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CASE REPORT

Detection of a unicentric type of Castleman-like mass at the site of adrenal grand: A case report and review of literature

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Abstract

We present a case of adrenal CD in a 26-year-old female. The patient was referred to our hospital because of left flank pain for 1 wk. A computed tomography scan revealed a 4 cm × 3 cm well-defined mass, considered as a paraganglioma. A preoperative diagnosis of left adrenal neoplasm and urinary tract infection was made. The patient underwent anti-inflammatory therapy followed by an open operation to remove the mass in the left adrenal. Through analysis of the morphological pattern and immunohistochemical markers, a diagnosis of CD was made. During the 12-mo follow-up, there was no evidence of metastasis or recurrence. This case reminds clinicians that CD should be considered in the evaluation of an adrenal mass. Surgery is suggested for its therapeutic management.

Key words: Adrenal; Castleman disease; Pathological classification; Therapeutic management

Core tip: Castleman disease (CD), also known as giant lymph node hyperplasia or angiofollicular lymph node hyperplasia, is a highly heterogeneous clinicopathological entity belonging to the family of lymphoproliferative disorders. CD is commonly found in the mediastinum.

Castleman-like masses in the adrenal grand are extraordinarily rare. We present a case of adrenal CD in a 26-year-old female. Through laparotomy, the tumor was removed. During a 12-mo follow-up, there was no evidence of metastasis or recurrence.

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INTRODUCTION

Castleman disease (CD), also known as giant lymph node hyperplasia or angiofollicular lymph node hyperplasia, is a highly heterogeneous clinicopathological entity that belongs to the family lymphoproliferative disorders[1-3]. The first description of CD was made by Castleman[1] in 1956. As a complex lymphoproliferative disease, CD has been divided into two subtypes depending on its pathological classification, namely the hyaline vascular variant and the plasma cell variant.

The median age of patients was reported to be 43-years-old[4], with a slight female predilection[5]. The distribution of CD was reported as 60% in the thorax, 14% in the cervical area, 11% in the abdomen, and 4% in the axillary region[6-9]. Extrathoracic sites of CD had been reported in the pararenal region. However, adrenal CD is very rare[10].

The final diagnosis of CD at the site of adrenal grand is primarily based on histopathology and immunohistochemistry, although imaging and clinical symptoms are crucial for the diagnosis. The clinical diagnosis of CD is difficult. The primary treatment strategy for CD of the adrenal gland is surgery. We present a case of CD at the site of the adrenal gland in a 26-year-old female.

CASE REPORT

A 26-year-old female was referred to our department because of left flank pain for 1 wk. No additional symptoms were present. Pulse and blood pressure were within the normal range. A physical examination revealed pain on percussion of the left lumbar region.

The biochemical findings were within normal ranges, including lactate dehydrogenase (2.45 mmol/L), albumin (44.4 g/L), creatinine (48 μmol/L), urinary catecholamine excretion, plasma renin activity, and plasma aldosterone. Routine urinalysis showed an increased number of leukocytes (181/μL), and fibrinogen (4.53 g/L) was also increased. Routine blood examination showed decreased hemoglobin (86 g/L) and hematocrit (28.6%). Ferritin (8.82 ng/mL) was low. The results of hormone measurements are listed in Table 1. Abdominal computed tomography (CT) revealed a 4 cm × 3 cm mass on the left adrenal region (Figure 1). No positive findings were revealed on chest X-ray.

A diagnosis of left adrenal neoplasm and urinary tract infection was made on the basis of physical examination, imaging, and laboratory tests.

Through laparotomy, the tumor was removed. The size of the adrenal tumor was approximately 4 cm × 3 cm, with a hard texture and unclear lesion margins. The adrenal tumor was adherent to the peritoneum as well as the renal artery and vein. Microscopically, hematoxylin and eosin staining showed angiofollicular lymph node hyperplasia, interstitial edema, cystic changes and hyperplasia of collagen fibers (Figure 2A and B). Immunohistochemistry staining showed that the tumor cells were positive for CD20, CD79α, Pax5, CD2, CD3, CD5, CD21, CD23 and CD68. The tumor was negative for cyclinD1, Bcl2, CD15, CD30, syn, ck and cga (Figure 2C and D). Through the analysis of morphological pattern and immunohistochemical markers, a diagnosis of CD was made.

During a 12-mo follow-up, there was no evidence of metastasis or recurrence.

DISCUSSION

CD was first reported by Castleman in 1954 in a patient with a large mediastinal mass. It is a highly heterogeneous clinicopathological entity belonging to the family of lymphoproliferative disorders[1-3].

The etiology and pathogenesis of CD is largely unexplored. The crucial function of interleukin-6 (commonly known as IL-6) has been revealed in lymphovascular proliferation and systemic symptoms of CD. Some studies showed that abnormal expression of IL-6 in CD may explain several clinical manifestations of CD, including autoimmune manifestations and hyperglobulinemia, acute-phase reaction, and hypodermic microcytic anemia[11]. Other researchers reported that the constitutive over-expression of IL-6 in mice led to the CD phenotype. Therefore, IL-6 may play an important part in the pathogenesis of CD, and anti-IL-6 receptor antibody could be a therapeutic strategy for CD[12].

