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LETTERS TO THE EDITOR

Clinical-scientific notes

Lymphangioma: an unusual cause for a non-functioning adrenal mass

Elucidation of the nature of large cystic adrenal masses mandates exclusion of malignancy, in addition to evaluation of secretory function. This case report describes the clinical history, imaging and pathology of a very rare cause of a large cystic adrenal mass, the adrenal lymphangioma.

A 23-year-old woman presented to the emergency room with sudden onset of right upper quadrant abdominal pain and loin tenderness. Physical examination demonstrated normal blood pressure and no features of hypercortisolism or hyperandrogenism.

Computed tomography (CT) showed a lobulated, septated mass arising from the right adrenal measuring 52 × 25 × 27 mm, with fluid attenuation and no enhancement (Fig. 1a). Serum electrolytes and androgens were normal, as were 24-h urinary catecholamines (metanephrine, normetanephrine, adrenaline and noradrenaline) and cortisol levels.

Surgical resection was undertaken using a laparoscopic approach. The multiloculated mass measured 37 × 27 × 22 mm, weighed 12.7 g, with cystic areas filled with clear gelatinous and haemorrhagic material (Fig. 1b). Postoperative recovery was uneventful.

Microscopy demonstrated an adrenal lymphangioma. The adrenal cortex and medulla were distorted by large

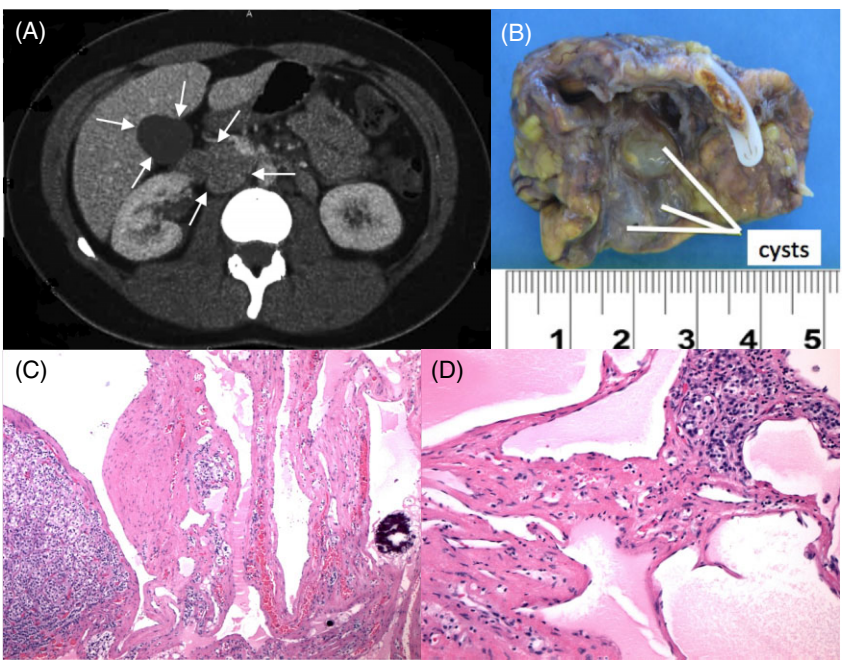

Figure 1 (A) Abdominal computed tomography showing a lobulated right adrenal mass arising from the right adrenal gland. (B) Excised mass weighing 12.7 g. (C) Haematoxylin and eosin stained section at ×40 magnification. (D) Haematoxylin and eosin stained section at ×100 magnification.

fibrous-walled cysts, lined by a single flattened layer of attenuated cells (Fig. 1c,d) without evidence of cytological atypia. There were lymphatic vascular channels, separated by fibrous material; some contained aggregates of adrenal cells. Immunoperoxidase staining showed positive staining for factor VIII and D2-40 immunostain; epithelial markers (AE1/AE3, CAM 5.2) and CD34 were negative.

Cystic adrenal masses have an incidence approximating 0.06% of the general population.[1] Within the differential diagnosis of these lesions, adrenal lymphangiomas are a very rare cause. Lymphangiomas are benign tumours of the lymphatic system, arising from malformations originating in embryogenesis.[2] They are characterised by thin-walled, cystic lesions lined by endothelial cells and filled with lymph fluid.[2] The literature suggests that adrenal lymphangiomas are more frequently right-sided, with a female predominance.[3]

While most adrenal lymphangiomas are clinically asymptomatic,[3] patients can present with flank pain, gastrointestinal symptoms or a palpable mass, relating to the size and position of the cyst.[4] The lack of distinctive symptoms and laboratory findings make preoperative diagnosis challenging.[5] The acute pain presentation of this case may have occurred due to bleeding into a cyst.

Adrenal lymphangiomas on CT imaging are characteristically non-enhancing, hypodense masses with an appearance indistinguishable to other cystic adrenal lesions, including cystic pheochromocytoma and adrenal cortical carcinoma.[6] Surgical resection is usually necessary to distinguish these benign lesions from potentially aggressive malignant lesions, in addition to relieving symptoms.[3]

A possible diagnosis of a lymphangioma should be considered when a hypodense, non-enhancing cystic, non-functioning adrenal mass is found in youth or middle age. Histological diagnosis of non-functioning adrenal masses must be undertaken with care, preferably in a unit specialising in endocrinology or endocrine surgery. As the differential diagnosis of larger non-functioning masses includes adrenal or metastatic carcinoma, excision biopsy is preferred, rather than fine-needle aspiration biopsy.[3,4] Due to the rarity of these tumours, their natural history is not known. Whether these tumours may be observed (after evaluation of functional status and exclusion of malignancy) is not known.

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Recurrence of Carney complex atrial myxoma causing embolic stroke

Carney complex is a rare genetic disease that can lead to the formation of multiple atrial myxomas. These tumours have the propensity to fragment, causing strokes and other embolic events. We present the case of a patient who suffered a stroke due to recurrence of a Carney complex atrial myxoma.

A 58-year-old woman presented to the emergency department following a collapse with no associated loss of consciousness. In the department, she was confused and agitated, and reported blurred vision. A neurological examination revealed a left-sided motor deficit, visual agnosia and perseveration.

The patient had a known diagnosis of Carney complex. She had previously undergone surgical excision of myxomas of her left atrium, bowel and uterus, as well as