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Plasma cell variant of Castleman lymphadenopathy presenting as an adrenal mass

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ABSTRACT

Adrenal masses are commonly found on radiographic studies performed for unrelated reasons. We report on a case of a non-functioning adrenal mass from which a needle biopsy showed a nonspecific infiltrate of polyclonal plasma cells and small lymphocytes. A definitive diagnosis of the plasma cell variant of Castleman lymphadenopathy was made only after surgical excision. While the hyaline vascular variant of Castleman lymphadenopathy has been identified in adrenal glands, this is the first report of the plasma cell variant in an adrenal mass. This case particularly underscores the importance of an excisional biopsy for proper diagnosis.

Introduction

Adrenal masses encompass both benign and malignant diseases and may include adenomas, metastases, pheochromocytomas, adrenocortical carcinomas, myelolipomas, cysts, ganglioneuromas, and other rarer entities.[1] The management of a newly identified adrenal mass begins with an endocrinological evaluation along with radiologic studies to decide whether or not proceed to surgical treatment. An attempt at a pathologic diagnosis via a needle biopsy may be fruitful in patients with a history of malignancy, but in the absence of a pre-existing cancer diagnosis, surgical excision is likely to be required for definitive characterization of the mass.[1]

Castleman lymphadenopathy is a rare but benign cause of an adrenal mass and may involve additional retroperitoneal locations. Castleman lymphadenopathy is classified as unicentric (localized) or multicentric (generalized) on the basis of radiographic and clinical findings, and histologic subtypes include the hyaline vascular, plasma cell and mixed variants.[2] The hyaline vascular variant has been described in association with the adrenal gland previously,[3,4] but in this paper we report on what we believe is the first case of the plasma cell variant of Castleman lymphadenopathy presenting as an adrenal mass.

Case presentation

A 70-year-old woman presented with a transient, vague sensation of abdominal fullness. A computerized tomography (CT) scan of the abdomen and pelvis with contrast showed a mass involving the left adrenal gland with calcifications along the superior margin (Fig. 1, panel A, arrow). The maximum diameter of the mass was 59 mm, and enlarged retrocrural, abdominal aortic, and right inguinal lymph nodes were also identified. Blood tests for estradiol, progestesterone, cortisol, dehidroepiandrostenedione (DHEAS), total testosterone, aldosterone, androstenedione, free normetanephrine, free metanephrines and renin activity showed no abnormalities, consistent with a non-functioning adrenal mass. A CT-guided core needle biopsy of the mass and a neighboring lymph node showed benign lymphoid tissue with an extensive polyclonal plasma cell infiltrate.

A laparoscopic left adrenal resection was subsequently performed for definitive diagnosis. Intra-operative exploration revealed that the adrenal lesion appeared largely to originate from a nodal mass adjacent and adherent to the adrenal gland. The dissection of the mass was particularly difficult related to the adjacent renal vessels, the size of the mass and the presence of peri-mass inflammation and fibrosis. After clipping of the blood vessels, the mass was excised completely.

The resected mass size was 7.2 × 4.0 × 3.8 cm and abutted the

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adrenal gland and posterior soft tissue. Microscopic examination of the tumor showed lymphoid tissue with numerous small to medium size round follicles throughout (Fig. 1, panel B). The germinal centers within many of the follicles were atrophic with hyaline deposits, sclerotic blood vessels and a paucity of small lymphocytes. Follicles showed "onion skinning" of the mantle zones around the germinal centers, and, rarely, "twinning" of germinal centers (Fig. 1, panel C). Sheets of CD138-positive plasma cells that showed polytypic expression of kappa and lambda immunoglobulin light chains dominated the interfollicular/paracortical regions (Fig. 1, panel D). Immunostaining for HHV-8 was negative throughout, and no infiltrating neoplastic lymphoid or metastatic cell populations were identified. Flow cytometry confirmed the polyclonal nature of both B-cell and plasma cell populations. A diagnosis of unicentric Castleman lymphadenopathy was made, and though features of the hyaline vascular variant were present in areas, the dense proliferation of polyclonal plasma cells in the interfollicular areas was most consistent with the plasma cell variant. The absence of systemic symptoms and the absence of HHV-8, excluded multicentric Castleman disease.

Discussion

Castleman lymphadenopathy was first described in the 1950's as a benign lymphoproliferation in the mediastinum.[5] It is defined clinically as unicentric (i.e., localized to a single lymph node region) or multicentric, and classified histologically into the hyaline vascular, plasma cell, and mixed variants.[2] While the hyaline vascular variant has been identified in the adrenal gland and elsewhere in the retroperitoneum,[3,4] this is, to our knowledge, the first report of the plasma cell variant in an adrenal mass and underscores the importance of an excisional biopsy for definitive diagnosis.

