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Trauma and reconstruction

Adrenal hemorrhagic pseudocyst - A case report of a rare presentation of pheochromocytoma

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Introduction

Pheochromocytoma is a rare tumour of the catecholamine-producing cells of the adrenal medulla. Although the incidence varies in several reports, approximately 1–2 per 100,000 individuals are diagnosed annually with pheochromocytoma.[1] Pheochromocytoma clinically is characterized by specific symptoms such as severe hypertension, tachycardia, palpitations, cardiac arrhythmia, weight loss, and sweating.[2] Today nearly 40% of cases of pheochromocytoma, are found as incidentaloma with no specific symptoms.[1,3,4]

Pheochromocytomas are known to vary in their enzymatic composition and in their ability to self-metabolize the catecholamines within each tumour's secretory vesicles. Therefore, this could lead to great variability in the amount and ratio of catecholamines secreted by pheochromocytomas. Indeed these differences in norepinephrine, epinephrine and dopamine secretion explain the heterogeneity in clinical behaviour of pheochromocytomas.[4] Definitive pre-operative diagnosis of pheochromocytoma is often difficult in the clinical setting. We report a case of pheochromocytoma in a patient presenting with abdominal pain and having a past history of trivial trauma to the abdomen six months earlier.

Case report

A 56 year old male presented to the urological services of the hospital with symptoms of on and off pain of six months duration. The severity of the pain was mild to moderate and was not associated with hematuria. The patient was a known hypertensive and was on calcium channel blockers for the same. Patient was also a known diabetic on treatment. Routine blood and urine tests were normal. Ultrasonography of the abdomen revealed a well-defined solid cystic mass measuring 8.2× 9.1 *cms* in the region of right suprarenal gland pushing the right kidney downwards. The fat planes were preserved and the lesion showed mild vascularity on Doppler study. Twenty four hour urinary excretion of vanillyl mandelic acid (VMA) was 44.60 mg (four times the normal values).

Computed Tomography (CT) imaging revealed a well-defined lobulated solid-cystic heterogeneously enhancing mass lesion in the right suprarenal region measuring 7.7 × 6.9 × 8.8 cms (Fig. 1). The right adrenal gland was not seen separately from this lesion. It was abutting the duodenum and IVC (inferior vena cava) medially, right crura of diaphragm posteriorly, segment I and V of the liver superiorly. Fat planes between the lesion and right kidney were maintained.

The patient was started on other anti-hypertensive drugs (alpha and beta blockers) and treated conservatively for about one week. In view of the raised urinary VMA levels, hypertension and the lesion in the right adrenal gland, the patient was advised right adrenalectomy. Patient was explored through a right loin incision and the lesion exposed. The lesion appeared like a mass of hemorrhagic pseudocysts with clotted blood within (Fig. 2). The mass was dissected out and excised. The

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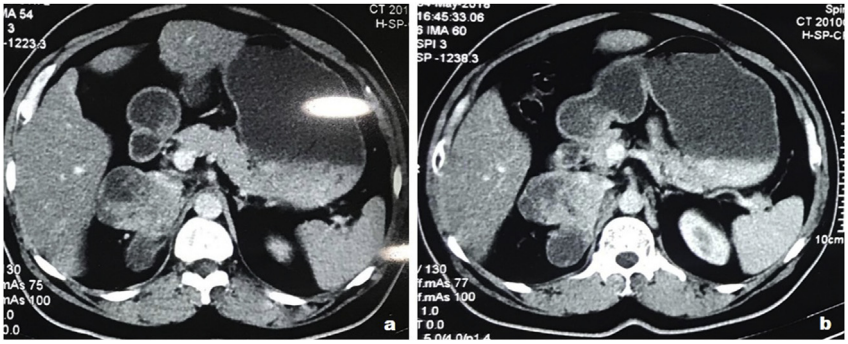

Fig. 1. a & b. Computed Tomography (CT) imaging revealed a well-defined lobulated solid-cystic heterogeneously enhancing mass lesion in the right suprarenal region measuring 7.7× 6.9 × 8.8 cms.

