

Bilateral adrenocortical carcinoma presenting as acute adrenal insufficiency with co-existing active pulmonary tuberculosis: A diagnostic dilemma

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ABSTRACT

We report a case of a 58-year-old male patient presenting in a state of drowsiness with recurrent hiccups who was found to have hypotension and hyponatremia. He was found to have bilateral adrenal masses with peripheral rim enhancement and central necrotic destruction on contrast enhanced computerized tomography scan of abdomen which accounted for the presentation of acute adrenal insufficiency. Histopathological examination of the adrenal mass was reported as bilateral adrenocortical carcinoma. He also revealed evidences of sputum-positive active pulmonary tuberculosis in absence of any lung mass. Bilateral adrenal mass is an extremely rare condition in its own right and co-existence of such pathological condition with tuberculosis both of which can predispose to acute adrenal crisis, is even rarer. This report emphasizes the need for histopathological diagnosis in every patient of adrenal mass as presumptions can potentially lead to fatal errors.

Key words: Acute adrenal insufficiency, adrenocortical carcinoma, bilateral adrenal mass, pulmonary tuberculosis

INTRODUCTION

Bilateral adrenal enlargement can occur as the result of disseminated malignant disease and this diagnosis should be considered in a patient with severe weight loss.^[1] Tuberculosis can also affect adrenal glands and can precipitate acute adrenal crisis when bilateral involvement destroys most of the functioning adrenal tissue. We report a patient who presented with features of acute adrenal insufficiency with active pulmonary tuberculosis who was later diagnosed to have bilateral adrenal cortical carcinoma. Such a combination is indeed very unusual in medical literature and can lead to diagnostic errors

unless there is a high index of suspicion on the part of the clinician.

CASE REPORT

A 58-year-old non-diabetic non-hypertensive male patient of low socio-economic strata, chronic smoker for last 30 years, presented to us in a drowsy state with recurrent hiccups. A detailed enquiry of his family members revealed that he had history of anorexia, weight loss, low back pain, low-grade intermittent fever with evening rise of temperature for 1 month. He had two episodes of hemoptysis in the recent past. Clinical examination revealed mild pallor, hypotension (blood pressure on admission: 70/40 mm Hg), Initial blood reports revealed microcytic anemia, raised Erythrocyte sedimentation rate and hyponatremia (Serum Na⁺: 122 mEq/L).

Following initial resuscitation and electrolyte replacement, he went on to have an ultrasonography of whole abdomen which revealed bilateral heterogeneous hypoechoic adrenal masses (size Rt. 4 cm × 5 cm, Lt. 3 cm × 4 cm).

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Contrast enhanced computerized tomography (CT) scan of abdomen showed bilateral adrenal masses with peripheral enhancement and central hypodensities [Figure 1].

Serum Cortisol (Morning sample) was <1 mcg/100 ml (Ref. range: 3.7-19.4 mcg/100 ml) and Plasma Adrenocorticotropic hormone (Morning sample) was 84.79 pg/ml (Ref. range: 7.2-63.3 pg/ml). Based on such findings, a diagnosis of primary Addison's disease with bilateral adrenal mass was made. A search for the cause was then started. Keeping in mind the history of hemoptysis in recent past, sputum was sent for microscopic analysis which was found to be positive for acid fast bacilli on two consecutive samples. Plain high-resolution CT scan of thorax showed focal fibrosis and cavitations in the apical region of left upper lobe suggestive of active pulmonary tuberculosis in absence of any neoplastic mass. His human immunodeficiency virus serology was negative. The patient was immediately put on intravenous corticosteroid replacement along with first line antitubercular quadruple therapy (Isoniazid, Rifampicin, Pyrazinamide, Ethambutol).

A CT guided core cut biopsy from the right adrenal mass however showed the presence of anaplastic cells with pleomorphic, hyperchromatic nuclei in the background of large areas of necrosis consistent with adrenocortical carcinoma [Figure 2]. So our final diagnosis was non-functioning bilateral adrenocortical carcinoma along with coexistent active pulmonary tuberculosis presenting as acute adrenal insufficiency. Despite initial good clinical response with fluid, electrolyte and steroid replacement, the patient succumbed to death before his performance status could be improved for any cancer directed therapy.

