

Oncology

Adrenal cavernous Hemangioma; A rare diagnosis of adrenal incidentaloma: A case report, and literature review

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ABSTRACT

Adrenal cavernous hemangioma is an extremely rare benign tumor. To date, less than 70 cases have been reported in the literature. In most of the cases, the tumor is hormonally silent, discovered incidentally and the diagnosis is made postoperatively. Pre-operative differentiation between this benign tumor and other malignant adrenal tumors is challenging. In this article we present a case of a non-functioning adrenal incidentaloma that was managed by laparoscopic adrenalectomy. The post-operative histological diagnosis was adrenal cavernous hemangioma.

Introduction

Adrenal incidentalomas are adrenal masses that are discovered inadvertently during the evaluation of conditions not related to the adrenals. Adrenal incidentalomas can be malignant or benign and can arise from the cortex, medulla, or extra-adrenal origin. The size of 1 cm is usually used as a cut-off for the evaluation of incidentalomas.¹

Cavernous hemangioma is a rare, non-functioning, benign adrenal tumor that originates from the endothelial lining of blood vessels. It can not be distinguished radiologically from adrenal cortical carcinoma. Hence, it is managed by surgical resection in most cases and the diagnosis of hemangioma is often made post-operatively.²

The first reported case in a live patient was in 1955. Since then, only 66 cases have been reported.³

Here, we present a case of adrenal incidentaloma that was managed by laparoscopic adrenalectomy and found to be an adrenal cavernous hemangioma.

Case presentation

A 58-year-old male patient, who is not known to have any chronic medical illnesses, presented to our emergency department due to left loin pain. A non-contrasted computed tomography (CT) scan showed a 6 mm distal ureteric stone, however, an oval-shaped 7 * 6.7 * 6.1 cm soft

tissue mass lesion in the left adrenal gland was identified, and a central area of fatty deposition and tiny calcification spots were also seen. [Figure \(1\)](#). Due to severe pain, a left double J stent was inserted.

The patient denied any symptoms related to the mass, and on physical examination, the mass was not palpable. The laboratory investigations of adrenal incidentaloma including a urinary free cortisol test, a urinary metanephrine, and a plasma renin-to-aldosterone ratio were normal.

A contrasted CT scan demonstrated the previously-mentioned mass with heterogeneous enhancement, ranging from 60 to 90 HU. The mass was gently compressing but not invading the upper pole of the left kidney. [Figure \(2\)](#).

Because malignancy cannot be ruled out, surgical resection was planned. He underwent laparoscopic adrenalectomy, at which, the tumor was smoothly dissected from the surrounding organs without invasion or dense adhesions.

The postoperative course was smooth and uneventful. The histopathological examination revealed a proliferation of dilated blood-filled spaces that are lined by flattened epithelium and surrounded by adrenal tissue. [Figure \(3\)](#).

Discussion and conclusion

The rate of adrenal mass detection has increased due to the

Abbreviations: CT, Computed Tomography; MRI, Magnetic Resonance Imaging; HU, Hounsfield Unit.

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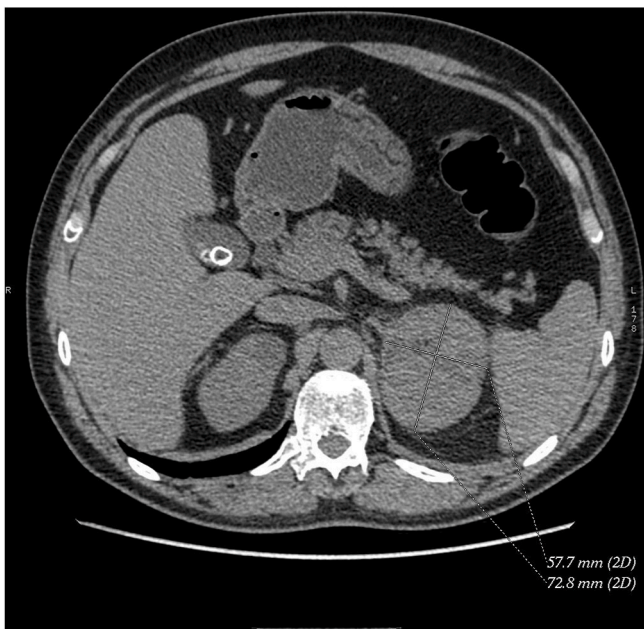


Fig. 1. Non-contrast Ct scan showing an oval-shaped soft tissue mass lesion in the left adrenal gland with fatty deposition and tiny calcification spots.

widespread utilization of abdominal ultrasonography, CT, and MRI in the evaluation of different abdominal conditions. This brings to the light rare types of tumors that can affect the adrenal glands including cavernous hemangioma.

