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Adrenal solitary fibrous tumor: A case report

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ABSTRACT

Solitary fibrous tumor (SFT) is a rare spindle cell neoplasm, originally described in the pleural cavity. It may also occur in extrapleural organs like adrenal gland. We present a case of a fifty two-year old Arab man who has been admitted in urology department for right lumbar pain. A suspect malignant adrenal mass has been detected. He underwent right adrenalectomy and histologically it was SFT. The follow-up three years after surgery is unremarkable. The prognosis of SFT after surgery is unpredictable. A long term follow-up is mandatory to detect local recurrence or distant metastasis.

Introduction

Solitary fibrous tumor (SFT) is a rare spindle cell neoplasm, originally described in the pleural cavity. But it may also occur in extrapleural sites of the body[1]. Solitary fibrous tumors involving the adrenal gland are extremely rare. According to a computed search of the medical literature, only seven cases of adrenal SFT have been reported to date. The clinical course of this tumor is indolent. A correct diagnosis is mandatory for a best management of the solitary fibrous tumor patients. The aim of this case report is to present the first adrenal SFT revealed by lumbar pain.

Case presentation

A fifty two-year old Arab man without previous medical history has been admitted in our department for urology. He has been complaining of right moderate lumbar pain for six months but not any hematuria, dysuria or fever. Physical examination was normal. His temperature was 37.4 °C, his blood pressure was 125/84 mmHg, and his pulse rate was regular at 76 beats per minute. On laboratory values, white blood cell count was 7.5×10^3/ml, red blood cell count 4.1 × 106/mL, hemoglobin 13.4 g/dl, platelets 285×10^3/ml, creatinine 78μmol/l, sodium 128 mEq/l, potassium 4 mEq/l, urinary creatinine 11.7 mmol/24h, normetanephrines 207nmol/creat, metanephrines 92 nmol/creat. Urines examination showed no leukocyturia or bacteriuria.

An abdominal ultrasound revealed a right heterogenous hypoechoic adrenal mass measuring 116 × 91 mm. For a best characterization of the mass he underwent a computed tomography (CT) scan. The CT scan showed a well delineated, lobular and encapsulated right adrenal mass measuring 110 × 100 × 90 mm (Fig. 1A). It was spontaneously heterogeneous hypodense and contrast-enhanced heterogeneously with a necrosis area at the lower pole. This mass was displacing the right kidney inferiorly but did not show invasion into any adjacent structures (Fig. 1B). The ipsilateral adrenal gland was normal. The patient underwent right adrenalectomy. He was asymptomatic and no recurrence of the tumor has been detected during three years of regular clinical and radiological follow-up.

Histology findings

Histological examination focused on a mesenchymal tumor proliferation well circumscribed by a peripheral thin and intact fibrous capsule surrounded by a laminated but normal adrenal gland. This consisted of spindle-shaped cells arranged either in short bundles, storiform or in most of cases without pattern distribution (Fig. 2A; 2B). The general aspect was heterogeneous with densely cellular areas alternating abruptly with hypocellular of hyaline fibrosis, oedematous or even mixoid areas (Fig. 2C). The whole was irrigated by branched vessels which were sometimes hyalinised or hemangiopericytic vessels. The nuclei were elongated or oval without mitotic activity (< 1 mitotic figure per 10 High Power Fields).

The immunohistochemical staining exhibited a net and diffuse

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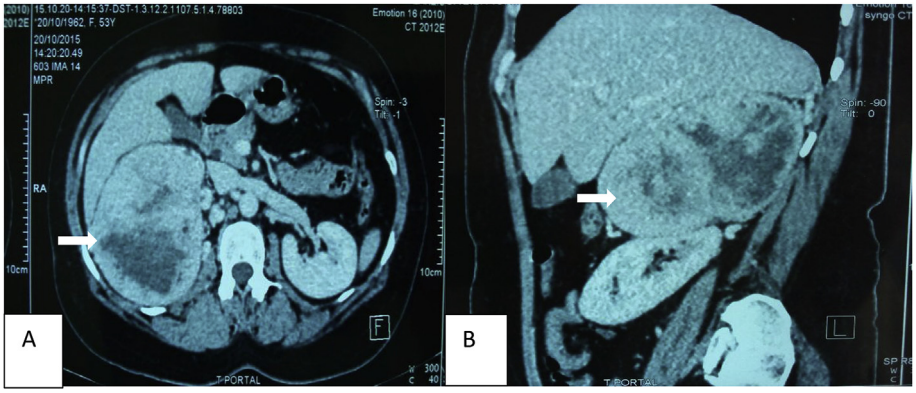

Fig. 1. A: Axial contrast-enhanced CT image showing a circumscribed right adrenal mass with heterogeneous enhancement and a necrosis area (arrow) B: Sagittal contrast-enhanced CT scan demonstrating the repression of right kidney upper pole by the adrenal mass (arrow).