DISCUSSION

Bilateral adrenal masses are very rare. Neoplastic diseases

presenting with bilateral adrenal masses include nonfunctioning adrenocortical carcinoma, lymphoma, adrenal metastasis from solid tumors e.g., breast, lung, melanoma, colon etc., Non neoplastic diseases reported as presenting with bilateral adrenal mass include tuberculous adrenalitis, histoplasmosis, cryptococcosis, coccidioidomycosis, blastomycosis. Bilateral adrenal masses are, more often than not, clinically inapparent. They are found incidentally when abdomen is scanned for some other reason (so called "incidentaloma"). Only a fraction of bilateral adrenal masses present as primary adrenal insufficiency, even fewer present with Addisonian crisis.

The signs and symptoms of a severe adrenal insufficiency include abdominal or back pain, palpable mass, shock, high fever, hypotension that is resistant to vasopressors, anemia, hyperkalemia, hyponatremia and sudden death occurring a few hours to several days after the insult.^[2] Few publications have described adrenal metastasis from lung cancer leading to an adrenal-deficient state.^[3-5] Most of these reports were secondary to adrenal hemorrhage. Whereas hemorrhagic adrenal tumor is identified on CT scans as a round or oval, large, suprarenal mass with a focal area of hyperdensity, necrosis secondary to tumor often appears hypodense.^[6,7]

The present patient was admitted with Addisonian crisis, later found to have large necrotic bilateral adrenal masses. As he had evidences of active pulmonary tuberculosis, the adrenal involvement was initially thought to be tuberculous in nature. However, histopathological examination clinched the final diagnosis. Our experience with this unfortunate patient highlights that the physician must be aware of varied etiologies that might cause bilateral adrenal enlargement and must not dwell upon a single pathology to explain all the manifestations of this rare entity.

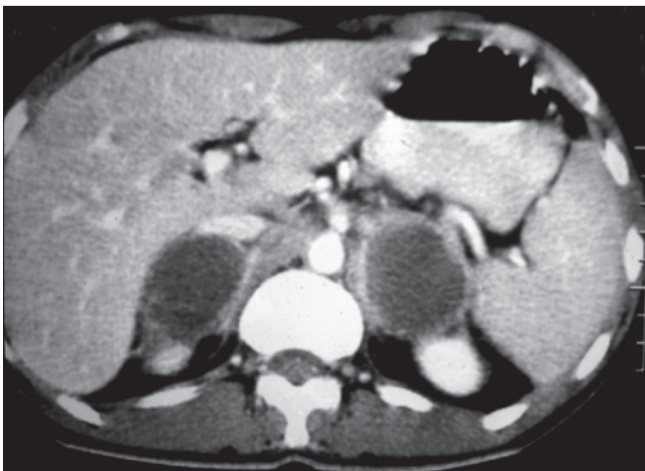


Figure 1: Contrast enhanced computed tomography scan of abdomen showing bilateral adrenal masses with peripheral enhancement and central hypodensities

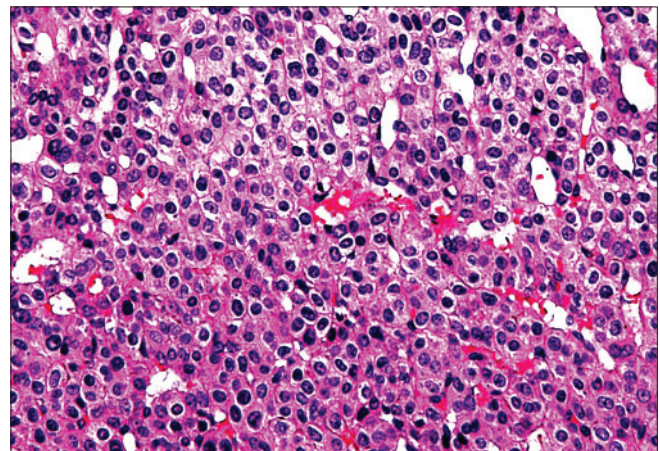


Figure 2: Photomicrograph (H and E, ×400) of histopathological examination of core cut biopsy from the adrenal mass showing the presence of anaplastic cells with pleomorphic, hyperchromatic nuclei with areas of necrosis consistent with adrenocortical carcinoma

CONCLUSION

The present report describes the incidence of bilateral adrenocortical carcinoma in a 58-year-old male presenting with acute addisonian crisis which is rather unusual. The patient also had coexistent pulmonary tuberculosis adding to the diagnostic dilemma. The present report highlights the need for complete evaluation of multiple probable pathological conditions in a patient with bilateral adrenal mass to pinpoint the proper diagnosis.

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