Adrenal cavernous hemangioma has a median age of 60 years at diagnosis. It has a female preponderance with a 3:2 female to male ratio.²

On a histological basis, adrenal hemangiomas are divided into cavernous and capillary subtypes. In the more common cavernous subtype, the tumor is composed of an enlarged mass of endothelially-lined sinusoids that are filled with blood and is displacing the normal tissues.⁴

The Hemangioma itself is not active endocrinologically. This can explain the findings of Degheiliet al. Review in which only 6 of the 66 reviewed cases were hormonally active.² Moreover, Noh et al. proposed that the cause of hormonal activity in those cases is not hypersecreting tumor cells, but the arteriovenous malformations within the lesions that facilitate the entry of hormonally active metabolites into the bloodstream.³

Most of the reported cases were asymptomatic and found incidentally. In symptomatic patients, vague abdominal pain and discomfort due to the mass effect were the most common presenting symptoms. Other presenting symptoms including retroperitoneal bleeding and hyperaldosteronism were infrequently reported.²

On contrasted CT, Noh et al. found that adrenal hemangiomas tend to be heterogeneous and hypodense in their internal structure (86% of the cases), with a peripheral patchy or centripetal pattern of enhancement (55% of the cases). Characteristic calcifications have also been reported in 51% of the cases. The calcifications were described as either speckled or centrally located with an irregular, stellate pattern.³ Those calcifications were described as being pathognomonic for adrenal hemangioma. However, it lacks specificity as it may present in various adrenal diseases including carcinoma, cyst, tuberculosis, neuroblastoma, and metastatic melanoma⁵

While some reports suggest that adrenal cavernous hemangiomas



Fig. 2. Cross-section (A) and Coronal (B) views Contrastd ct scan showing a heterogeneouslyenhancing mass in the left adrenal gland compressing the upper pole of the left kidney.

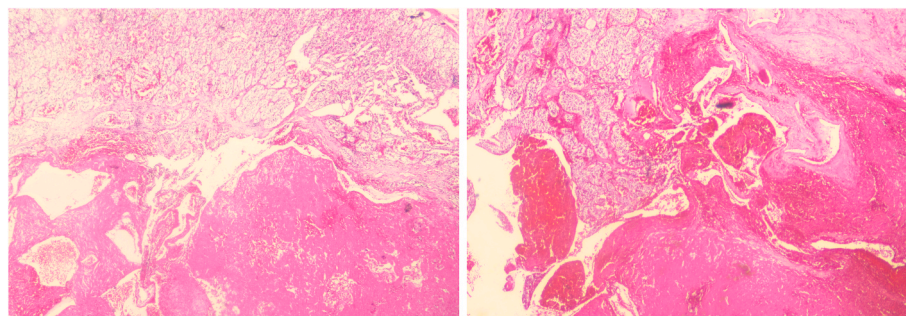


Fig. 3. Hematoxylin and eosin stain (× 40) showing Large thin-walled, cystically dilated blood vessels lined by endothelial cells.

tend to appear hypointense on T1-weighted MRI,² Noh et al. review found no strong clinical evidence to back this suggestion. Because of the large amount of blood contained inside the mass, the majority of the cases were hyperintense on T2-weighted MRI.³

Nevertheless, none of these radiological findings was specific enough to diagnose adrenal cavernous hemangioma, hence, most of the cases including ours were diagnosed retrospectively on postoperative histological examination.

Because of the rarity of adrenal cavernous hemangioma, optimal treatment guidelines have not been established. Surgical resection of adrenal incidentaloma is indicated if the tumor is hormonally active or exceeding 6 cm, due to the risk of malignancy.¹ Because most of the reported cases were hormonally silent, the size of the mass was the main indication of adrenalectomy. However, other indications including mass effect symptoms have been also reported.² On the other side, when the tumor is small and is not associated with a hormonal disturbance, active surveillance with endocrinological and radiological evaluation may be considered.³

Laparoscopic adrenalectomy was reported in 16 of the 63 cases reviewed by Degheili et al.² This can be explained by the fact that the risk of malignancy and bleeding from a hypervascular tumor stand against minimally invasive approaches. However, Noh et al. argued that laparoscopic adrenalectomy in cavernous hemangioma is a safe and feasible modality. Furthermore, they proposed that the risk of bleeding in adrenal cavernous hemangioma is less than in hepatic hemangioma because of the presence of a rigid fibrotic capsule in the former.³

In conclusion, adrenal cavernous hemangioma is one of the rare differential diagnoses of adrenal incidentaloma. Pre-operative differentiation between this benign tumor and other malignant adrenal tumors is challenging. Hence, most of the cases require surgical resection due to the risk of malignancy, mass effect symptoms, or the risk of spontaneous bleeding. Laparoscopic adrenalectomy seems to be safe modality of treatment.

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Availability of data and materials

Not applicable.

Ethics approval and consent to participate

The study abides by the guidelines for human studies and is

conducted in accordance with the World Medical Association Declaration of Helsinki.

Consent for publication

The patient has given his written informed consent to publish his case including publication of clinical data and images.

Author contributions

Samer Al-Rawashdah: Conception and design, drafting the article, critical review, and final approval of the version to be submitted.

Hammam Mansi: Acquisition of data, drafting the article, and final approval of the version to be submitted.

Antonio Luigi Pastore: Acquisition of data, critical revision of the article, and final approval of the version to be submitted.

Antonio Carbone: Conception and design, critical revision of the article, and final approval of the version to be submitted.

Declaration of competing interest

The authors declare that they have no competing interests.

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