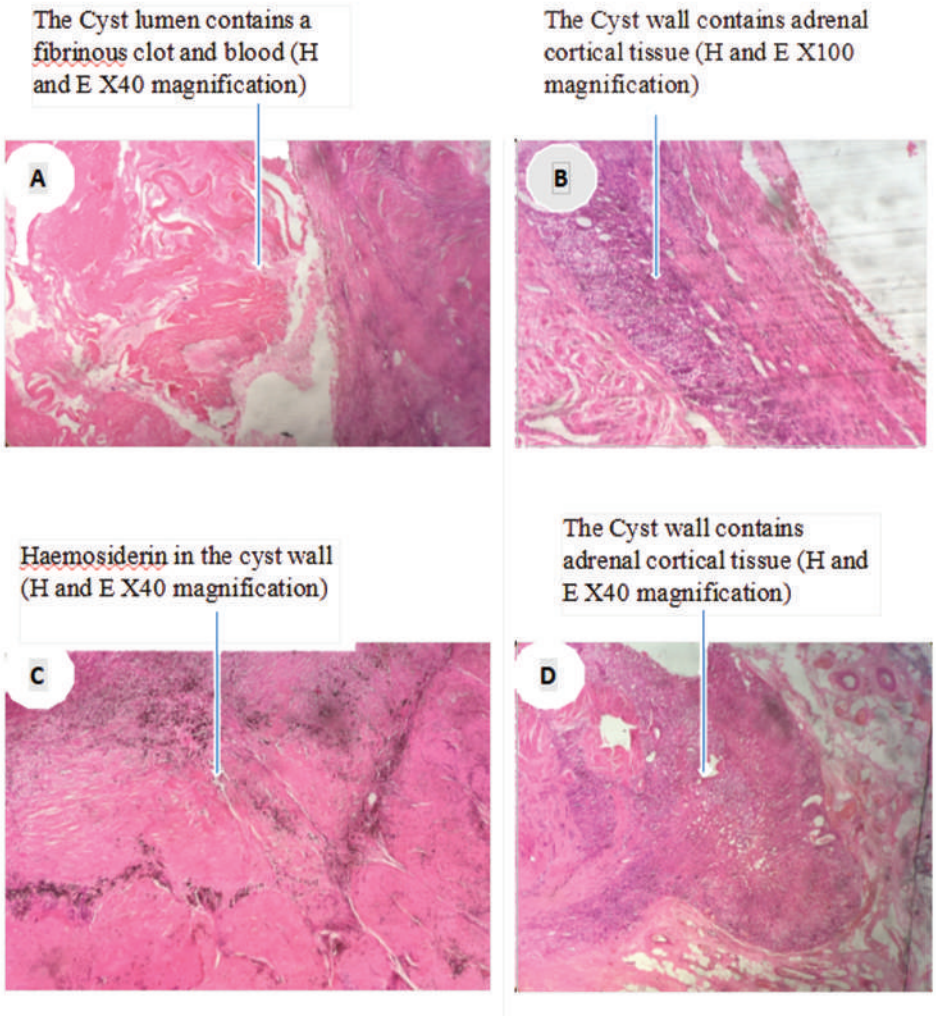

Figure 3: Histopathological characteristics of the endothelial cyst of the adrenal gland

Cystic pheochromocytoma is a different clinical entity resulting in adrenal cyst-associated hypertension [10]. However, a subclinical functional adrenal tumour is not unusual and cystic pheochromocytoma may not exhibit the typical symptoms, a 24-hour urine fractionated metanephrine level should be acquired prior to surgery to rule out this possibility.

As in the case of solid adrenal lesions, cystic adrenal lesions should also follow the common evaluation protocol, in order to exclude underlying hormonal activity and precise imaging technology to exclude malignancy with CT or MRI or functional studies (MIBG-scan for suspected pheochromocytoma). Due to its capability of providing coronal and sagittal projections that help identify the origin of the

retroperitoneal cyst, particularly for a larger cyst, MRI has benefits over CT and sonography [11]. As it can distinguish between metastases, simple cysts, functional tumours, and intra-adrenal bleeding, MRI is also claimed to have higher specificity [11]. An adrenal cyst should appear as an avascular lesion on an adrenal angiography [12]. The nature of an adrenal cyst may be determined by aspiration, which has a sensitivity and specificity of 85% and 100% in excluding malignancy [13]. The index patient had a clear diagnosis of a left adrenal cyst without any malignant features or endocrine activity although she has unresolved symptoms of abdominal origin for which she underwent left side adrenalectomy.

Management is often based on the presence of hormonal hypersecretion or features of malignancy. Percutaneous cyst aspiration has been recommended as an alternative to surgery if the cyst is not hormonally active and there is no suspicion of malignancy [10] and even for obtaining cytology [14]. However, in our opinion, this technique should only be used in situations where a patient's clinical condition makes an elevated risk of adverse surgical events unavoidable.

Adrenal cysts that are small, asymptomatic, or dysfunctional can be monitored clinically without treatment. However, surgery is advised when they are beyond >5 cm diameter, symptomatic or functional, or when malignancy is suspected. Open surgery with cyst enucleation, en bloc adrenalectomy, or laparoscopic surgery with cyst decortication and partial or complete adrenalectomy are the surgical options available [3]. The most preferred method is a straightforward laparoscopic enucleation of the cyst with preservation of the kidney and adrenal gland [16]. However, the procedure should be performed in accordance with the rules of surgical oncology for possibly malignant or complicated adrenal cysts. Laparoscopic therapy with en bloc excision via a lateral transperitoneal technique is safe and practicable while keeping the advantages of low invasiveness, according to two studies from Taiwan. Numerous minimally invasive surgical approaches have been suggested for adrenalectomy [14,15]. When the liver, spleen, and pancreatic tail can be adequately mobilized on both the right and left sides of the abdomen, the lateral transabdominal approach, first proposed by Gagner in 1992 [14], offers the advantages of optimal displacement of vessels and visceral structures, easy identification of anatomic landmarks, and a large surgical field [14,15]. Open surgery should only be used in complex instances where mini-invasive techniques may fail due to increased cyst size, pre-operative signs of enormous compression, or infiltration of adjacent tissues. Complete adrenalectomy, preventing fragmentation of adrenal tissue, and cyst rupture with subsequent fluid leakage are all deemed essential [7,10]. The female lead underwent laparoscopic left adrenalectomy successfully and is currently being followed up with no additional complications or recurrence to date.

Conclusions

With the increased use of imaging modalities for patient evaluation, it is anticipated that adrenal cysts will be identified more frequently by accident. When the conditions are satisfied, surgical removal of these cysts is a viable technique of therapy.

Consent

Oral consent was given by the patient for this case report.

Conflicts of interests

The authors have no conflict of interest to declare.

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