Adrenal cavernous haemangioma

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ABSTRACT

The adrenal gland is a rare location for haemangioma. Approximately 52 surgical cases have been reported in the literature. We report a huge non-functioning adrenal haemangioma presenting in a 50-year-old woman with flank pain. This was illustrated by computed tomography. It was surgically removed and diagnosed postoperatively as adrenal haemangioma. Although rare, adrenal haemangioma should be included in the differential diagnosis of adrenal neoplasms. The risks of haemorrhage, necrosis and thrombosis impose in the majority of cases surgical excision, particularly in tumours more than 3 cm in diameter.

Keywords: adrenal gland, adrenal haemangioma, adrenal tumour

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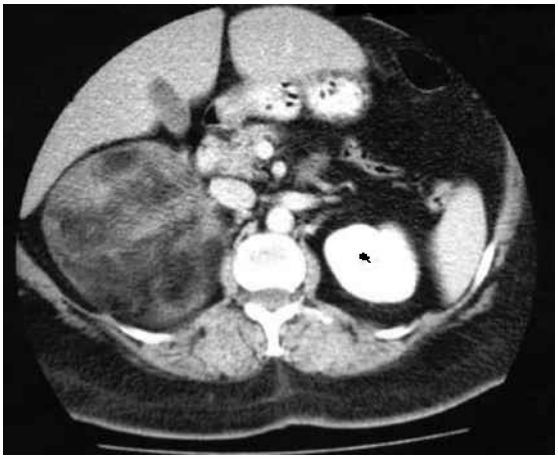

Fig. 1 Contrast-enhanced axial abdominal CT image (portovenous phase) shows heterogeneous spotty peripheral enhancement of a mass lesion in the right adrenal gland suggestive of adrenal cavernous haemangioma. Histopathological examination confirmed that this mass was a cavernous haemangioma of the adrenal gland.

INTRODUCTION

Cavernous haemangiomas most commonly affect the skin and liver. Adrenal gland cavernous haemangioma is an extremely rare benign vascular tumour composed of angioblastic cells that is mostly discovered incidentally on ultrasonography, computed tomography (CT), or magnetic resonance (MR) imaging. Johnson and Jeppesen reported the first case of surgical resection of adrenal haemangioma.[1] Only 52 surgical cases have been reported in the literature.[1-4] These adrenal haemangiomas usually present with chronic flank pain or as an incidental radiological finding.[5,6] We report a huge non-functioning cavernous haemangioma in a patient who presented with flank pain and was illustrated by CT. It was surgically removed and diagnosed postoperatively as adrenal haemangioma.

CASE REPORT

A 50-year-old woman complained of dull right flank pain. Physical examination revealed a nonpulsatile mass in the right upper quadrant. Ultrasonography showed a large soft tissue tumour in the upper part of the right kidney. Radiograph of the abdomen was normal. Abdominal CT showed a well-circumscribed, heterogeneous, retroperitoneal mass located at the upper pole of the right kidney, which was 10 cm in diameter, clearly demarcated from the right lobe of the liver with enhancement of intravenous contrast (Fig. 1). The abdominal CT was performed in the portovenous phase. Although the diagnosis of haemangioma is usually facilitated by additional phases, in this case, no arterial or delayed phases was performed. There were no abnormalities suggesting endocrine dysfunction on either physical or biochemical tests. Plasma and urinary steroid and catecholamine levels were within the normal range and hypertension was not observed. The non-specific radiological features, the vague clinical presentation, in addition to the malignant tumour, which could not be excluded due to its huge size, led to mandatory surgical resection.

After a right subcostal transperitoneal incision, right adrenalectomy was performed. The mass was found to be smooth and well-delineated. An adrenal tumour approximately 400 g in weight was opened, and found to have a cystic cavity with haemorrhage and necrosis. Histopathology revealed a cavernous haemangioma that ruptured within the adrenal gland, and there were no signs of malignancy. Immunohistochemistry with anti-CD34 antibody confirmed the diagnosis of cavernous haemangioma of the adrenal gland. The patient did well postoperatively and was discharged after five days.

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DISCUSSION

The increase in the detection rate of the adrenal masses is due to the widespread use of the modern imaging techniques. While adrenal tumours are relatively common, adrenal haemangiomas are extremely rare and pose problems in the differential diagnosis, because preoperatively, they can be confused with adenomas or malignant tumours of the adrenal gland, and their preoperative diagnosis is very difficult.[7] Cavernous adrenal haemangiomas are usually unilateral, become apparent in the sixth to seventh decade of life, with a 2:1 female-to-male predilection.[8] When patients come to clinical attention, they present usually with a large tumour. They are usually non-functioning tumours, as only two published reports described functioning adrenocortical haemangioma.[9,10] These rare benign tumours are usually discovered as incidental radiological findings or as a result of dull pressure- and mass-related symptoms. Spontaneous life-threatening haemorrhage from adrenal haemangioma has been reported.[8]

It is very difficult to distinguish a large adrenal haematoma from a malignant tumour; the final diagnosis has to be made by histological study of the resected specimen. The clinical picture is not specific and precise preoperative diagnosis remains difficult, even though the tumour shows some characteristic radiological findings.[11-14] In up to two-thirds of cases, radiographs show speckled calcification throughout the entire neoplasm.[8] Generally, CT and MR imaging are useful for the diagnosis of haemangioma.[6,11-14] Contrast-enhanced CT displays a characteristic peripheral patchy enhancement and highly dense peripheral rim. This pattern of peripheral spotty contrast enhancement with centripetal enhancement is crucial for diagnosing adrenal haemangioma.[13] When the haemangioma is seen as only a thin-rim enhancement without centripetal enhancement, the preoperative diagnosis is very difficult because this pattern is seen in other adrenal tumours. Marked hyperintensity on T2-weighted images and focal hyperintensity on T1-weighted images that showed focal haemorrhage and calcification may be seen. These findings can also be seen in other adrenal tumours and are not pathognomonic of adrenal haemangioma. However, observing the combination of these findings can lead to the correct diagnosis.[13,14] Angiography usually reveals peripheral pooling of contrast that persists well into the venous phase of the study.[15]

The indications for resection of this rare neoplasm are to relieve the mass-effect-type symptoms, to exclude malignancy, and to treat complications such as haemorrhage.[8]

The best way to determine the surgical indication is based on tumour size as determined by CT or MR imaging. Deckers et al reported that it is safe to conservatively manage tumours smaller than 3.5 cm in size by regular follow-up imaging.[12] In general, these tumours are large and most reported cases were treated with open surgery.[2,3] Removal of the tumour via retroperitoneoscopic procedure may be used.[4] In conclusion, although rare, adrenal haemangioma should be included in the differential diagnosis of adrenal neoplasms. The risks of haemorrhage, necrosis and thrombosis impose in the majority of cases surgical excision, particularly in tumours more than 3 cm in diameter.

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