CASE REPORT

Giant adrenal gland pseudo-cyst: a case report with literature review

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Abstract

Adrenal Gland Cysts are rare among all the pathologic cysts that occur in human beings; the pseudo-cyst variety even rarer. Adrenal pseudo-cysts are asymptomatic, non-functional, small, and incidentally discovered disease entities. Their clinical presentation is usually the result of their mass effects. Thanks to the advanced diagnostic technology, more such cases are being discovered timely and managed surgically, before life-threatening complications occur. Open surgical treatment remains the treatment of choice for giant cysts.

Keywords: Adrenal Gland Cyst, Pseudo-, Lumbar region.

DOI: 10.47391/JPMA.6896

Submission completion date: 25-04-2022
Acceptance date: 16-12-2022

Introduction

Adrenal Gland Cyst (AGC) is a rare clinical condition.[1] It can present with vague upper abdominal pain, vomiting, and other uncommon symptoms like haemorrhage in the cyst cavity and signs of hypertension.[2] Histologically, there are four main types of adrenal cysts, of which endothelial type is the most common one, followed by pseudocysts.[3] Women are more likely to develop AGCs than men and are diagnosed at the mean age of 39.5 years.[4] Computerised Tomography (CT) scan is the imaging modality of choice to identify the accurate location of the cyst and delineate its anatomy from the surrounding structures.[5] The optimum treatment of choice depends upon several factors, including size, symptoms, and endocrine function of the adrenal cyst. Laparoscopic enucleation of the cyst with preservation of adrenal gland is the most favoured approach,[6] while open surgery is highly reserved for malignant and giant AGC s(>5cm).[3] AGCs can be revealed by complications such as infection, haemorrhage and rupture.[6] We present a unique case of Giant Adrenal Gland Pseudo-cyst. After extensive literature search, it is believed that it is probably the first case of this size, type, and such presentation, to be reported in English scientific literature from Pakistan.

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Case Profile

A 55-year-old normotensive, non-diabetic and non-smoker man presented at the surgical outpatient clinic at Lahore General Hospital in October 2018 with a history of dull pain in the left lumbar region for the past two years. The patient ignored the symptoms and did not consult any clinician, owing to his uneducated background and meagre earnings. He described that the pain was gradual in onset, mild in intensity, radiating to the back, aggravated on walking, relieved on resting, and had no relevant associated factors. He stated to have lost appetite since the past two years and had lost five kilograms over one year. There was no significant past medical or surgical history. He was not taking any medications. On examination, his abdomen was soft and mildly tender in the left lumbar region. Upon deep palpation, a 10x10cm mass was palpable in the left lumbar region, not moving with respiration. Although it was felt to be spherical, it's lower limit was not reachable. The rest of the systemic examination was normal. Ultrasound (US) of the abdomen and pelvis showed a mass in the left hypochondriac region measuring 16x13x6cm. Contrast enhanced CT of the abdomen also confirmed "a well-defined, left retroperitoneal mass with peripheral enhancement and few enhancing septa" (Figure 1). Serum amylase, urinary vanillyl mandelic acid, and blood metanephrines and dopamine levels were normal. All baselines laboratory tests were also within the normal physiologic range.

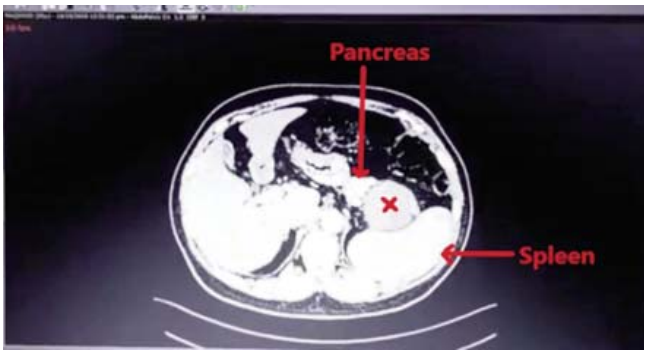

Figure-1: Axial slice of contrast enhanced CT of the abdomen showing structure, labelled "x" which is a giant adrenal gland/supra-renal cyst. A clear plane of fat is preserved between the pancreatic body and the adrenal cyst and between splenic hilum and the adrenal cyst.

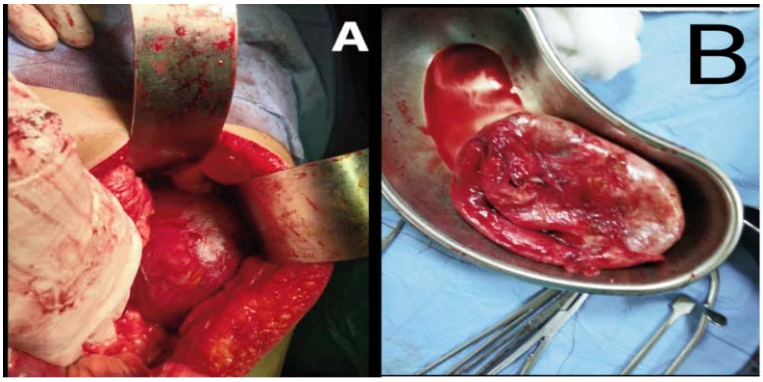

Figure-2: A. View of Left Adrenal Gland Cyst in-situ. B. Excised cyst

The senior consultants planned to explore the patient by open surgery owing to the very large size of the mass lesion. On exploratory laparotomy via a midline incision a cyst measuring 14x12cm was observed to arise from the left suprarenal area with no left adrenal gland seen separately (Figure 2). Pancreas and spleen were also not adherent to the cyst. The rest of the abdominal cavity and retroperitoneum were normal. The excised mass was sent for histopathology, which documented an adrenal pseudo-cyst.