Enlarged lymph nodes in the plasma cell variant typically show a mixture of normal and hyperplastic follicles with occasional follicles showing hyaline vascular features. As we describe in the current patient, a prominent, dense interfollicular proliferation of polyclonal plasma cells is invariably present. The unicentric plasma cell variant represents less than 20% of all Castleman disease cases, with a roughly equal incidence in men and women, and patients are frequently asymptomatic or minimally symptomatic as in this case.[2] However, there is clinical and histologic overlap with multicentric disease which is commonly accompanied by systemic symptoms, including fever, night sweats and weight loss, may be associated with a polyclonal hypergammaglobulinema and an increase in inflammatory cytokines and usually requires systemic treatment.[2] Human immunodeficiency virus (HIV) infection seems to predispose to multicentric Castleman disease, often demonstrating co-infection with human herpes virus-8 (HHV-8). The presence of HHV-8 is important to assess as its presence may portend transformation to the plasmablastic variant of Castleman disease or plasmablastic lymphoma, an aggressive malignant neoplasm.

The initial needle biopsy result in this case was nondiagnostic. As the diagnostic considerations for a mass in which polyclonal lymphoid cell populations are present include both benign and malignant processes, an excisional biopsy was critical for definitive diagnosis. Among the potential alternative diagnoses include primary adrenal malignancies, Hodgkin or T-cell lymphomas, metastatic carcinomas or rheumatoid lymphadenopathy, and the distinction among these entities has important ramifications for treatment.

Surgical excision is frequently curative for unicentric Castleman disease, and clinical and radiographic follow up at regular intervals will identify the minority of patients who subsequently relapse or develop systemic disease.[2] As the lesion in our patient was benign, asymptomatic and localized, surgical resection was the definitive treatment. She has been under clinical follow-up every 6 months with no signs of recurrence, and CT imaging studies are planned for every 2–3 years.

Conclusion

We report on a heretofore never-described presentation of the plasma cell variant of Castleman lymphadenopathy as an adrenal mass. This case emphasizes the value of surgical excision for accurate

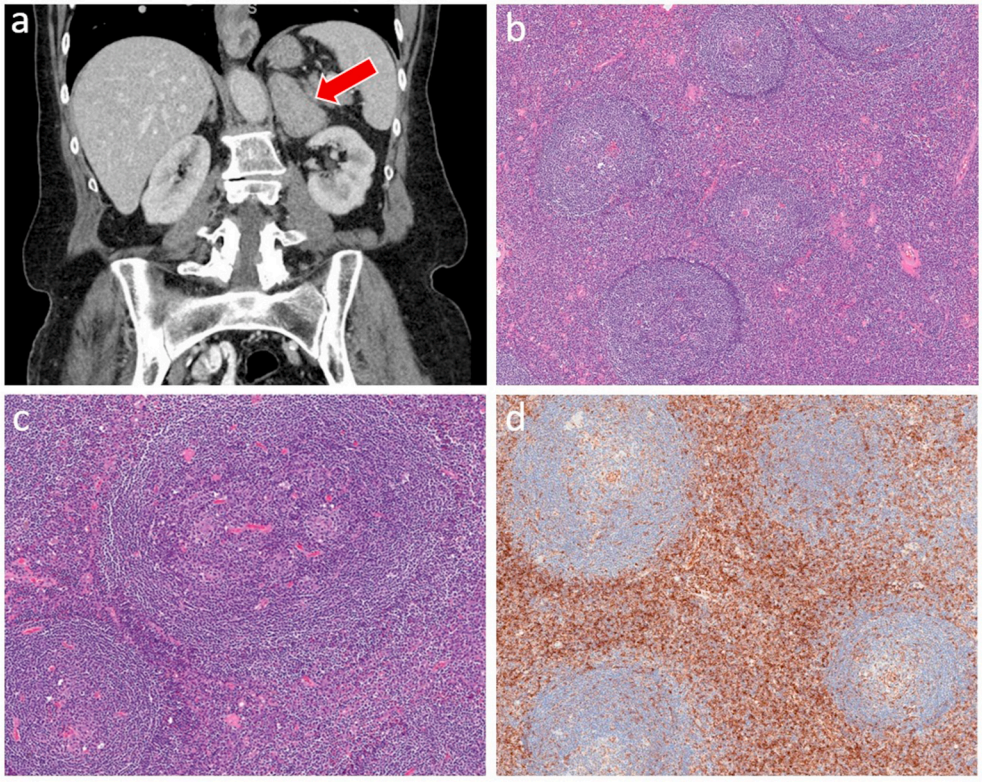

Fig. 1. Radiographic and microscopic appearance of adrenal tumor. a, Computerized tomography image of the abdomen and pelvis with a 59 mm left adrenal mass indicated by the arrow. **b,** Histologic appearance of the adrenal mass, H&E stain, lymphoid tissue comprised of small to medium size, round follicles with largely atrophic germinal centers (original magnification ×200). **c,** Lymphoid follicle showing "twinning" of germinal centers (original magnification ×400). **d,** Immunohistochemical stain for CD138 identifies a dense interfollicular proliferation of plasma cells, which displayed polytypic cytoplasmic immunoglobulin light chain expression (original magnification ×400).

diagnosis and characterization of lymphoproliferative processes.

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Authors' contributions

All authors participated in drafting and reviewing the manuscript, and contributed equally to this work.

Declaration of competing interest

No author has any conflict of interest or competing interest with respect to this work.

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