Histological evaluation plays a vital role in the exact diagnosis of CD. Histologically, the disease is primarily subclassified into three types: Hyaline vascular type, plasma cell variant, and mixed type. The characteristics of the hyaline vascular type include considerable enlarged lymphoid follicular proliferation at various levels of maturity[13]. In addition, lymphoid follicles are normally scattered throughout the tissue. The plasma cell type is almost related to the multicentric form of CD. This type shows less vascularity and is characterized by sheets of mature plasma cells within the interfollicular tissues surrounding larger germinal centers[14].

CD is ubiquitous but predominates at lymph node sites. The localized form is the most frequent. Localized CD of the hyaline vascular type is commonly asymptomatic. As in this case, adrenal CD has no specific presenting symptoms, including flank pain and hypertension[15]. In another study, the clinical manifestations of adrenal

Table 1 Results of hormone measurements

Hormone	Result	Normal or abnormal
Cortisol	285.10 nmol/L	normal
ACTH	23.90 pg/mL	normal
ALD (lying position)	410.20 pmol/L	normal
ALD (standing position)	527.90 pmol/L	normal
DA	25.53 ng/mL	normal
E	16 ng/mL	normal
NE	38.68 ng/mL	normal

ACTH: Adrenocorticotrophic hormone; ALD: Aldolase; DA: Dopamine; E: Epinephrine; NE: Norepinephrine.

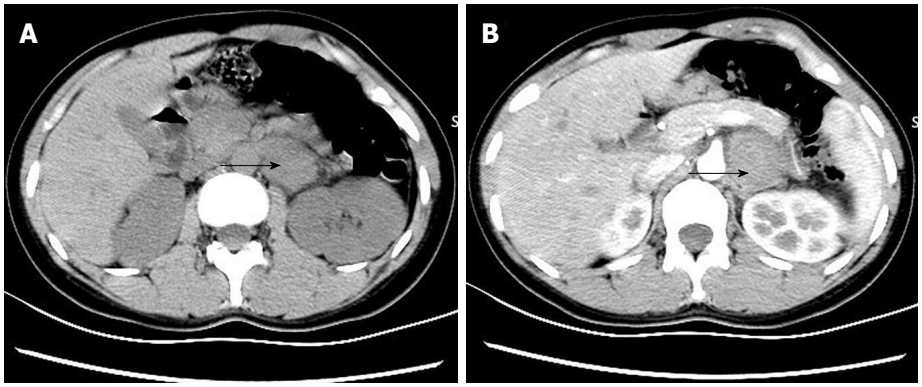

Figure 1 Abdomen computed tomography-scan demonstrating a 4.2 cm mass in the left adrenal. A: Computed tomography (CT) plain scan showed the mass in the left adrenal; B: CT enhancement scan showed the mass in the left adrenal with no enhanced uniform.

CD patients included poor appetite and weight loss[8]. However, the plasma cell type of localized CD and multicentric CD present systemic symptoms, including fever, night sweats, peripheral lymphadenopathy, weight loss, anemia, hepatosplenomegaly, and renal insufficiency. Disseminated CD has been described in association with polyneuropathy, organomegaly, endocrinopathy, monoclonal gammopathy and skin changes (known as POEMS) syndrome that results from a disorder of underlying plasma cells. It often displays aggressive behavior, including autoimmune anemia, sarcoidosis and amyloidosis[16].

The radiological appearance of CD is nonspecific. A homogeneous hypoechogenic formation is commonly shown on ultrasound[17]. Masses that are smaller than 5 cm have a homogeneous vascular appearance on CT, while larger masses are often mixed with the central low presence of fibrosis and necrosis[18]. According to the literature, on CT, adrenal CD often is rounded, well circumscribed and possesses some small calcifications without cystic or fatty areas. After intravenous injection of iodinated contrast agent, the tumor did not enhance and the adrenal gland was not visualized[19]. There are rare descriptions of MRI characteristics of CD. The signal characteristics of adrenal CD were similar to those reported for adrenal lymphoma: Hypointense compared with hepatic parenchyma on T1-weighted images and hyperintense on T2-weighted image[20]. Because the hyaline vascular type has three patterns of manifestation on CT, including solitary noninvasive mass, dominant infiltrating mass accompanied by lymphadenopathy and matted lymphodenopathy[21], it is difficult to distinguish CD from other malignant diseases solely on the basis of radiological methods.

Adrenal diseases include classic endocrine syndrome diseases, including Cushing syndrome, hyperaldosteronism and pheochromocytoma. Tumors of the adrenal gland may be accompanied by abdominal pain or an abdominal mass. The diagnosis of these diseases requires cautious endocrine assessment, and in many patients adrenal anatomy must be defined using adrenal imaging methods. If the examination of pulse, renal function, serum cortisol, electrolytes and urinary catecholamines are normal, the adrenal tumor may be diagnosed as nonfunctional. Under these circumstances, adrenal CD should be considered during the process of evaluating an adrenal mass[22]. Nevertheless, diagnosis of adrenal CD relies on histopathological examination.

