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Case report

An acute complication of adrenal pseudocyst – a rare phenomenon

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ABSTRACT

Pseudocysts of the adrenal gland are a rare entity and are usually discovered incidentally. They result from haemorrhage within a normal adrenal gland and can become massive due to large amount of fluid collection. Pseudocysts constitute 6% of adrenal incidentalomas and may be associated with pheochromocytoma, other primary adrenal tumors, and cysts. Therefore, all cases of adrenal pseudocysts warrant a comprehensive evaluation to exclude any underlying functional tumor.

1. Introduction

Adrenal pseudocysts are benign cystic masses enclosed by a fibrous wall.[1] Epidemiologically, women aged 30–60 years are commonly affected.[2] Trauma, infection, or bleeding may be the underlying pathology for their formation. Bleeding occurs secondary to underlying primary or metastatic adrenal tumors, anticoagulant/hormonal therapy, immune thrombocytopenic purpura (ITP), and severe sepsis.[3–5]. They are usually detected incidentally. Rarely, hemorrhage within the pseudocyst occurs, and maybe it is the first presenting symptom.[6] We present a case of a young female who presented with nonspecific symptoms and was diagnosed as a case of acute hemorrhage within a right adrenal pseudocyst.

2. Case report

A girl, aged 22 years, presented with complaints of sudden-onset severe pain in the right upper abdomen associated with nausea and multiple episodes of nonbilious vomiting, 10 days before consultation in our hospital. She was treated for ITP at the age of 10 years. After administration of steroids, she was cured of the disease, with no further episodes of bleeding or deranged coagulation profile.

During the current episode, she was admitted elsewhere, and evaluation there revealed a steady decline in hemoglobin levels over a span of one week (11 g% to 7 g%). Computed tomography (CT) scan of the abdomen reported a large cystic lesion in the right suprarenal region. She was then referred to our center. Upon presentation to our hospital, she appeared pale but hemodynamically stable, with a heart rate of 88 beats per minute, blood pressure of 110/60 mm Hg, and oxygen saturation of 98% at room air. Tenderness in the right lumbar region was noted on abdominal examination. Rest of the systemic examination was unremarkable. Films of CT scan performed at the hospital of previous admission were unavailable due to insurance policies. Because the patient was stable, magnetic resonance imaging (MRI) of the abdomen was performed which reported an acute-subacute hemorrhage in an underlying right adrenal cyst measuring 9.35 cm in diameter with perinephric extension (Fig. 1). Serial measurements of hemoglobin levels did not show further decline. Rest of the biochemical and hematological parameters were within normal limits including platelet count (3 lakh/mm^3) and coagulation profile. A thorough endocrinology workup comprising plasma and urine aldosterone, plasma and urine catecholamines, free cortisol, and dehydroepiandrosterone excluded an underlying functional adrenal tumor. There was also no evidence of adrenal insufficiency.

A large symptomatic pseudocyst warranted excision, and so a laparoscopic right adrenalectomy was planned. Intraoperatively, a large cyst replacing the right adrenal gland and densely adherent to the underlying inferior vena cava was noted, hindering the dissection. The procedure was converted to open, and the cyst was excised in toto after ligating the adrenal vessels.

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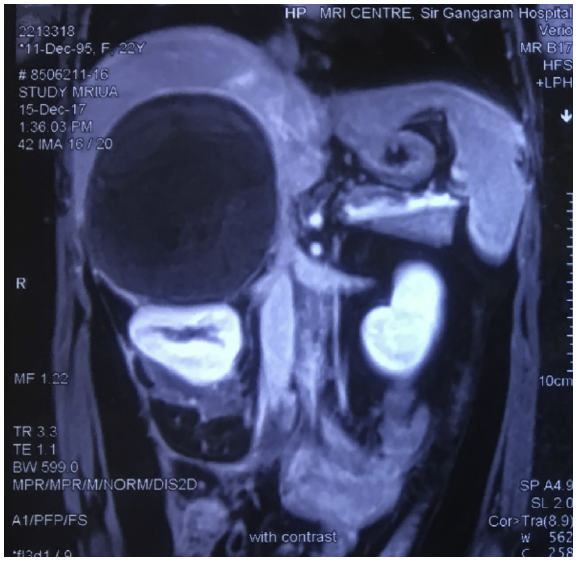

Fig. 1. Abdominal MRI showed a lesion of 9.35 cm in diameter in the right suprarenal region. The lesion is indenting upon the inferior surface of hepatic parenchyma and displacing the kidney anteriorly and inferiorly. MRI, magnetic resonance imaging.