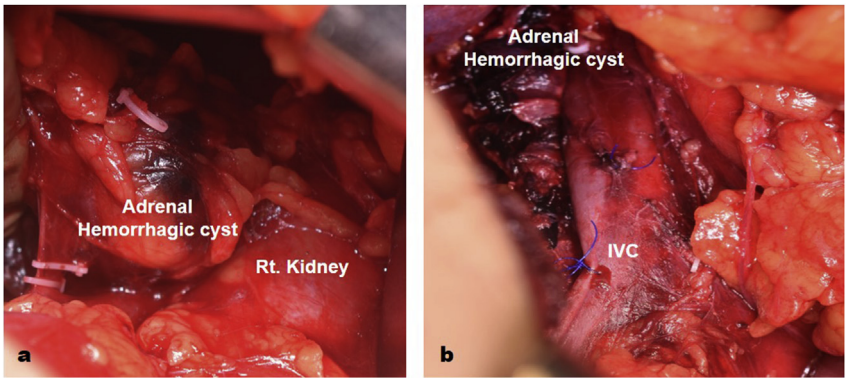

Fig. 2. a & b. The lesion appeared like a mass of hemorrhagic pseudo-cysts with clotted blood inside.

intra-operative and post-operative period was uneventful. On repeated questioning in the post-operative period, the patient gave a history of a trivial fall from a two wheeler about six months prior and sustained blunt injury to the chest, loin and upper abdomen on the right side. The patient then had taken pain killers on the advice of a local doctor.

The patient was discharged on the 5th post-operative day with advice of continuing anti-hypertensives. Histopathological examination of the surgical specimen revealed a pheochromocytoma (Fig. 3). The alpha blocker was discontinued a week later and the patient's blood pressure was well controlled and maintained. Urinary VMA repeated three months later was within the normal range.

Discussion

Our patient presented with no classical symptoms of Pheochromocytoma. Except for elevated levels of urinary VMA and the CT findings of a space occupying lesion, there were no obvious telltale findings. The CT too showed a well-defined lobulated solid-cystic heterogeneously enhancing mass lesion which further confused the diagnosis.[3] The operative findings of a hemorrhagic pseudocyst raised the doubt of adrenal hemorrhage following trauma and in this case hemorrhage could have occurred in a pre-existing adrenal lesion i.e. pheochromocytoma.

Isolated adrenal hemorrhage following blunt abdominal trauma is very rare. Adrenal gland can be injured as an uncommon consequence of blunt abdominal trauma, such as in motor vehicle collisions, falls, or injuries in sports. The estimated incidence of adrenal hemorrhage is approximately 2–3%. Isolated adrenal hemorrhage is a very rare subset of this type of injury and usually has limited clinical significance.[5] Majority of adrenal gland hemorrhage following injuries can be treated conservatively.

Kyoda et al.[2] reviewed clinical and pathological findings of 31 cases with radio-graphically diagnosed pheochromocytoma, including three cases of hemorrhagic pseudocysts (HPC). Biochemical testing showed

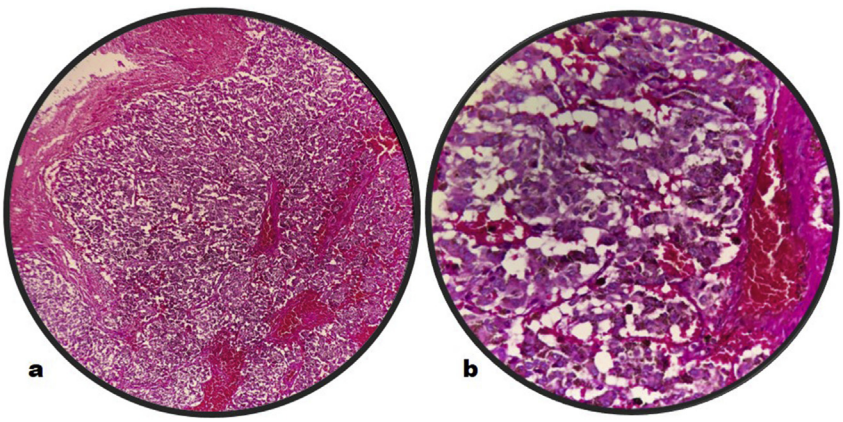

Fig. 3. a. Sections from Rt. adrenal mass showing features of pheochromocytoma (low power 20 X).
b. The neoplasm appears well encapsulated and the cells are round to polygonal with eosinophillic granular cytoplasm. The nuclei are centrally placed. Hemorrhage and hemosiderin pigment is present. Zell Belen pattern is evident (40X).

no definitive excessive catecholamine secretion in any of the three patients with HPC and I^{131} - metaiodobenzylguanidine scintigraphy was negative too in all the three with HPC. All HPC patients had concomitant disease or symptoms suggestive of pheochromocytoma, and two had received an anti-coagulant or anti-platelet agent. The authors concluded that adrenal HPC should be considered as a differential diagnosis of pheochromocytoma.

Surgical treatment of pheochromocytoma includes proper preoperative control of blood pressure, blood transfusions if necessary and all measures to prevent complications. Adrenalectomy be it open, laparoscopic[1] or robot assisted is the treatment of choice. Laparoscopic adrenalectomy is safe, feasible and efficient means of resecting large (> 8 cm) adrenal pheochromocytomas.[5]

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.eucr.2018.08.005.

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