Localized CD is primarily benign[23]. Localized CD usually requires surgical excision of the enlarged lymph node without additional treatment and it always responds well to surgical excision[24]. A systematic review of the role of surgical resection in localized CD showed that surgical resection was the most effective treatment for localized CD[25]. All general symptoms disappear after surgery and there were almost no relapses postoperatively[26].

CD is a rare disorder that is hard to diagnose early because of its nonspecific manifestations and radiological characteristics. Adrenal CD is extraordinarily rare

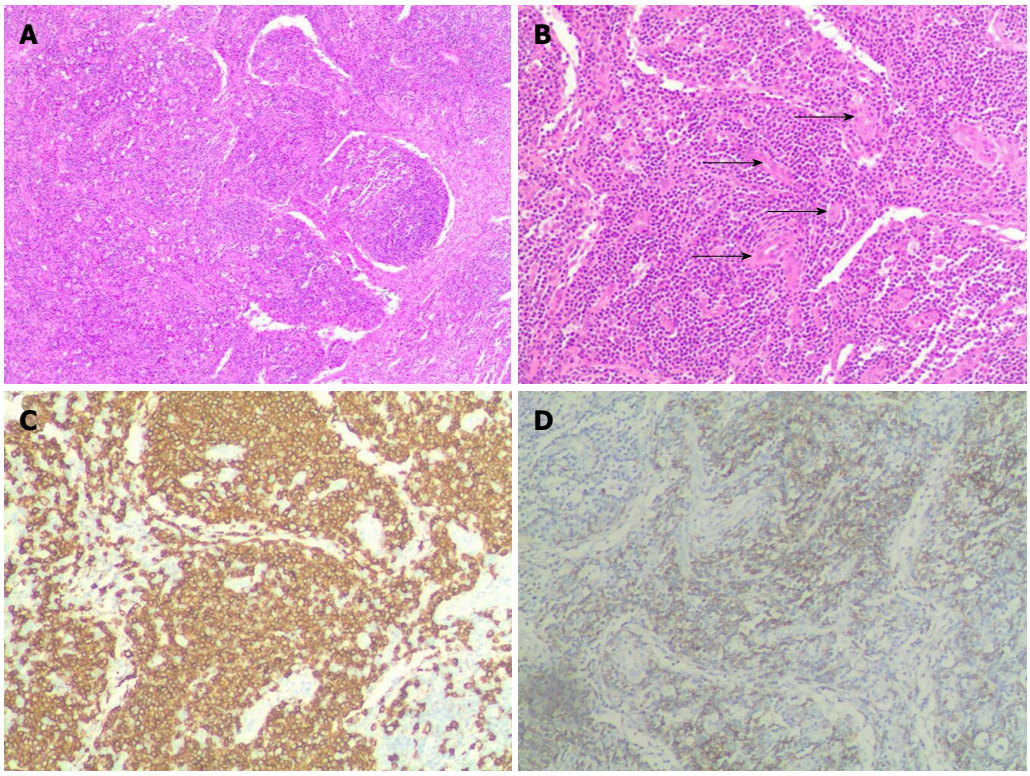

Figure 2 Pathological diagnosis of castleman disease was made by the analysis of morphological pattern and immunohistochemical markers. A: hematoxylin-eosin (HE) (40 × magnification) staining showed a large number of enlarged lymphoid follicle scattered in the distribution; B: HE (100 × magnification) staining showed hyperplasia and wall thickening of the small blood vessels; C: Immunohistochemical staining (100 × magnification) showed CD20 was ubiquitously expressed; D: Immunohistochemical staining (HE 100 × magnification) showed CD21 was ubiquitously expressed. CD: Castleman disease; HE: Hematoxylin-eosin.

and should be considered in the process of evaluating an adrenal mass.

ARTICLE HIGHLIGHTS

Case characteristics

This patient was referred to our department due to left flank pain for 1 wk, with no additional symptoms.

Clinical diagnosis

A 4 cm × 3 cm mass was found in the left adrenal region.

Laboratory diagnosis

The findings of biochemical and hormone measurements were within the normal range.

Imaging diagnosis

Abdominal computed tomography (CT) of this patient revealed a 4 cm × 3 cm mass in the left adrenal region, and CT enhancement scan showed a mass in the left adrenal with no enhanced uniform.

Pathological diagnosis

Through the analysis of morphological pattern and immunohistochemical markers (the tumor cells were positive for CD20, CD79α, Pax5, CD2, CD3, CD5, CD21, CD23 and CD68), a diagnosis of CD was made.

Treatment

The patient was treated by laparotomy.

Related reported

CD was first reported by Castleman in 1954, in a patient with a large mediastinal mass. Adrenal CD is very rare. Interleukin-6 may play an important part in the pathogenesis of CD and anti-interleukin-6 receptor antibody could be a therapeutic strategy for CD.

Experiences and lessons

CD is a rare disorder, for which is hard to achieve early diagnosis due to its nonspecific manifestation and radiological characteristics. Adrenal CD is extraordinarily rare and should be considered in the process of evaluating adrenal mass.

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