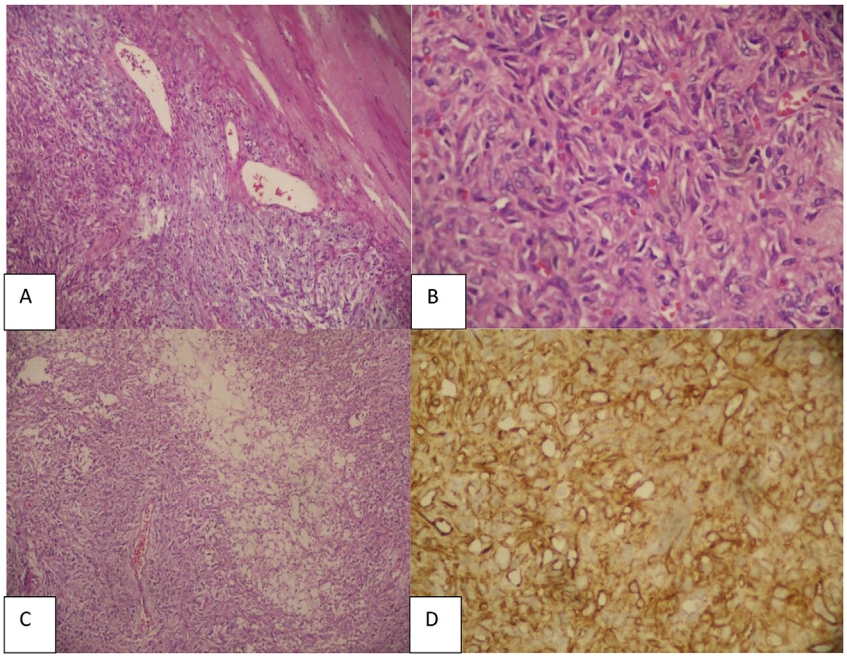

Fig. 2. A: Well limited spindle-shaped cells tumor (hematoxylin and eosin × 200). B: Spindle-shaped cells in "patternless" distribution (hematoxylin and eosin × 400). C: Cellular proliferation of densely cellular areas alternating with hypocellular areas (hematoxylin and eosin × 200). D: Diffuse staining for CD34 (immunohistochemical × 400).

positivity for CD34, a focal positivity for Actine and negativity for Desmine, S-100 protein, HMB 45 (Fig. 2D).

Discussion

Clinical feature

Solitary fibrous tumor is a mesenchymal neoplasm originally described in the pleura, but thereafter reported in several anatomic sites. SFT is most often incidental finding mass during health screening for other diseases. It occurs as a slowly indolent growing mass but symptoms may be associated because of the pressure effects on adjacent organs[2]. Among the seven adrenal SFT reported in the literature none have been revealed by lumbar pain. This is the first case revealed by lumbar pain.

Diagnosis and treatment

In this patient, the CT scan showed a well delineated, and encapsulated right adrenal mass. Microscopic structure is composed of spindle-shaped cells with densely cellular areas alternating abruptly with hypocellular areas. On immunohistochemistry, the tumor showed strong and diffuse positivity for CD34. Macroscopically, conventional SFT is usually a well-circumscribed and often capsulated mass[2]. Histologically, SFT has ovoid or spindled fibroblastic cells arranged in "patternless" distributions while immunohistochemically it typically shows diffuse and strong CD34 positivity[3]. Our patient underwent right adrenalectomy. Wide surgical resection represents the gold standard therapy for SFT[2,3].

Prognostic

In this patient there is neither recurrence nor metastasis after three years follow-up. On histology findings we do not have high likelihood of aggressive behavior[4]. However the prognosis of SFT is unpredictable even if in our patient there were not atypical histological features. It is not strictly depend on histologic features. Local recurrence or distant metastatic may occur despite adequate negative surgical margins[5].

Conclusion

Adrenal solitary fibrous tumor is a rare tumor. It indolent character makes it being often asymptomatic incidental finding. However adrenal SFT may also be revealed by lumbar pain. The prognosis after surgery is substantially unpredictable because does not depend only on histological features. A long-term follow-up is then required to detect local

recurrence or distant metastasis.

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