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Case Series

Unilateral left spontaneous adrenal haemorrhage in a middle aged male. A case report

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ABSTRACT

Background: Adrenal haemorrhage is a rare condition, traditionally diagnosed at autopsy. Aetiology of adrenal haemorrhage can be traumatic or non-traumatic related. Rare cases of spontaneous adrenal haemorrhage, mostly from the right adrenal gland have been documented without any identifiable predisposing aetiologies.
Case presentation: A middle aged male with no comorbidities presented to the emergency department with non-specific signs and symptoms. There was no history of trauma and the haemoglobin was dropping. Initial investigations showed abdominal haematomas with no clear source. Taken to theatre, a diagnosis of a left spontaneous adrenal haemorrhage was made and left adrenalectomy was performed.
Conclusion: Adrenal haemorrhage is associated with severe complications such as adrenal insufficiency, sepsis and death. The following report will explore one such case, of a patient diagnosed with spontaneous left adrenal haemorrhage that was complicated by sepsis and death.

1. Introduction

This case report is compliant with the SCARE guidelines and has been reported in line with the SCARE checkline [15]. Adrenal haemorrhage (AH) is a rare condition, occurring in 0.14–1.1 % of cases at autopsy [6]. It is a serious condition with multiple aetiologies which may be trauma or non-trauma related [1,5]. The single vein which is responsible for the drainage of this highly vascular structure, the adrenals, makes it particularly sensitive to changes in venous pressure and subsequent haemorrhage [11]. AH can occur bilaterally or unilaterally. Bilateral AH can result in adrenal insufficiency which, when catastrophic, can lead to adrenal crisis and subsequent death [2]. Unilateral AH is more commonly secondary to trauma (blunt or penetrating) whereas bilateral AH is usually associated with systemic conditions causing adrenal vein spasm or thrombosis [2]. Idiopathic adrenal haemorrhage is rare and the mechanism is unclear [2]. Predisposing factors of non-traumatic AH include; underlying adrenal tumour, anticoagulation, heparin induced thrombocytopenia, disseminated intravascular coagulation, liver transplant, sepsis (commonly meningococcaemia, Waterhouse-Friderichsen syndrome), steroid use, thromboembolic disease, pregnancy, primary antiphospholipid syndrome and in neonates (sepsis, hypoxia, adrenal insufficiency, birth trauma, diabetic mothers and haemorrhagic disorders) [2–5]. Most patients presenting with unilateral AH have non-specific signs and symptoms, and rarely have adrenal insufficiency [1–3]. Diagnosis is usually incidental on imaging (ultrasound, computed tomography/magnetic resonance imaging). This report will document the presentation, management and outcome of a middle aged male presenting with non-specific signs and symptoms, diagnosed as idiopathic left unilateral adrenal haemorrhage.

2. Case presentation

A 57 year old, caucasion male, presented with a one week history of abdominal pains. It was localised to left flank and hypochondrium regions, and was associated with pleuritic chest pain. The pain was of spontaneous onset and there was no history of trauma. He had no known comorbidities and was not on any medication, including anticoagulants. He had no significant family history of ischemic heart disease or coagulopathies. His social history included a ten-pack-year history of smoking.

On examination, he was hypertensive with tachycardia, his vitals were, blood pressure of 176/90 millimetre of mercury, and heart rate of 112 millimetre of mercury. Was apyrexial to touch with

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temperature of 36.6° Celsius. General examination showed mild pallor. Cardiovascular system and respiratory systems were unremarkable. Abdominal examination, was soft, with tenderness over the left lower quadrant with localised guarding. Incidentally, he was noted to have an uncomplicated umbilical hernia. He had no renal angle tenderness. Direct rectal examination was normal. Other systems were unremarkable.

Electrocardiogram and urine dipsticks were normal. Initial blood results upon presentation showed renal dysfunction with a mild elevated C-reactive protein (Table 1). Haemoglobin, electrolytes, cardiac enzymes, pancreatic enzymes, international normalized ratio, liver function tests, were all within normal limits. Repeat bloods the next day, showed worsening renal dysfunction, haemoglobin dropped and had elevated white cell count and a C-reactive protein (Table 1).

