CASE REPORT

A unique presentation of a complex haemorrhagic adrenal pseudocyst

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SUMMARY

A man aged 33 years presented with a 3-day history of left-sided testicular discomfort. Clinical examination revealed a large left-sided varicocele. He was referred for a testicular ultrasound, which demonstrated a moderate left-sided varicocele and an associated large suprarenal mass. An urgent (CT) scan was arranged which confirmed this, with associated compression of adjacent structures, including the left kidney and left renal vessels. The patient was admitted 1 week following the CT scan with features of sepsis. An inpatient adrenal MRI scan suggested the presence of a large left-sided haemorrhagic adrenal cyst. This was removed surgically through a left subcostal incision. Histology confirmed there was no malignancy. The patient was discharged and a follow-up CT scan at 6 months showed complete resolution of the cyst with no further testicular discomfort. This is the first known case of an adrenal cyst presenting with a varicocele.

BACKGROUND

Adrenal cysts are a rare find in clinical practice. Many are picked up incidentally, but presentations may include abdominal pain, back pain, sepsis and occasionally shock.^{1–4} There have been a range of sizes of cysts observed in medical literature. However, it has been reported that the size of the cyst does not directly correlate with the presence of symptoms.^{5–6} Many cysts may not cause significant symptoms or signs, despite being substantial in size. It must be noted, however, that the normal presenting features are often related to mass effect and underlying endocrine abnormalities are rare.^{7–8} Although often difficult to diagnose with certainty on radiological imaging, surgically excised cysts are often found to be benign.⁹ A literature review revealed that there are <100 documented cases of haemorrhagic adrenal cysts. A large, inoperable adrenal carcinoma has been shown to present with a varicocele, but there have been no documented incidents of adrenal cysts (haemorrhagic or otherwise) presenting in this way.¹⁰

CASE PRESENTATION

A previously fit and well man aged 33 years presented to his general practitioner (GP) with a 3-day history of left-sided testicular discomfort and scrotal swelling. An initial clinical examination by the patient's GP suggested a left-sided varicocele; a testicular ultrasound was arranged on an urgent basis. This was performed and supported the clinical diagnosis of a left-sided varicocele, which was moderate in size. Given that the left-sided

varicocele had presented suddenly, an ultrasound scan of the upper part of the urinary tract was carried out at the same session to investigate for underlying renal malignancy. The patient was found to have a large left-sided suprarenal hyperechoic mass. As a result of this unusual finding, follow-up imaging was arranged.

A CT scan was performed 4 days later confirming ~13.5×13 cm predominantly cystic lesion involving the left suprarenal region with mass effect. This was of indeterminate origin and follow-up MRI and Multidisciplinary Team (MDT) discussion were arranged. These investigations were arranged on an urgent outpatient basis, as an underlying malignant process could not be confidently ruled out.

One week following this CT scan and prior to any further imaging or MDT discussions, the patient presented to accident and emergency (A&E) with left-sided loin pain and fever of 24 hours duration, with a temperature of 38.3°C and a heart rate of 103 bpm. White cell count (WCC) was normal on admission ($8.0 \times 10^9/L$) with a C reactive protein (CRP) of 351 mg/L. Clinical examination of the patient identified a large, ill-defined fullness in the left flank indicative of the underlying suprarenal mass found on previous imaging. The patient was admitted to hospital and

Figure 1 Admission CT scan showing congestion of the left gonadal vein and haemorrhage into the adrenal pseudocyst.

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broad-spectrum intravenous antibiotics were started. A further inpatient abdominal CT scan was performed to establish any changes in the cystic lesion that may have accounted for the patient's A&E presentation. This showed perinephric and peria-adrenal fat stranding and haemorrhage into the suprarenal cyst, which was not present on the original CT scan. This CT also showed congestion of the left gonadal vein (figure 1).

As part of the patient's septic work up, repeated blood cultures were sent for analysis. Serial blood cultures were negative. The patient's WCC also remained normal throughout admission with a persistently raised CRP level.

A subsequent MRI of the adrenal glands was arranged based on the previous imaging. This was reported as showing a large left-sided suprarenal cyst with irregular internal septations, mildly thickened walls and significant multifocal internal soft tissue components (figure 2). There were no convincing abnormal internal enhancements on the non-subtracted images. The cyst was thought to be left adrenal in origin and compatible with a complex haemorrhagic cyst.

Figure 2 T2-weighted MRI image of a large left-sided adrenal pseudocyst with associated compression of surrounding structures.

Figure 3 A 14x12x3 cm completely excised and collapsed adrenal pseudocyst.

DIFFERENTIAL DIAGNOSIS

During the patient's clinical journey, a number of differential diagnoses were considered, including but not limited to: adrenal myolipoma, cystic adrenocortical carcinoma, pheochromocytoma and complex renal cyst. Initial ultrasound scans, CTs and MRIs gave some clue to the underlying diagnosis but were not conclusive. In order to fully diagnose the lesion, histological evaluation was required following surgical excision.

