Rare disease

Adrenal haemangioma

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Summary

Adrenal haemangioma is a rare, benign, non-functioning neoplasm. Haemangiomas are tumours mainly affecting the liver. In 1955, Johnson and Jeppesen described the first adrenal cavernous haemangioma. Here the authors report a large adrenal haemangioma presenting in a 75-year-old woman who had experienced left flank pain for 5 months. Laboratory examinations and the plasma levels of tumour markers in the patient were within normal limits. Imaging with ultrasound and CT showed a heterogeneous $15 \times 14 \times 18$ cm mass located in the left adrenal gland. The tumour showed irregular peripheral enhancement after bolus intravenous injection of contrast medium. The mass was removed surgically and histopathology revealed infarcted cavernous haemangioma. No signs of malignancy were detected. Although rare, haemangioma should be included in the differential diagnosis of adrenal neoplasms.

BACKGROUND

Haemangiomas are tumours mainly affecting the skin and liver. Adrenal gland haemangiomas are rare and often diagnosed after surgery with histologic examination, but rarely

before. Since the first adrenal cavernous haemangioma was reported by Johnson and Jeppesen,¹ approximately 57 cases have been reported. Here we report a large non-functioning cavernous haemangioma in a patient who presented with

Figure 1 Abdominal CT scan left adrenal mass.

Figure 2 Large adrenal haemangioma.

left flank pain, as illustrated by a CT scan. It was surgically removed and diagnosed as adrenal haemangioma.

CASE PRESENTATION

A 75-year-old female being treated for hypertension, complained of vague abdominal pain of the left upper quadrant for 5 months, and this was accompanied by weight loss for 9 months. Physical examination revealed a soft abdomen with a large non-tender mass in the left side of the abdomen, of around 10×9 cm.

INVESTIGATIONS

Laboratory examinations, plasma levels of free metanephrine and 24-h urinary excretion of catecholamines and total metanephrines in the patient were all within normal limits. An abdominal CT scan showed a well capsulated retroperitoneal, heterogeneous mass of around 15×14×18 cm. The tumour showed irregular peripheral enhancement after bolus intravenous injection of contrast medium. No lymph nodes were observed (figure 1). Although the MRI usually give more information about this mass and it will help in the diagnosis of this case before surgery, we are convinced after doing a CT scan, which showed that the large size of the tumour is to be surgically removed. An FDG-PET usually help preoperatively to elucidate the functional features of the mass, But unfortunately it is not available in our hospital.

DIFFERENTIAL DIAGNOSIS

Non-functioning adrenal neoplasms. Malignancy could not be excluded clinically.

TREATMENT

Because of the large size of the tumour and the fact that malignancy could not be excluded clinically, surgical resection was mandatory. The patient underwent a left adrenalectomy through a left subcostal incision. The patient recovered well postoperatively and was discharged 5 days later. The tumour was well delineated and smooth, and approximately 2230 g in weight (figure 2).

OUTCOME AND FOLLOW-UP

Histopathology revealed an infarcted cavernous haemangioma, with no signs of malignancy.

DISCUSSION

Adrenal haemangiomas are rare, benign, non-functional tumours, with only three cases of hormone-secreting adrenal haemangiomas reported to date.²⁻⁴ Affected individuals are usually between 50 and 70 years of age, and there is a strong predilection for females. Adrenal haemangiomas are usually discovered as incidental radiological findings after abdominal imaging for various other reasons (incidentaloma) or as a result of dull pressure and mass-related symptoms. However, spontaneous retroperitoneal life-threatening haemorrhages from adrenal haemangioma have been reported in two cases.^{5 6} On microscopic analysis, adrenal haemangiomas are usually cavernous and contain peripheral dilated vascular spaces, the larger of which contain central necrosis, calcification and haemorrhages. It is difficult to distinguish large adrenal haemangiomas from malignant tumours preoperatively. In most cases, the final diagnosis is made by histopathology after

surgical resection. However, there are some radiological features that raise the suspicion of adrenal haemangioma. The ultrasound features are not specific, not permitting the differentiation of haemangiomas from other benign lesions.⁶ CT scans usually display a characteristic irregular peripheral enhancement with progression to the centre of the tumour. This pattern is similar to that of liver haemangioma. A CT scan is able to demonstrate calcification in the tumours, and calcification is one of the characteristics of adrenal haemangiomas appearing as a speckled pattern throughout the entire mass. This pattern is related to multiple phleboliths that are located in the dilated vascular spaces. However, calcification can occur in other adrenal tumours, including pheochromocytoma, carcinoma and adenoma. Therefore, this finding is suggestive but not pathognomonic of adrenal haemangioma.^{6 7} MRI has proven to be the best diagnostic tool for adrenal haemangioma to date. The most characteristic finding is peripheral spotty and centripetal enhancement on dynamic studies. Marked hyperintensity on T2-weighted images in combination with focal hyperintensity in T1-weighted images indicates areas of calcification and haemorrhage that are associated with adrenal haemangiomas.^{6 7}

It is important that cavernous haemangioma should be included in the differential diagnosis of adrenal neoplasms. Cavernous haemangioma requires conservative treatment by periodical follow-up if the tumour is small and asymptomatic. Surgical resection is required for excluding malignant disease, removing pressure-related symptoms, and precluding spontaneous haemorrhaging. Laparoscopic resection of adrenal tumours has become the standard procedure. However, in general, these tumours are large and most reported cases have been treated with open surgery.

Learning points

- Although rare, adrenal haemangioma should be included in the differential diagnosis of adrenal neoplasms. Surgical resection is required for excluding malignant disease, removing pressure-related symptoms and precluding spontaneous haemorrhaging. Laparoscopic resection of adrenal tumours has become the standard procedure when possible.

Competing interests None.

Patient consent Obtained.

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