Abdominal ultrasound showed free fluid in the abdomen and pelvis (Fig. 1), an apparent mass on the upper pole of the left kidney/suprarenal measuring 83 × 87 × 74 mm (Fig. 2), a left iliac fossa mass/collection 104 × 64 mm non vascular (Fig. 3). A non-contrast computed tomography (CT) of the abdomen showed left suprarenal hyperdense mass and retroperitoneal haematoma with perinephric fatty stranding (Fig. 4).

The patient continued to drop his haemoglobin and become more haemodynamically unstable and so, he was taken for an exploratory laparotomy. Intra-operative findings were retroperitoneal clots with no evidence of major vessel injury; a small ooze in the retroperitoneum of which the origin could not be identified and a normal spleen and liver. The patients abdominal cavity was packed as a damage control mechanism and vac dressing placed in order to facilitate a mandatory relook. Intra-operatively he was transfused 4 units of packed red cells and 2 units of Fresh Frozen Plasma (FFP) and was haemodynamically unstable. He was transferred to the Intensive Care Unit (ICU) intubated, for continued ventilatory support and resuscitation prior to his relook. He was started on broad spectrum antibiotics in ICU.

Post-operatively, a CT angiogram of chest and abdomen was requested. The angiogram of the chest revealed bilateral pleural effusions and a small pericardial effusion but no major vascular injury. The CT Abdomen showed improvement of the retroperitoneal haematoma, the adrenal gland haematoma was removed and there was no active bleeding (Fig. 5). He was then taken back to theatre for a relook laparotomy the next day. Intra-operatively a haematoma was noted in the region of the left kidney with venous bleeding from the left adrenal gland. A Left adrenalectomy was performed and he was transferred back to ICU.

Post adrenalectomy in ICU, he was started on intravenous hydrocortisone for suspected acute adrenal crisis. Serum adrenocorticotropic hormone (ACTH) was 0.4 pmol/L which was low and cortisol 753 nmol/L was high and likely due to the patient being post adrenalectomy. ACTH stimulation test, corticotropin releasing hormone or insulin tolerance test were not done. He had worsening renal dysfunction requiring dialysis. He had an elevated parathyroid hormone (PTH) of 18.1 pmol/L thought to possibly be secondary to his renal dysfunction. Urine metanephrines and vanillylmandelic acid were normal. His Initial blood cultures showed no growth, but his septic markers continued to worsen on empiric therapy. His repeat blood and urine cultures were positive for the gram negative bacilli, Acinetobacter baumannii, and was started on the appropriate therapy.

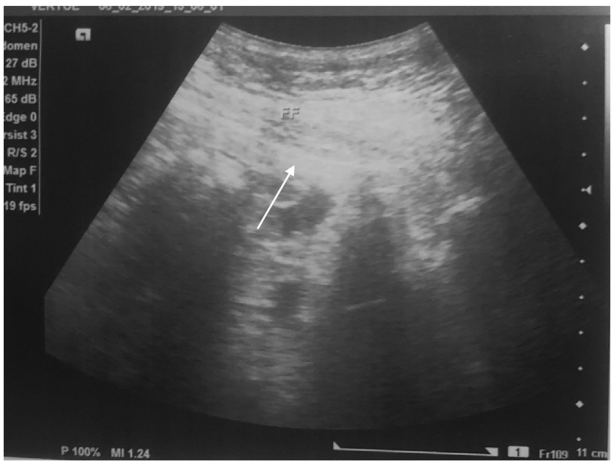

Fig. 1. Abdominal ultrasound showing free fluid in the abdomen and pelvis, white arrow.

Eighth day post theatre, while still receiving supportive management in ICU, the patient had a sudden cardiac arrest and subsequently demised despite attempts at resuscitation. He was assessed to have demised from sepsis associated with primary adrenal insufficiency. Post mortem was not done.

Histology of the left adrenal gland showed adrenal infarction and suppurative inflammation, with surrounding blood clots. There was no evidence of malignancy or any other specific pathology. Microscopic examination was negative for acid fast bacilli and fungi, and no granulomas were seen.