TREATMENT

Owing to diagnostic uncertainty and ongoing symptoms secondary to the supraadrenal mass, the patient was counselled for surgical intervention. The patient was under the joint care of the urological and the general surgical team and underwent an open excision of large left adrenal cyst. This was performed through a left subcostal incision. A large retroperitoneal cyst was slowly extracted with the aid of mobilisation of the left colon and division of splenic and coeliac vein attachments. The cyst was decompressed at the end of the procedure to ease extraction from the surrounding structure. The cyst was eventually completely excised and sent for histological evaluation (figure 3).

OUTCOME AND FOLLOW-UP

The patient developed a postoperative fever, which was successfully treated with a short course of antibiotics. Histological analysis of the lesion revealed a collapsed, focally ruptured cyst 14x12x3 cm weighing 363 g. The cyst wall was up to 2 mm thick and lined by ragged haemorrhagic material. A small quantity of adrenal gland tissue was noted at one end on the outer surface. Microscopic evaluation confirmed this to be a pseudocyst that appears to have developed around an inflamed haematoma (figure 4). The patient was discharged 1 week after surgery. A follow-up CT was performed 6 months later and showed no evidence of recurrence. The patient was subsequently seen in the urology outpatient clinic and was found to have a normal testicular examination with clinical resolution of the left-sided varicocele.

DISCUSSION

Adrenal cysts can be categorised as endothelial, pseudocyst, epithelial and parasitic.¹¹ Over 600 cases of adrenal cysts have been documented worldwide in medical literature. Most of these are found incidentally on imaging.¹² The presentation of

Figure 4 Lining of the blood-filled pseudocyst adjacent to the adrenal gland (blood at bottom, inflamed granulation tissue at top). H&E stain. Original magnification x40.

symptomatic cysts varies considerably from abdominal fullness to hypovolaemic shock. Patients presenting with fever and tachycardia secondary to an underlying pseudocyst is a rare finding and often requires surgical intervention.¹³

An increasing incidence of adrenal cysts has been suggested by some authors, this is thought to be secondary to increasing use of radiological modalities for diagnostic purposes.⁸ Frequent use of ultrasound or CT scans on patients with non-specific symptoms or signs is now commonplace in medicine, and it can now be said that the vast majority of patients diagnosed with adrenal cysts occur as incidental findings on CT scans. Clinicians familiar with adrenal cysts often recommend surgical removal as opposed to percutaneous drainage due to the hypothesised high recurrence rates as found with other cystic lesions.¹⁴ There is often an element of diagnostic uncertainty as these lesions are not frequently encountered in clinical practice or seen routinely by reporting radiologists. Although one of the primary reasons for surgical removal of incidental cysts is to rule out malignancy,^{15–16} one review article suggests only 7% (39 of 515 cases) of excised cysts are proven to be malignant on histological analysis.⁹

The incidence of varicocele in the general population is estimated at 15%.¹⁷ A left-sided varicocele is associated with a small risk of malignant obstruction of the renal vein secondary to renal carcinoma.¹⁸ This is often discussed as a theoretical possibility in medical textbooks and relayed to medical students as an important consideration in patients presenting with a left-sided varicocele. However, the actual likelihood of finding a varicocele secondary to an obstructing renal lesion is low.

Up to 90% of all varicoceles are located in the left scrotum. There may be up to a 35% incidence of bilateral varicoceles on radiological assessment.¹⁹ However, very few are located on the right side due to the anatomical differences in left testicular drainage. The angle where the left testicular vein enters the renal vein is acute and therefore impedes venous return. There is also a lack of sufficient valves between these two veins. Another often-cited reason is the so-called 'nutcracker syndrome'—the superior mesenteric artery and the aorta compresses the renal vein reducing venous flow into the inferior vena cava.²⁰

Learning points

- ▶ Adrenal cysts are a rare find in clinical practice but may present in a number of different ways.
- ▶ Most often the presenting features are due to local mass effect. Endocrine abnormalities are rare.
- ▶ If a patient presents with a varicocele, the clinician should also be mindful that an obstructing renal, pancreatic or even adrenal lesion might be the cause.
- ▶ MRI imaging seems to be the best imaging modality.
- ▶ Histological evaluation for diagnostic purposes is strongly recommended, despite the low rate of malignancy in these masses.

Although NICE guidelines do not suggest referring all patients with a left-sided varicocele, patients with a suspicion of underlying malignancy or with concerning symptoms should be referred to an urologist for further evaluation. This also includes patients with a sudden presentation of a varicocele or patients where there is uncertainty about the nature of the swelling.²¹ The association between a left-sided varicocele and a corresponding left adrenal cyst has never been made previously and represents a unique finding in this case report.

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Contributors RJG gathered the relevant clinical information about the case with the aid of SS who was the consultant responsible for the patient's care. RB, who was also involved in the management of the patient, provided additional case-based information and report writing guidance. The report was constructed and written by RJG and SS. Significant additional editorial work was provided by both RB and DF.

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