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A Rare Cavernous Hemangioma of the Adrenal Gland

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ABSTRACT

Adrenal cavernous hemangiomas are rare nonfunctioning benign tumors. This case report presents a patient with a huge nonfunctioning adrenal cavernous hemangioma presenting as an adrenal incidentaloma suspicious for adrenal myelolipoma. Although adrenal cavernous hemangiomas are rare, they should be considered as a part of the differential diagnosis of adrenal neoplasms. The proper treatment is surgical excision due the risk of spontaneous tumor rupture and the difficulty of ruling out malignancy.

Introduction

Adrenal cavernous hemangioma is a rare benign tumor. Most of these tumors are asymptomatic. Others present with pain abdomen or hypovolemic shock. We report a case of a left adrenal cavernous hemangioma. It presented as an adrenal incidentaloma suspicious for adrenal myelolipoma. We review the literature on the adrenal cavernous hemangiomas, focusing on the clinical presentation, imaging features and pathology of these lesions.

Case Presentation

A 71-year-old woman complained of chronic abdominal distension was admitted to our hospital on November 14th, 2014.

The patient has no specific signs and symptoms including hypertension, weakness and osteopenia. Physical examination revealed an about 8.0 cm mobile mass on the left flank without tenderness. All laboratory values were within the normal range. All endocrinology tests were normal, so a functional tumor would be ruled out. There were no clinical signs of Cushing's syndrome or adrenogenital syndrome. An incidental large left adrenal mass ($9.4 \times 8.1 \times 8.9$ cm) was found in a computed tomography (CT) scan during a routing control of her disease. The CT scan revealed a well-circumscribed, heterogenous, irregular mass with fat component and peripheral speckled calcifications. Contrast enhanced CT showed no enhancement was found in all phase. Imaging characteristics were suspicious for adrenal myelolipoma as the tumor was well defined and contained fat component (Fig. 1a and b). There was no overt evidence of invasion. On November 25, 2014, the patient underwent laparoscopic left adrenal gland resection via the transperitoneal approach due to diagnostic uncertainty and the risk of spontaneous tumor rupture or bleeding. There was an alternative plan to convert to conventional open surgery if the tumor was too big and difficult to resect. The tumor was resected successfully by laparoscopic procedure. Pathological examination displayed a well encapsulated and moderately firm irregular mass, measuring $9.5 \times 8.0 \times 7.5$ cm, with a cross section of ash-gray organized hematoma (Fig. 2). Microscopic evaluation showed some dilated blood filled vascular spaces with single lining of endothelial cells (Fig. 3). Areas of coagulative necrosis and hemorrhage were seen. Immunohistochemical examination revealed that CD34 was specifically positive in vascular endothelial cells, which demonstrating their endothelial nature. The patient recovered without surgical related complication and was discharged on December 1, 2014.

Discussion

Cavernous hemangioma is a rare, benign, and endocrinologically inactive neoplasm. Adrenal hemangiomas are mostly cavernous, unilateral lesions of the adrenal glands that appear between the ages of 50 and 70 years.[1]

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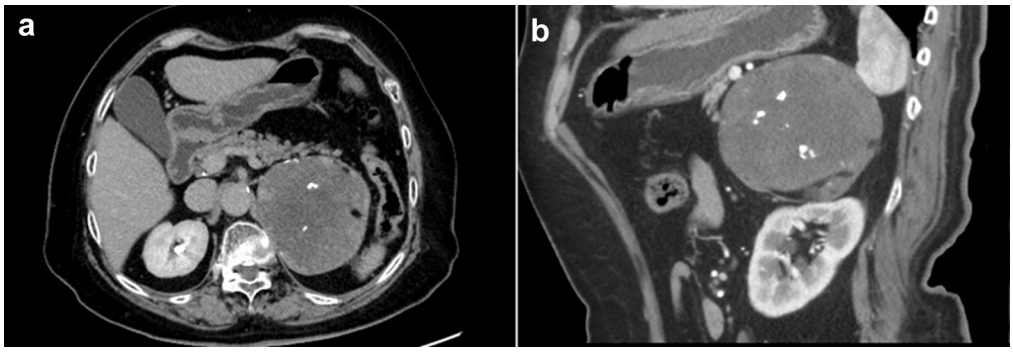

Figure 1. Large left adrenal lesion (9.4 × 8.1 cm) including regions of dystrophic calcification and mature fat but well circumscribed with no obvious areas of invasion (a) axial and (b) sagittal.

Patients usually have no signs and symptoms and have been noticed only when they have large palpable masses or hypovolemic shock caused by spontaneous rupture. This was similarly observed in our case, without associated symptoms of either adrenalism or hypoadrenalism except the presenting of chronic abdominal distension due the oppression by the huge tumor.

While ultrasonography is not helpful in differentiating cavernous hemangiomas from other adrenal lesions, CT and magnetic resonance imaging (MRI) are felt to be contributory. The main findings of adrenal cavernous hemangioma on CT include a hypodense, heterogenous lesion with calcifications, as was seen in our patient. Characteristic calcifications appear speckled throughout the entire mass, which have also been reported in 28–87% of cases.[2] These represent phleboliths within the dilated vascular spaces of the lesion. However, calcifications lack specificity, a variety of other adrenal lesions including adrenal cortical carcinoma, hemorrhage, tuberculosis and metastatic melanoma also show it.[3] It makes diagnosis so difficult that cavernous hemangiomas also tend to exhibit delayed washout of contrast on enhanced computed tomography as the adrenal cortical carcinoma. While the absence of enhancement on enhanced computed tomography in our case, there is a possibility that the contrast agent can not enter the mass due to massive necrosis. MRI shows low intensity signal on T1 weighted images and high intensity signal on T2 weighted images.

