

## Giant Cavernous Hemangioma of the Adrenal Gland: Case Report and Review of the Literature

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### Abstract

Adrenal tumors are nowadays being detected with increasing frequency due to the widespread use of various radiological imaging techniques (CT, MRI, US). Incidentally discovered adrenal masses (incidentalomas) are shown in 1% to 5% of all abdominal CT scans performed. Cavernous hemangiomas of the adrenal gland are extremely rare, benign in nature and most usually non-functioning lesions. We report a case of a 50-year-old female who presented with flank pain and abdominal discomfort. MRI of the abdomen revealed a large, oval, adrenal tumor mass, embedded between the upper pole of the right kidney and inferior surface of the liver. Surgery was performed, and tumor was excised completely. Histopathological examination revealed a cavernous hemangioma of the adrenal gland.

**Keywords:** Incidentaloma; Hemangioma; Cavernous; Adrenal gland; Surgical excision

### Introduction

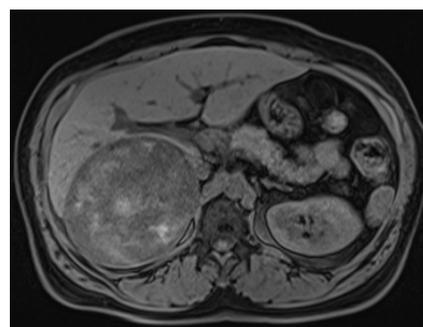
Uncommon adrenal tumors include cystic lesions (endothelial cyst, hydatid cyst), solid lesions (ganglioneuroma, hemangioma, angiosarcoma) and solid fatty lesions (collision tumor, myelolipoma). Adrenal hemangiomas are non-functioning benign, solid tumors and they are most usually cavernous, unilateral and encapsulated. To date, only 3 cases of hormone-secreting cavernous hemangiomas have been reported [1-4]. These tumors are more frequent in the sixth and seventh decade of life with female-to-male ratio being 2:1 [3]. Great majority of adrenal hemangiomas are diagnosed postoperatively after histopathological examination due to the rarity of these tumors and the lack of specific symptoms. Histologically, these tumors are characterized by the presence of blood-filled, dilated vascular spaces lined with mature endothelial cells. First report was published in 1955 by Johnson and Jeppesen [5]. Performing a literature review, using Embase and Medline, we have found 63 documented cases of adrenal cavernous hemangioma in English literature [6-10]. The size of reported tumors ranged from 2 to 25 cm in diameter, with majority measuring of more than 10 cm, probably because most of these tumors are usually asymptomatic until they start showing mass effects due to the tumor growth and compression on adjacent structures [11-13]. When large, tumor can cause flank pain, early satiety or can be evident as palpable abdominal mass on physical examination. Complete surgical excision is advised in all adrenal lesions greater than 6 cm because of the significant risk of malignancy which goes up to 25% [1,14]. Symptoms related to tumor mass effects as well as the risk of spontaneous tumor rupture and hemorrhage are also indications for surgical treatment.

### Case Report

A 50-year-old female was admitted to our hospital due to intermittent flank pain and abdominal discomfort. Symptoms lasted for the past 2 months. Laboratory findings were within reference ranges, as well as tumor markers. MRI of the abdomen and pelvis was performed and large tumor mass of the right adrenal gland was detected. Tumor was 11.5 cm × 11 cm × 11 cm in size, well delimited and encapsulated with no infiltration of surrounding structures, showing a dislocation and malrotation of the right kidney as well as the compression on the inferior surface of the liver (Figure 1). Interior of the lesion was non-homogeneous with zones of necrosis and hemorrhage (Figure 2). Endocrine secretion tests were done and showed all parameters to be within the normal range, confirming non-functioning adrenal



**Figure 1:** MRI in coronal section showing a large, oval, encapsulated tumor mass embedded between the visceral surface of the liver and the superior pole of the right kidney.



**Figure 2:** MRI in axial section showing a large, well delimited, non-homogeneous mass with central zones of necrosis and hemorrhages.

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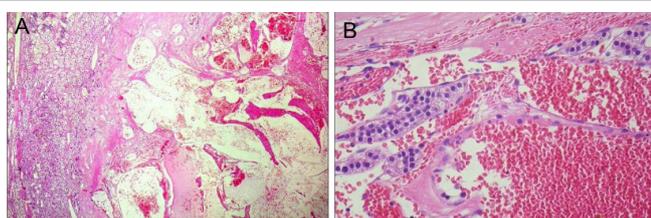
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**Figure 3:** Completely enucleated tumor mass with destructed parts of adrenal gland stretched over the surface of the lesion.



