Case Study of the Month

Clinically Inapparent Bilateral Adrenal Masses Due to Histoplasmosis


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1. Case report

A 50-yr-old, nondiabetic, normotensive man presented with fatigue, body ache, weight loss, and epigastric discomfort for 3 mo. Previous medical history was unremarkable. On examination, he was hemodynamically stable, mild hepatomegaly was noted, and there was no generalized lymphadenopathy.

Lab findings disclosed a hemoglobin of 12.9 gm%, white cell count of 6200 cells/cumm, erythrocyte sedimentation rate (ESR) of 50 mm per first hour, serum sodium 133 mEq/l, serum potassium 4.2 mEq/l, serum bilirubin 0.8 mg%, alanine aminotransferase 45 IU/ml, aspartic aminotransferase 38 IU/ml, serum alkaline phosphatase 410 IU/ml, and calcium of 9.2 mg%. His hormonal and catecholamine levels were normal. Serum cortisol was 25 μg/dl (9–25 μg/dl), dehydroepiandrosterone (DHEA) was 14 μg/dl (133–440 μg/dl). Plasma epinephrine was 23.80 pg/ml (<67 pg/ml), norepinephrine was 238 pg/ml (95–446 pg/ml), 24-hr urinary epinephrine was 2.65 μg/24 hr (1.70–22.40 μg/24 hr), and 24-hr urinary norepinephrine was 35.71 μg/24 hr (12.10–85.5 ug/24 hr).

A work-up for tuberculosis, including enzyme-linked immunosorbent assay (ELISA) and Mantoux...
test, was negative. His human immunodeficiency virus (HIV) serology was negative and chest roentgenogram was normal. A blood buffy coat test and ELISA for histoplasmosis were also negative.

Transabdominal ultrasonography revealed bilateral heteroechoic adrenal masses with mild hepatomegaly. Contrast-enhanced computed tomography (CT) of the abdomen revealed mixed attenuating adrenal masses of size 6 × 3.56 cm and 4.1 × 2.5 cm on the right and left side, respectively. The masses had hypodense areas with rim enhancement (Fig. 1). Magnetic resonance imaging (MRI) revealed bilateral oval, well-defined adrenal mass lesions mildly hyperintense on T2-weighted MRI and homogeneously hypointense on T1 without any evidence of intrallesional fat (Fig. 2).

Whole body F-2-fluoro-D-deoxyglucose positron emission tomography CT (FDG-PET/CT) showed increased FDG uptake in both adrenal masses. The
maximum standardized uptake value (SUV max) was 13.46 in the left and 8.49 in the right adrenal gland (Fig. 3). There was abnormal uptake by a lymph node in the left paratracheal region with a SUV of 3.12; a subsequent chest CT revealed a small 1.1-cm left lower paratracheal lymph node. CT-guided fine needle aspiration cytology (FNAC) of the adrenal lesion was done to characterize the lesion; however, it turned out to be inconclusive.

In view of a large adrenal mass of a size >6 cm and with a suspicion of adrenal metastasis/adrenal tumor, the patient underwent laparoscopic right adrenalectomy plus biopsy of the left adrenal gland. Findings included a 6-cm right adrenal mass full of necrotic caseous material densely adherent to the liver and inferior vena cava (Fig. 4). Frozen section revealed a chronic inflammatory lesion with no evidence of malignancy. Histopathology revealed multiple histiocytes and multinucleated giant cells. Gomori methenamine silver staining demonstrated multiple spores suggestive of disseminated histoplasmosis. Postoperative recovery was uneventful, and the patient was started on oral itraconazole (200 mg twice a day) with low-dose prednisolone (5 mg/day).

2. Discussion

The differential diagnosis of bilateral adrenal masses with fever, loss of weight, malaise, fatigue, and generalized weakness include tuberculosis, histoplasmosis, metastatic carcinoma, and lymphoma [1]. CT features of adrenal histoplasmosis include bilateral symmetric enlargement with preservation of normal contour, peripheral enhancement, and central hypodensity. These features are not specific to histoplasmosis and may occur with disseminated infections like TB, cryptococcosis, and blastomycosis and in metastasis and lymphoma [1]. There is no reliable imaging feature to distinguish between various causes of bilateral adrenal masses [2].

FDG-PET is a useful modality for distinguishing malignant from benign adrenal lesions in patients with incidentally detected adrenal tumors on CT or MRI. There is increased uptake of FDG by malignant adrenal lesions, with a reported sensitivity of 100% and specificity of 80–100% [3]. Intense uptake of FDG can also be observed in inflammatory conditions and infectious diseases because of the increased glucose metabolism of activated inflammatory cells [3]. Umeoka et al [4] reported a case of bilateral adrenal histoplasmosis showing a significantly high uptake of 18F-FDG on PET study. In our case, there was increased FDG uptake in both the adrenals and a small paratracheal lymph node, suggestive of either primary adrenal malignancy or adrenal metastasis.

Percutaneous imaging-guided biopsy of the adrenal gland remains a safe and useful procedure in diagnosing adrenal metastasis, adenomas, and infections in patients with nonspecific features on imaging [5]. FNAB may be inconclusive at times, and an open adrenal biopsy may be indicated. The nondiagnostic biopsy rate of adrenal FNAB ranges from 6–17% in different series, and potential disadvantages include a high false-negative rate, bleeding, or a hemodynamic catastrophe if the lesion is a pheochromocytoma [6].

There are no clear guidelines for management of inapparent bilateral adrenal masses. Even the National Institutes of Health Consensus Development Program [7], which addressed the causes, prevalence, and natural history of clinically inapparent adrenal masses, did not specify any guidelines for the management of such cases. It is in this subset of patients with indeterminate features on imaging and FNAB that laparoscopic adrenalectomy (LA) should be employed as a diagnostic and therapeutic tool.
LA is currently considered the procedure of choice for benign adrenal disease [8]. The role of LA for adrenal gland metastasis remains undefined at present. Current data suggest that well-selected patients with adrenal metastasis can undergo LA without any oncologic disadvantage, and it may offer a survival benefit, especially in patients with a significant disease-free interval [9].

Although LA has been contraindicated in cases of proven or suspected primary adrenal cancer because of concern for a high incidence of locoregional recurrence [8], certain authors have performed laparoscopic radical adrenalectomy with acceptable outcomes in carefully selected patients [10]. LA is advantageous in terms of less intraoperative blood loss, decreased analgesic requirement, shorter length of hospital stay, brief convalescence, and superior cosmesis as compared to open adrenalectomy [8].

Laparoscopic adrenal exploration is an effective diagnostic and therapeutic modality for the management of inapparent bilateral adrenal masses.

Conflicts of interest: The authors have nothing to disclose.

References


EU-ACME question

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Question:
The generally accepted recommendation regarding nonfunctional adrenal masses is:

A. Excise lesions >6 cm.
B. Follow up lesions >6 cm.
C. Excise all lesions regardless of size.