Like their more common counterparts in the liver and skin, they generally contain areas of hemorrhage, calcification, necrosis and degeneration. Cavernous hemangiomas are enlarged masses of blood filled spaces. On pathologic inspection, cavernous hemangiomas of the adrenal gland involve the adrenal cortex and consist of multiple dilated vascular channels lined by a single layer of vascular endothelium surrounded by a collagenous wall. Furthermore, the presence of multiple vascular cavities at the periphery is a vital feature, which accounts for the characteristic peripheral nodular contrast enhancement pattern seen on imaging.

The mass size is the main surgical indication. Surgical resection remains necessary for larger adrenal incidentalomas exceeding 6 cm due the risk of malignancy is 35–98%[4] and their propensity to

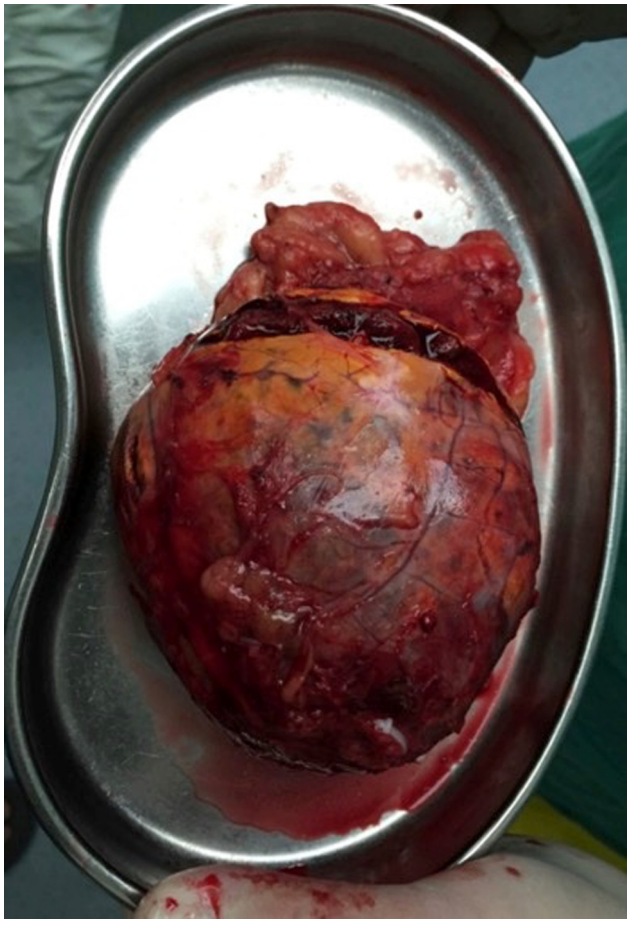

Figure 2. Resected adrenal gland showing a smooth surface and consisting of multiple dilated vascular channels.

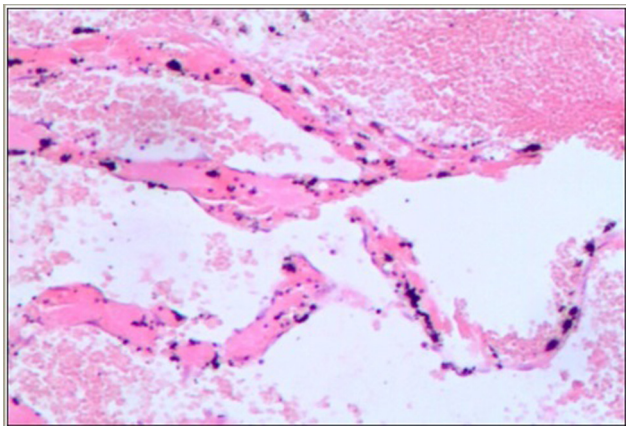

Figure 3. Optic microscopic image showing blood filled dilated vascular spaces with single lining of endothelial cells (hematoxylin eosin staining, 200×).

bleed. For those masses measuring 4–6 cm, history of extra adrenal malignancy, hormone function of tumor, patient's age and physical condition should be taken into consideration. Other indications for surgery include mass effect type symptoms from neighboring organs. Several open techniques have been described, including transabdominal, flank and posterior approaches. The laparoscopic adrenalectomy is a safe and effective approach for removal of benign lesions. However, we should prepare for conversion to an open surgery if it was found with incapability of accomplishing the procedure with laparoscopic technique. In our case, the laparoscopic approach was undertaken after careful preparation and completed uneventfully with blood loss of 20 ml. It seems that the laparoscopic surgery is a reasonable option in treating this particular case.

Conclusion

In summary, we reported a case of nonfunctional adrenal cavernous hemangiomas discovered by imaging studies incidentally. Although the adrenal cavernous hemangiomas is rare, benign and a laparoscopic approach in experienced hands was a safe and effective option, we suggest individualizing the decision based on patient factors, tumor factors, and the experience of the operator.

Conflict of Interest Statement

None declared.

Acknowledgment

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