Gross examination of the specimen revealed a cystic mass with congested surface and wall thickness of 0.2–0.4 cm. Microscopically, no surface epithelial lining was noted, and there were areas of hemorrhage in the fibro collagenous wall with a periphery of normal adrenal cortical tissue. No evidence of atypia was noted. Thus, a diagnosis of pseudocyst of the right adrenal gland was concluded.

The patient made a steady recovery postoperatively and was discharged on postoperative day 6 in a satisfactory condition.

3. Discussion

Only 125 cases of adrenal pseudocysts have been reported in literature so far.[7] Even rarer are cases reported on complications of pseudocysts. The adrenals are more prone to hemorrhage because of the rich vascular bed, formed by three arteries but only one vein[8]. The hypothesis stands that stress increases the adrenal vascularity and in turn the adrenal venous pressure due to vasoconstriction, thereby causing intraglandular hemorrhage.[9]

The clinical presentation is varied and ranges from nonspecific symptoms to shock secondary to profuse bleeding. Unilateral adrenal hemorrhage does not cause adrenal sufficiency. The initial diagnostic workup is based on 4 conditions that can cause adrenal hemorrhage, namely, tumors, hemorrhagic diathesis, stress, and idiopathic diseases. More emphasis is given to detecting underlying tumors, if any, by imaging and laboratory evaluation. CT scan and MRI are the ideal imaging techniques for diagnosing Pseudocysts and their complications. Both precontrast and post contrast images are included in CT/MRI protocol to differentiate between primary and tumor-related hemorrhage. Absence of gadolinium enhancement in MRI is an important proof of benign hemorrhagic pseudocyst,[10] as was seen in this patient [Fig. 2].

The management depends on the underlying pathology, severity of hemorrhage, and presence of active bleeding. In cases of massive hemorrhage within the pseudocyst, immediate adrenalectomy is necessary. Benign adrenal pseudocysts more than 5 cm in size should be excised after exclusion of a secretory tumor.[11]

Despite a thorough evaluation in our patient, the cause of hemorrhage could not be found. She had been previously cured of

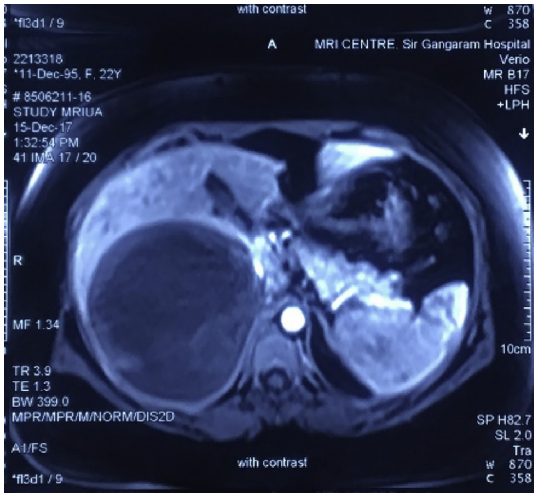

Fig. 2. No contrast enhancement noted in arterial, portal, or delayed contrast images. There is significant pressure effect in the inferior vena cava (displaced medially and anteriorly) due to the lesion, suggestive of hemorrhage.

ITP, as was also proven by hematological tests. We assume that the pseudocyst formed previously secondary to ITP became symptomatic now as a result of the acute hemorrhage. Thus, it is appropriate to add this case to the small list of complicated adrenal pseudocysts reported in literature.

Conflict of interest

The authors declare that they have no conflict of interest.

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