3. Discussion

Spontaneous adrenal haemorrhage is a rare entity whose incidence ranges between 0.14 and 1.1 % when looking at autopsy studies [6]. Prior to the widespread use of imaging investigations, given the non-specific signs and symptoms, post-mortem diagnosis was usually made. In recent years, image detected haemorrhage has been found to have an incidence up to 5 % of hospital admitted patients [12]. This rate is even higher when looking at post-mortem studies of people who died from shock specifically, which have been reported up to 15 % [6].

Adrenal haemorrhage has several risk factors. These are typically separated into traumatic causes (blunt or penetrating) and non-traumatic causes. Non-traumatic causes include anatomical pathologies such as tumors, cysts, haemangioma's, metastases [9,12]; coagulopathic disorders (such as antiphospholipid syndrome) and anticoagulant medication; stress related disorders such

Table 1
Blood panel results on admission and day 1 in hospital.

Blood test	Unit	Normal value	Admission	Day 1 hospitalised
Urea	mmol/L	2.1–7.1	14.5	16.9
Creatinine	Umol/L	64–104	351	404
eGFR (estimated glomerular filtration rate)	MDRD formular (mL/min/1.73 m^2)	65	12	10
CRP (C-Reactive Protein)	Mg/L	<10	18	148
WCC (White cell count)	x10^9/L	3.92–10.40	10.20	14.29
Hb (Haemoglobin)	g/dL	13.4–17.5	11.5	6.7

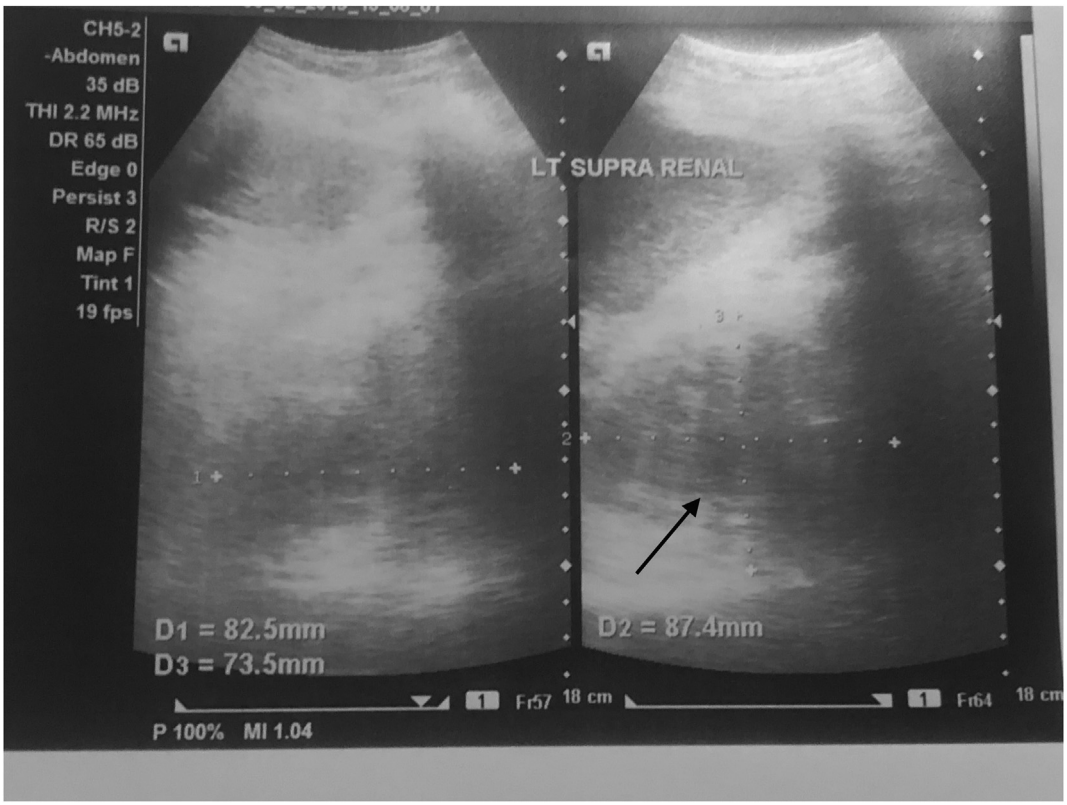

Fig. 2. Abdominal ultrasound. Black arrow, showing an apparent mass on the upper pole of the left kidney/suprarenal measuring 83x87x74 millimetres.

