Primary Hodgkin lymphoma of the adrenal gland: a unique case presentation

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Adrenal "incidentalomas" are commonly found on body imaging, and treatment of these lesions 4 cm-6 cm in size is controversial. Most of these lesions are benign adrenal cortical adenomas. Lymphoma is a rare disease manifestation in the adrenal gland, and the overwhelming majority are metastatic lesions. Hodgkin lymphoma has never been reported as a primary adrenal lesion. We present a very unique case report of a 5 cm adrenal "incidentaloma" that represents the first reported case of primary Hodgkin lymphoma in the adrenal gland.

Key Words: adrenal gland neoplasm, laparoscopy, Hodgkin lymphoma

Introduction

Adrenal lesions are commonly discovered as an incidental finding during CT and MRI for unrelated issues. Adrenal "incidentalomas" are encountered in up to 5% of individuals who undergo body imaging studies,[1,2] and are discovered in nearly 6% of autopsies. Most of these lesions are benign adrenal cortical adenomas.[3] The adrenal gland is also a common location for metastatic disease.[4] Furthermore, up to 25% of patients with non-Hodgkin lymphoma will be found to have adrenal involvement by lymphoma.[5] In contrast, Hodgkin lymphoma only very rarely involves the adrenal gland and in the few reported cases, the adrenal involvement was a secondary manifestation of the disease.[4] To our knowledge, Hodgkin lymphoma has not been reported as a primary tumor of the adrenal gland. We report a unique case of primary Hodgkin lymphoma in the adrenal gland.

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Case report

Presentation and clinical course

A 76-year-old female presented for urologic consultation after an incidental adrenal mass was discovered on CT scan of the abdomen and pelvis

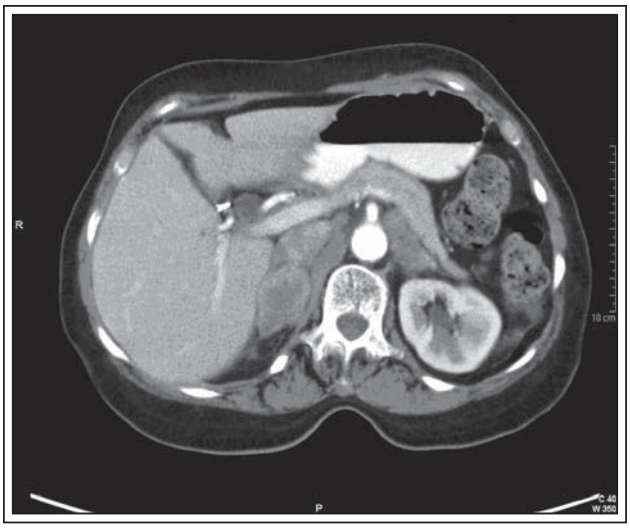

Figure 1. Abdominal CT with IV and PO contrast showing a 4.7 cm right adrenal mass.

performed for a complaint of back pain, Figure 1. There had been no adrenal abnormality noted on a CT scan performed 2 years earlier. Plasma metanephrines, plasma renin activity, serum aldosterone, DHEAS and basic metabolic panel were all normal. A CT guided biopsy of the adrenal mass was performed but was non-diagnostic as it revealed only necrotic tissue and chronic inflammation. A CT scan of the chest revealed indeterminate lung lesions, and a FDG-PET scan showed uptake in these lesions alone. These were excised thoracoscopically and showed granulomatous inflammation. Further metastatic work up showed no evidence of adenopathy in the chest, abdomen or pelvis. The patient underwent laparoscopic right adrenalectomy. Intraoperatively, the right adrenal gland was enlarged, nodular and fibrotic with adhesions to the inferior vena cava. The liver and spleen were normal. Pathologic examination of the resection specimen showed classical Hodgkin lymphoma involving the adrenal gland. A bone marrow aspiration and biopsy were negative.

Pathology

The adrenalectomy specimen weighed 39 gm and measured 5.5 cm x 4.5 cm x 3.0 cm. Sectioning demonstrated a tan pink nodular tumor with marked fibrosis and areas of necrosis as well as a thin rim of compressed normal adrenal gland, Figure 2. On microscopy, there were portions of normal adrenal gland and a nodular infiltrate composed of an admixture of small mature lymphocytes, plasma cells, epithelioid histiocytes and large atypical cells (Hodgkin/Reed-Sternberg cells), Figure 3. Interstitial fibrosis and abundant necrosis were also present. The neoplastic Hodgkin/Reed-Sternberg cells exhibited large irregular often multilobated nuclei with prominent nucleoli, Figure 4. Immunohistochemical stains showed that the large atypical neoplastic cells were strongly positive for CD15 and CD30 and variably positive for the B-cell marker CD20. The background contained numerous CD3 positive T cells and few CD20 positive B cells. These findings are consistent with classical Hodgkin lymphoma. In addition, in situ hybridization for Epstein-Barr virus (EBER) was positive.

