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Rare Adrenal Tumor: Cavernous Hemangioma. A Case Report and Review of the Literature

Zineb Ait Si Ali[1*], Rania Dassoufi[1], Sana Rafi[1], Ghizlane El Mghari[1], Nawal EL Ansari[1]

[1]Department of Endocrinology, Diabetology, Metabolic Diseases and Nutrition Cadi Ayyad University, Mohammed VI University Hospital, Marrakech, Morocco

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***Corresponding author:** Zineb Ait Si Ali
Department of Endocrinology, Diabetology, Metabolic Diseases and Nutrition Cadi Ayyad University, Mohammed VI University Hospital, Marrakech, Morocco

Abstract **Case Report**

***Introduction*:** Adrenal hemangioma is a very rare, benign vascular tumor, the cavernous type is the most common. ***Case report*:** We present the case of a 66-year-old female patient, diagnosed with an adrenal left mass of 56× 65 ×58 mm. The patient underwent laparoscopic tumoral resection. Histopathological examination revealed adrenal cavernous hemangioma. ***Discussion & Conclusion*:** Adrenal cavernous hemangioma is a benign tumor, generally unilateral, often asymptomatic, discovered incidentally or after autopsy. it can be misdiagnosed preoperatively as adenomas or malignant tumors of the adrenal gland. Diagnosis of certainty remains the prerogative of anatomopathological examination. The treatment of choice is surgical excision due the difficulty of excluding malignancy and hemorrhagic risk.

Keywords: Cavernous hemangioma - Adrenal - Diagnosis - Management - Surgery.

INTRODUCTION

Adrenal hemangioma is a very rare, benign tumor, arising from endothelial cells lining the blood vessels, first described in 1955 by Johnson and Jeppesen. It is generally asymptomatic and often voluminous, frequently discovered as incidentaloma during imaging by ultrasonography, computed tomography or magnetic resonance imaging. It presents a diagnostic problem with malignant tumors, especially as its imaging aspects are not always specific [1-4].

In this work, we present a new case of histologically confirmed cavernous hemangioma of the adrenal gland, we highlight its clinical and para-clinical features, and we discuss the challenges of its management in the light of literature data.

CASE REPORT

We report the case of a 66-year-old female patient, her medical history included a cholecystectomy 10 years prior to presentation. She had intermittent left lumbar moderate intensity pain becoming permanent, evolving for one year in a context of conservation of general condition with no other associated signs including no menard's triad or recent weight gain. Clinical examination revealed normal blood pressure and normal heartbeat rate, a moderate obesity BMI = 34kg/m2 with no cushing syndrome and slight tenderness of the left hypochondrium.

The radiological work-up included abdominal ultrasound, which visualized an adrenal left mass. Complementary CT scan (Fig 1) revealed a voluminous left adrenal mass measuring 56× 65 ×58 mm, with spontaneous density of 31 HU, and a moderate enhancement after injection of contrast media, it contains a small well-limited internal fatty component, and it's pushing down the upper pole of the left kidney without local invasion.

Biological findings: Kalemia: 3.6 mmol/l, negative 1mg dexamethasone suppression test at 4.59 µg/dl, normal urinary free cortisol, ACTH: 5.1ng/l, normal SDHEA, normal urinary catecholamines, normal urinary methoxylated derivatives.

Due to the non-specific radiological and biological findings and the size of the tumour, surgical resection was decided upon to establish the final diagnosis. The patient underwent laparoscopic surgery; left adrenalectomy, the mass appeared heterogenous, hypervascularised and mesured 7cm in maximum diameter. Postoperative course was simple and the patient was discharged on the fourth day.

Anatomopathological study showed vascular proliferation in favor of a cavernous hemangioma with no sign of malignancy.