**Figure 4:** Histopathology. A: Histopathological findings revealed cavernous type of haemangioma with remnants of stretched adrenal gland tissue at the periphery (A, H&E, original magnification 13x); B: Areas of intermingled adrenocortical and haemangiomatous elements (B, H&E, original magnification 64x).

tumor: aldosterone 377 pmol/L; renin 8.1 pg/mL; noradrenaline 142 nmol/24 h; adrenaline 22 nmol/24 h; normetanephrine 0.78  $\mu$ mol/24 h; metanephrine 0.42  $\mu$ mol/24 h; dopamine 2112  $\mu$ mol/24 h; chromogranin A 79  $\mu$ g/L; adrenocorticotrophic hormone (ACTH) 3.1 pmol/L; DHEA-S 3.98  $\mu$ mol/L; 17-OH-progesterone 6.6  $\mu$ mol/L; serum cortisol (1 hour after 250  $\mu$ g ACTH stimulation test) 729 mmol/L. Due to impossibility of ruling out the malignancy and tumor mass effects on adjacent organs, we decided to perform surgery. Large size of the tumor was a contraindication for the laparoscopic approach. A right adrenalectomy was performed through the right subcostal incision, with complete excision of the tumor mass. Tumor weighted 650 gr with destructed parts of adrenal gland stretched over the surface of the lesion (Figure 3). On macroscopic section, tumor mass was encapsulated while interior of the lesion was formed of sponge-like, dark brown tissue, containing lacunas and sinuses filled with blood. Histopathology revealed cavernous type of haemangioma with remnants of stretched adrenal gland tissue at the periphery (Figure 4A) and areas of intermingled adrenocortical and haemangiomatous elements (Figure 4B). Postoperative course was uneventful, and patient was discharged on the 6<sup>th</sup> postoperative day.

## Discussion

Hemangiomas are benign vascular tumors usually affecting liver and skin. An involvement of genitourinary organs (ureter, prostate, bladder) is rare while adrenal localization is extremely uncommon. Adrenal hemangiomas are non-functioning, benign tumors and their preoperative diagnosis can be quite challenging. Most often, they are discovered as incidentalomas, either during imaging or at autopsies. These lesions are almost always of cavernous type with unilateral localization while bilateral involvement has been reported only twice in literature [15]. Most commonly, these neoplasms are asymptomatic or with vague clinical presentation such as non-specific abdominal pain or symptoms related to the compression effects of tumor on the

surrounding structures. Abdominal CT and ultrasound exams most frequently reveal an encapsulated, heterogeneous tumor mass with areas of calcification and cystic spaces [16,17]. An enhanced CT may show irregular enhancement in the peripheral areas of the mass where residual tissue of the compressed adrenal gland still persists. Pooling of contrast material to the peripheral areas of the lesion may be shown on dynamic CT which correlates with the large venous sinuses usually seen on histological examination [16,17]. We would like to emphasize that MRI should be preferred imaging modality with peripheral enhancement of the tumor being the most common finding [8]. Other typical findings include multiple areas of hemorrhage and necrosis, as well as areas of calcification [18]. Other diagnostic modalities such as angiography may be also useful. Vascular channels that retain contrast material in delayed films are characteristic for the neovascularization of hemangiomas. Although the incidence of adrenal incidentalomas have increased lately with the increased utilization of cross-sectional imaging (CT and MRI), detailed scans that are being performed nowadays rarely require percutaneous biopsy. Unfortunately, a negative biopsy (core-needle) does not exclude a malignancy. It is important to differentiate incidentalomas compared to adrenal lesions identified during diagnostic workup for staging cancer patients or symptomatic patients because the treatment can be crucially different. Adrenal cavernous hemangiomas are typically asymptomatic until they reach sizes greater than 10 centimeters in diameter [17]. Indications for the removal of adrenal tumors are size, impossibility to exclude malignancy, tumor mass effects and complications such as hemorrhage, necrosis, or thrombosis. The treatment for smaller and asymptomatic cavernous hemangiomas is conservative with periodic follow up. Surgical resection is therefore required to exclude malignancy, relieve pressure related symptoms, and prevent hemorrhage [19].

## Conclusion

In our case, we decided to perform surgery due to clinical symptoms caused by tumor size and impossibility to rule out the malignancy. Laparoscopic approach should be taken into consideration if the lesion is less than 6 cm [19,20]. Partial adrenalectomies can be performed with robotic, image-guided surgery. Larger tumors of uncertain etiology should be removed through laparotomy. Finally, if performed, bilateral adrenalectomy, postoperatively should include consideration of supplementation of glucocorticoids, mineralocorticoids and adrenal androgens. We consider reporting of this rare entity to be useful as it highlights the disease itself and also emphasize the need to consider this rare etiology among the differential diagnosis in symptomatic adrenal masses.

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