The patient had an uneventful recovery in the ward, and was discharged on the fifth post-operative day. The patient was followed-up on monthly basis for six months, with repeated CT scans. He continued to live a normal routine life.

Discussion

Cysts of adrenal gland are rare; with a prevalence of 0.064% - 0.18% in autopsy studies and 5-6% in clinical series. However, due to the advancement in technology and better imaging techniques, more of these AGCs are discovered incidentally.[1] The pseudo-cyst variety of AGC is the largest group to be discovered intra-operatively and, hence, incidentally.[7] In our case, the AGC came to light due to the nature of symptoms and discomfort caused by it to the patient. It was a giant adrenal cyst. Contrary to their predisposition among female patients, our patient was a 55-year-old male.[4]

There are no specific risk factors for AGC; however, the vast majority of these cysts are unilateral and small.[3] The same pattern of unilaterality with a paradoxically giant size of the cyst was seen in our patient, in whom the cyst measured 14x12cm per-operatively. The various pathophysiologic mechanisms responsible for the development of pseudo-cyst include cystic degeneration of any solid or vascular adrenal neoplasm, vascular malformation, or rupture of adrenal vein into the gland.[8]

Patients with AGCs are mostly asymptomatic, while the symptomatic patients present with typical symptoms of vague pain in the lumbar region, gastrointestinal symptoms, and a palpable mass in the abdomen.[1] Our male patient also had these complaints. Some other non-specific symptoms include right or left hypochondriac pain, pain in epigastrium and back, and hyperemesis.[2] It should be noted that small-sized cysts are usually silent, while large cysts present with symptoms due to mass effect.[6] Moreover, these cysts could be complicated by infection, haemorrhage and hypertension.[1] AGCs could be malignant in approximately 7% of the cases.[6]

The differential diagnosis under consideration should include malignant adrenal tumours, splenic, hepatic and renal cysts, and pancreatic neoplastic cysts.[7]

It is recommended that any kind of suprarenal mass should be investigated for its possibility of malignancy. Functional masses can be well picked by their clinical manifestations and laboratory tests for pheochromocytoma, Cushing's syndrome and Conn's tumour.[8] Serum levels of amylase, blood metanephrines and dopamine levels and urinary vanillyl-mandelic acid were also done, which were all within physiological ranges with normal baselines tests.

Imaging studies play a key role in the diagnosis of AGCs. Currently, CT scan is considered the gold standard investigation.[7] The sensitivity of Magnetic Resonance Imaging (MRI) is 100% in detecting AGCs, while that of CT scan and US is 80% and 66.7%, respectively.[3] US of the abdomen and pelvis were used as the initial investigation, followed by CT of the abdomen with intravenous contrast as the definitive investigation.

Histologically, four main varieties of AGCs have been studied: endothelial type (45%), which could either have angiomatous or lymaphangiomatous origin; pseudo-cyst (39%), which are often highly vascularised and haemorrhagic; epithelial cysts (9%), and parasitic cysts (7%), which are often hydatid.[3] The post-operative anatomopathological assessment of the specimen revealed pseudo-cyst variety of adrenal mass in our case.

The surgical management of adrenal cysts is dictated by many clinical factors, especially size, symptoms, endocrine function, and correctly differentiating it from other cysts. Clinical surveillance is indicated for small (<5cm), asymptomatic, and non-functioning cysts, while surgery is recommended for large, symptomatic, and

complicated cases.[6] Both laparotomy and laparoscopic surgery could be performed.[5] In case of parasitic cysts, antiseptic precautions, using hypertonic saline solution, are recommended for ruptured cyst during surgery, to avoid larva dissemination and anaphylactic shock.[5]

Laparoscopic approach should be avoided when the cysts are large or complicated, although it is most favoured for small cysts.[5, 6] Laparotomy can be performed by either intercostal lumbar access or transperitoneal anterior access.[2] In our patient, midline laparotomy was performed, peritoneum and retroperitoneum were explored, and the left suprarenal mass was excised and delivered intact. There was no need for larvicidal solution.

Conclusion

This case report highlights the clinical features of a rare variety of adrenal gland cyst, which is an incidental lesion in majority cases. Our patient was managed successfully with open approach owing to the giant size of the cyst. His intra-operative and post-operative course was free of complications.

AGCs of the giant variety occur rarely and there is no specific clinical or radiographic sign to pick them up. The bilateral lumbar regions of the abdomen are known to have cystic diseases of spleen, pancreas, liver, kidneys, and, yet uncommonly, adrenal gland. Hence, the histopathologic confirmation of the cyst is the only way to be certain about its diagnosis after surgical removal of the symptomatic cysts. The possibility of malignancy in lumbar masses should be considered as a rule, along with relevant history. It is recommended that the clinicians should include Giant Adrenal Gland Pseudo-cyst to the list of differential diagnoses in cases of adrenal gland mass during relevant work-up, irrespective of the patient's gender.

Ethical considerations: Written informed consent was taken from the patient. Detailed history was taken by the resident in charge of the bed and operating surgeon, after consent.

Disclaimer: None.

Conflict of Interest: The in-charge of Departmental Approval Committee, is one of the co-authors of this article.

Source of Funding: None.

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