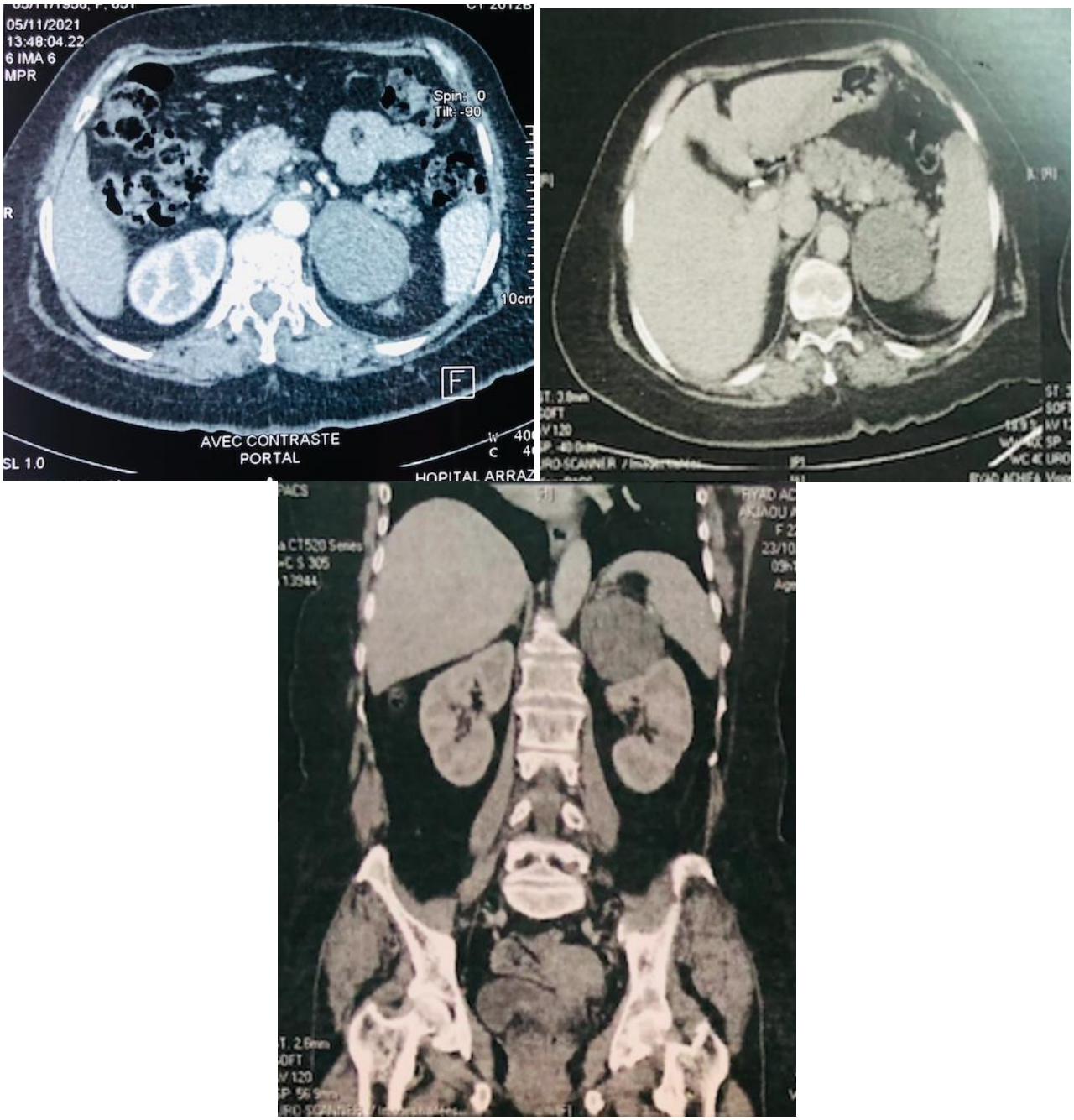

Figure 1: Abdominal CT images showing heterogeneous left adrenal mass of our patient

DISCUSSION

Hemangioma is a benign vascular tumor, representing a group of vascular malformations arising from the endothelial layer of blood vessels, of which the cavernous type is the most common [1, 3]. It develops preferentially in the liver, skin and central nervous system [2, 3, 5]. Involvement of the adrenal glands is extremely rare (0.01% of adrenal tumours) [2].

According to Chua Y *et al*., [3], only 74 cases of adrenal hemangioma were reported between 1955 and 2022. It is generally a unilateral tumor, preferentially affecting women (2/1) between the ages of 50 and 70 [2, 6, 7], often asymptomatic, rarely palpable, painful or complicated [1, 8].

The majority of reported cases were discovered incidentally or after autopsy. A rare case of cavernous hemangioma of the adrenal gland has been described as rupturing and causing severe retroperitoneal hemorrhage

and hyopovolemic shock [8]. Most often, these are non-functional tumors, although rare cases of hormone-secreting adrenal hemangiomas have been described in the literature [4].

They present problems of differential diagnosis, as they can be misdiagnosed preoperatively as adenomas or malignant tumors of the adrenal gland [4, 9]. Enhanced medical imaging has made it possible to diagnose adrenal hemangioma, without however being able to totally exclude the diagnosis of a malignant adrenal lesion: pheochromocytoma, adrenocortical carcinoma or metastasis [2]. Ultrasonography is mostly not helpful, on CT scan Adrenal hemangiomas are usually heterogeneous, hypodense, with peripheral contrast enhancement. Characteristic calcifications due to phleboliths within the dilated vascular spaces of the lesion, have also been reported, but lack specificity as they are also seen in a variety of other adrenal lesions [9, 10].

Diagnosis of certainty remains the prerogative of anatomopathological examination, particularly as surgical treatment is often indicated to prevent the risk of bleeding [1, 5, 11]. On pathological analysis, adrenal cavernous hemangiomas are characterized by multiple dilated vascular channels, lined by a single layer of vascular endothelium surrounded by a collagenous wall. They also contain extensive central necrotic areas mixed with sinusoidal dilatation and fibrotic septa. Like their homologues in the liver and skin, they generally present not only areas of hemorrhage, thrombosis and necrosis, but also degeneration and calcification [9, 12].