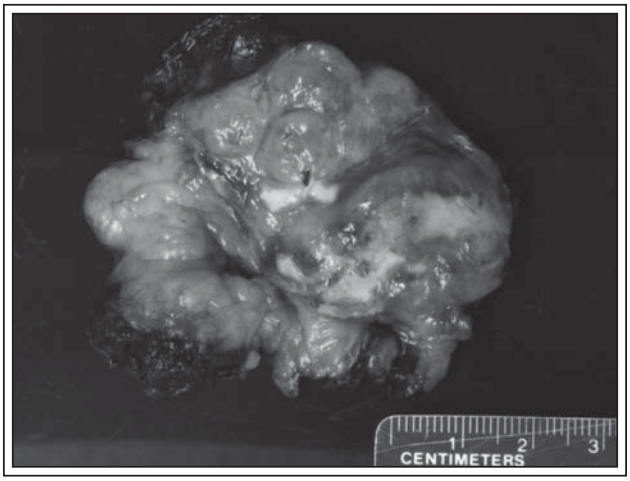

Figure 2. Nodular tumor with fibrosis and areas of necrosis. Note compressed rim of normal adrenal gland (top left).

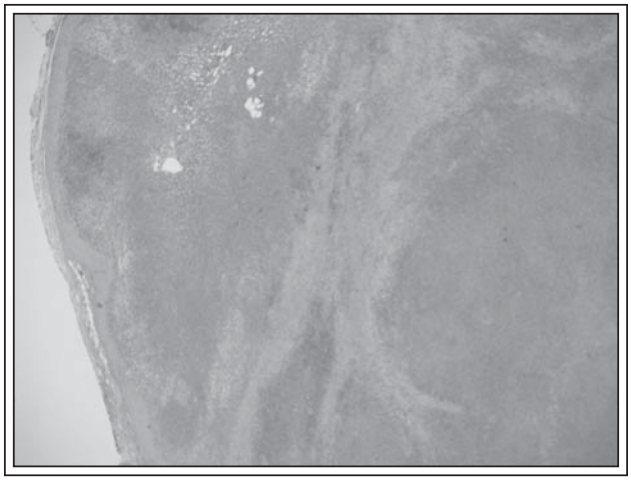

Figure 3. Nodular tumor infiltrate (right) with surrounding fibrosis and normal adrenal gland (left).

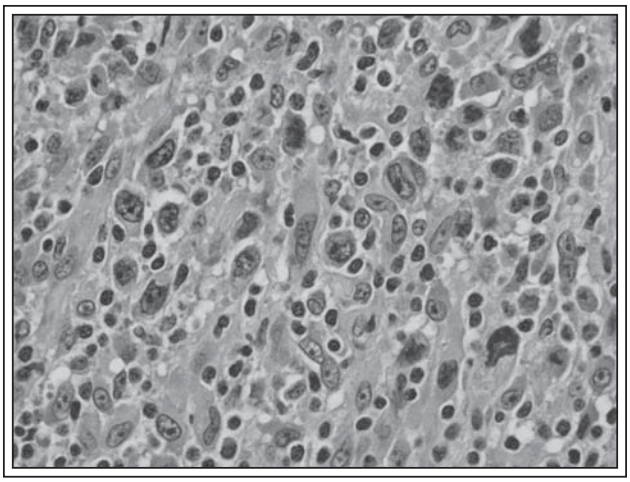

Figure 4. Hodgkin-Reed Sternberg cells with large irregular nuclei and prominent nucleoli. The background is composed of small mature lymphocytes and few plasma cells.

Discussion

Incidental adrenal lesions are commonly found on abdominal CT scans performed for other indications. Although there is a wealth of literature discussing the occurrence of adrenal "incidentalomas", the appropriate management of incidentally discovered adrenal lesions 4 cm to 6 cm in size is controversial.[6-9] Interestingly, this case of primary Hodgkin lymphoma of the adrenal gland presented as an incidental 5 cm lesion on CT scan. Hodgkin lymphoma involving the adrenal gland is very rare and in those few cases where it has been reported, the adrenal involvement was a secondary manifestation of systemic disease. In this case, the primary extranodal location of the tumor, age of the patient and positive test result for Epstein-Barr virus (EBV) suggest that this unusual presentation of primary Hodgkin lymphoma in the adrenal gland may be related to senile EBV positive lymphoproliferative disorders. Oyama et al recently described 22 Epstein-Barr virus-associated B-cell lymphoproliferative disorders (LPDs) occurring in elderly patients without predisposing immunodeficiencies.[10] Interestingly, 15 of the 22 cases showed large atypical cells with an appearance reminiscent of Reed-Sternberg cells. However, in none of these 15 cases were the large atypical Reed-Sternberg-like cells positive for CD15. This is in contrast to our case in which the Hodgkin/Reed-Sternberg cells demonstrated strong CD15 staining, characteristic of Hodgkin lymphoma. Furthermore, the LPDs in Oyama's series presented in a variety of nodal and extranodal sites but none occurred in the adrenal gland. To our knowledge, this is the first reported primary adrenal classical Hodgkin lymphoma. □

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