as sepsis, pregnancy and surgery or rarely it occurs spontaneously [9,12]. No risk factor was found in the initial workup of the patient presented in the case, he had no macroscopic anatomical defects detected on imaging and his subsequent histopathology did not reveal any microscopic pathology. He had no known history of trauma and was not on anticoagulant medication prior to his presentation. He was not worked up for an underlying coagulopathy that, in retrospect, would have been indicated. Despite the absence of an underlying risk factor, his clinical course was complicated by surgery and sepsis. This undoubtedly compounded the problem, as they are independent risk factors themselves. AH is associated with a 15 % mortality rate in isolation, climbing to 50 % in the presence of sepsis [2]. When bilateral, it is associated with a 50 % progression rate to adrenal insufficiency and death [2,6,13]. This high mortality rate was clearly reflected in the difficulty in management and subsequent demise of the patient in the case.

There is limited evidence as to a clear cause of spontaneous AH. However, a few theories and considerations have been postulated. The anatomy of the adrenal gland and its blood supply make it particularly vulnerable [12]. It has a rich vascular inflow, with branches from the inferior phrenic, renal and aortic arteries

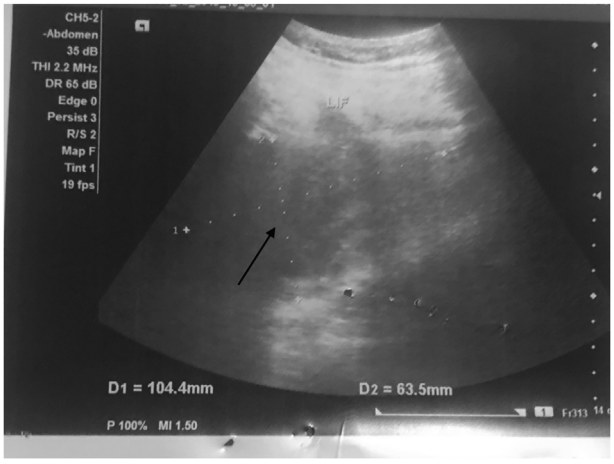

Fig. 3. Abdominal ultrasound. Black arrow, showing a left iliac fossa mass/collection 104x64 millimetres non vascular.

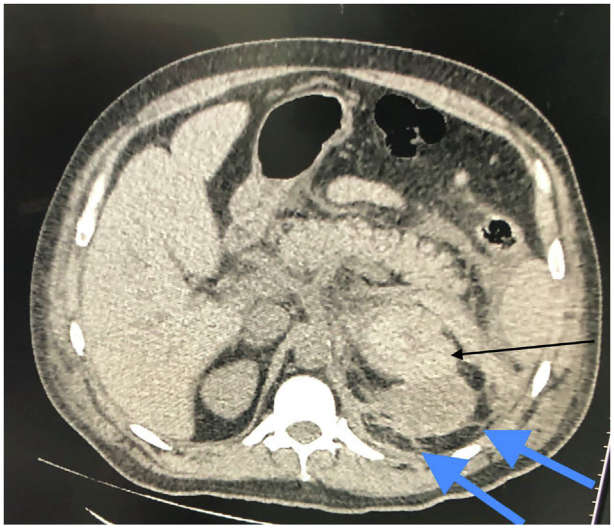

Fig. 4. Pre-operative CT. Black arrow showing a left suprarenal hyperdense mass. Blue arrows showing retroperitoneal haematoma with perinephric fatty stranding.