The risk of primary adrenal malignancy is related to the size of the mass, with a diameter greater than 4 cm having a sensitivity of 90% but low specificity. Adrenal incidentalomas larger than 4 cm are an indication for surgery, even when they are suspected to be angiomatous due to the bleeding risk and the inability to adjudicate on malignancy [11, 13]. most authors recommend surgical removal of any hemangioma, regardless of the tumoral size because of necrosis, hemorrhage and thrombosis risks [1].

Several approaches to tumor excision in open surgery have been described, namely the anterior, lateral and thoracoabdominal approaches, but laparoscopic adrenalectomy has become the procedure of choice [11]. It offers significant advantages; lower perioperative morbidity, less operative blood loss, less postoperative pain and shortened hospital stay than open adrenalectomy. However, its major limitation for large, potentially malignant adrenal tumors is incomplete resection and capsular rupture, with an increased risk of local recurrence and intra-abdominal neoplastic dissemination [12].

In our case, the patient underwent a laparoscopic surgery, we agree that it's an advantageous, feasible and secure modality for resecting adrenal cavernous hemangiomas with a reduced risk of surgical bleeding.

CONCLUSION

In conclusion, we reported a case of left adrenal cavernous hemangioma which is an extremely rare entity. The non-specific radiological aspects of these tumors make their diagnosis challenging, they are exclusively confirmed after anatomopathological examination. Surgery and especially laparoscopy being the preferred approach when possible, plays an important role in the management of these tumors, ruling out any risk of malignancy and preventing complications related to retroperitoneal haemorrhage.

REFERENCES

1. Harzallah, L., Zouari, L., Chérifa, L. B., Harzallah, F., Sriha, B., Bakir, D., & Kraiem, C. (2006, December). Hémangiome surrénalien: à propos d'un cas. In *Annales d'endocrinologie* (Vol. 67, No. 6, pp. 624-627). Elsevier Masson.
2. de la Villéon, B., Goudard, Y., Peroux, E., Jacquet, S. F., Aubert, P., & Duverger, V. (2011). L'hémangiome caverneux: un incidentalome rare de la surrénale. *Progrès en urologie*, *21*(13), 961-964.
3. Chua, Y., Quake, S., Prasad, K., Elsaify, W., & Quake, S. Y. L. (2022). A Rare Case of Cavernous Haemangioma of the Adrenal Gland: A Case Report and Review of Literature. *Cureus*, *14*(10), e29917.
4. Zemni, I., Haddad, S., Hlali, A., Manai, M. H., & Essoussi, M. (2017). Adrenal gland hemangioma: A rare case of the incidentaloma: Case report. *International journal of surgery case reports*, *41*, 417-422.
5. Feo, C. V., De Troia, A., Pedriali, M., Sala, S., Zatelli, M. C., Carcoforo, P., & Feo, C. F. (2018). Adrenal cavernous hemangioma: a case report. *BMC surgery*, *18*, 1-5.
6. Testini, V., Eusebi, L., Giannubilo, W., Ferrara, V., Bartelli, F., & Guglielmi, G. (2021). Imaging findings and 4 years follow-up of adrenal cavernous hemangioma: a case report. *Egyptian Journal of Radiology and Nuclear Medicine*, *52*, 1-5.
7. Noh, J. J., Choi, S. H., Hwang, H. K., Kang, C. M., & Lee, W. J. (2014). Adrenal cavernous hemangioma: a case report with review of the literature. *JOP. Journal of the Pancreas*, *15*(3), 254-257.
8. Nishtala, M., Cai, D., Baughman, W., & McHenry, C. R. (2019). Adrenal cavernous hemangioma: a rare tumor that mimics adrenal cortical carcinoma. *Surgery Open Science*, *1*(1), 7-13.
9. Edwards, J. P., Stuart, H. C., Urbanski, S. J., & Pasieka, J. L. (2014). A rare cavernous hemangioma of the adrenal gland. *International journal of surgery case reports*, *5*(2), 52-55.
10. Gupta, A., Gupta, A., Gupta, S., Durgapal, P., Sideek, R. A. T., & Verma, S. (2020). Cavernous hemangioma of the adrenal gland: a rare adrenal

incidentaloma. *Nigerian Journal of Surgery*, *26*(2), 159-161.

11. Noureddine, N., Nabil, J., Mohammed, L., Faicel, M., Mohammed, N., Hicham, I., & Aziz, Z. (2017). Hémangiome surrénalien: à propos d'un cas. *The Pan African Medical Journal*, *28*.
12. Antonino, A., Giorgio, R., Juliana, D. L., Giuseppe, A., Roberto, C., Laura, V., ... & Pasquale, G. G. (2016). Adrenal cavernous hemangioma: which correct decision making process?. *Acta Medica*, *32*, 385.
13. Oishi, M., Ueda, S., Honjo, S., Koshiyama, H., Yuba, Y., & Takabayashi, A. (2012). Adrenal cavernous hemangioma with subclinical Cushing's syndrome: report of a case. *Surgery today*, *42*(10), 973-977.