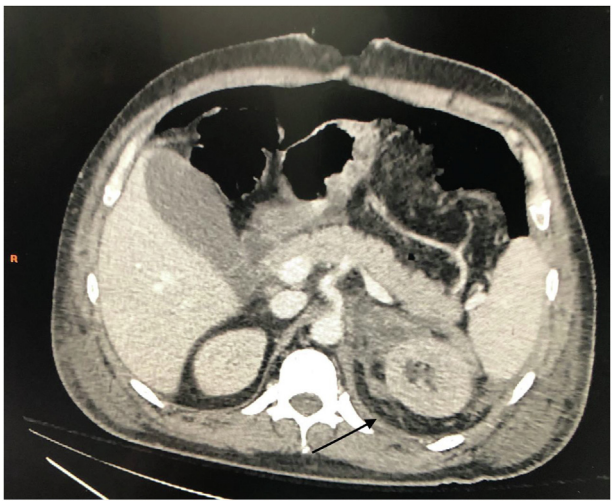

Fig. 5. Post-operative CT. Black arrow shows retroperitoneal haematoma with perinephric fatty stranding. Adrenal haematoma has been removed.

supplying the gland. These drain into a capillary network and sinusoids that essentially constitute to a reservoir [9,12]. The reservoir then drains into a single adrenal vein [9,12]. This means that any increase in blood supply or pressure, impacts venous drainage disproportionately and may lead to haemorrhage. The catecholamines and ACTH released from the adrenal medulla in times of stress also accentuate bleeding. Their release leads to vasoconstriction and platelet aggregation that ultimately increases viscosity, turbulence, venous pressure and subsequent haemorrhage [7,8,12].

Adrenal haemorrhage has been documented in vulnerable groups such as neonates and pregnancy [14]. This mechanism is similarly due to a relative state of stress as well as physiologic changes in pregnancy such as cortical hypertrophy of the adrenal gland and hypercoagulability [4,12,14]. Risk factors in the neonatal group include difficult deliveries (prolonged labour, malpresentation), neonatal sepsis and coagulopathies [12,14]. In these patients, it is most commonly documented in the right adrenal gland but up to about 10 % of cases are bilateral [1]. In pregnancy, the incidence of which is unknown, it also typically involves the right adrenal gland [4,10]. Although less common, there has been a case documented with left spontaneous AH in the third trimester of pregnancy, which was managed with left adrenalectomy post caesarean section [4]. There is limited literature documentation of unilateral left spontaneous AH, other than in pregnancy, as was seen in this case report. Our case report is of a left spontaneous adrenal haemorrhage, making it one of the first documentation other than in pregnancy.

Despite the challenges in making the diagnosis, investigations are targeted at making the diagnosis, excluding hormonal activity and searching for lethal complication i.e., adrenal insufficiency [13]. Imaging studies include ultrasound, Computed Tomography (CT) scan or Magnetic Resonance Imaging (MRI), the latter of which is the investigation of choice [2,3]. These are used to make the diagnosis and identify any tumors. Hormonal assays typically include urine vanillylmandelic acid (VMA) to exclude an active Pheochromocytoma and cortisol and ACTH to workup for adrenal insufficiency [13]. The diagnosis in our patient was suggested on sonar but definitively made on CT. He had negative urine metanephrines and VMA. Following diagnosis, it can be managed conservatively with angioembolization and/or adrenalectomy [9]. Our case was managed with left adrenalectomy. Despite the method selected for management, it is associated with a high mortality rate and due to the systemic effects and secondary sepsis.

In conclusion, spontaneous adrenal haemorrhage is rare with non-specific presentation and requires high index of suspicion to diagnose. Imaging (CT/MRI) is the investigation of choice for diagnosing of adrenal haemorrhage. Unstable non-traumatic patients that have peritonitis requiring laparotomy with haemoperitoneum and have no obvious source of bleeding, consider possible adrenal haemorrhage. Combination of adrenal haemorrhage and sepsis is associated with a high mortality rate, despite early use of steroids, as seen in this case report.

"Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request".

Ethical approval

Ethics approval has been obtained by the University of Witwatersrand Ethics Committee M190482.

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Author contribution

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Conflicts of interest statement

No conflicts of interest.

Guarantor

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Registration of research studies

Not applicable.

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The following information is required for submission. Please note that failure to respond to these questions/statements will mean your submission will be returned. If you have nothing to declare in any of these categories then this should be stated.

Consent

Consent obtained from the next of kin, the father of the patient.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijso.2021.100373.

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