CT FOLLOW-UP OF BILATERAL ADRENAL DISSEMINATED CRYPTOCOCCOSIS IN IMMUNOCOMPETENT PATIENT WITH ADDISON’S DISEASE: CASE REPORT

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ABSTRACT

Disseminated cryptococcosis rarely occurs in immunocompetent hosts. Here, we describe a case of an immunocompetent patient with Addison’s Disease caused by bilateral adrenal disseminated cryptococcosis which presented as adrenal enlargement without enhancement in computed tomography (CT). No changes were found in CT follow-up (10 months) while an improvement of the serum cryptococcal antigen titers was performed, which suggested that non-enhancing masses denoted the stable stage of adrenal fungemia. We conclude that bilateral adrenal cryptococcal masses is a cause of Addison’s Disease in immunocompetent patients and CT may be valuable on assessing the activity of the masses.

Key Words: Cryptococcus spp., Primary adrenal insufficiency, fungemia, immunocompetent
INTRODUCTION

Cryptococcosis is an opportunistic infection for the tendency of occurring in immunocompromised individuals. The most commonly target organs are meninges and lung.[1, 2] There are few reports about cryptococcal adrenal glands in immunocompetent patients. [3-7] Peripheral enhancement of adrenal masses were presented in the most of these cases. Here in, we describe a case of an immunocompetent patient with Addison’s Disease caused by bilateral adrenal cryptococcosis which presented with adrenal masses without enhancement and continued in CT follow-up for 10 months.

CASE REPORT

A 71-year old man presented walking unsteadily and decline of audition for 3 months. He had a history of hepatitis B (HBV) forty years ago and did not use drugs in recent decades. Bilaterally adrenal masses were found by CT scan. A percutaneous adrenal gland puncture biopsy of left adrenal gland was performed for followed histopathology, Cryptococcus spp. were identified within the adrenal tissue. Addison’s Disease was verified by hormone examination in some hospital.

Then he came to central hospital (jingzhou, China) for an antifungal therapy. Eczema of neck and sporadic skin pigmentation were found in physical examination. A neurological examination revealed no focal or lateralizing deficits. His blood pressure was 100/72 mmHg, heart beats, 76 per minute, and temperature, 36.1°C.

The laboratory data at the time of admission showed in table 1. His white cell count was 1015/μL which was a little high than normal at 1000/μL. Although having a history of HBV, his HBV surface antigen (HBsAg) was negative as well as HIV and HCV. The red blood cell count and hemoglobin were lower than normal but not low enough to anemia. The liver function was slightly abnormal. Kidney function tests and levels of serum electrolytes were normal.
A culture of the colorless cerebrospinal fluid (CSF) revealed no growth of Cryptococcus spp. The CSF and serum cryptococcal antigen titers were 1:80 and 1:1280, respectively. Contrast-enhanced abdomen CT demonstrated bilateral adrenal masses (right: 3.7×2.5cm, left:3.0×2.7cm) without enhancement. And liver cirrhosis was not revealed (Fig 1).

Figure 1: CT imaging on admission. (a) Pre-contrast CT of abdomen showed bilateral adrenal masses without calcification. (b,c) Enhanced CT of arterial phase (b) and venous phase (d), no enhancement was observed. (d) CT in multiplane reformation (MPR).

Based on the diagnosis of cryptococcal infection and adrenal insufficiency, the patient was treated with intravenous liposomal amphotericin B combined with oral flucytosine and cortisone.
acetate. Regular CT follow-up were performed in the third, fifth and tenth month. During treatment, he stopped taking cortisone once for 1 month. It was notable that the plasma adrenocorticotropic hormone (ACTH) ascended from 91 pg/ml to 236 pg/ml without any change of CT images in this period. Meanwhile, the CSF and serum cryptococcal antigen titers declined continuously (Fig. 2).

**Figure 1:** (a-c) CT follow-up in the third (a) fifth (b) and tenth month (c) respectively. The size of adrenal masses had no changes. (d) Cryptococcal antigen titers of serum and CSF showed a tendency of decline in 10 months. A rise of ATCH(*) was revealed when the patient stopped taking cortisone.

**DISCUSSION**

Addison’s disease, as known as primary adrenal insufficiency, is a relatively uncommon disease with hypoactive adrenal glands, which may present with muscular weakness, loss of weight, hypotension and pigmentation. Hyponatremia, hyperkalemia and hypoglycemia can show at the time of crisis[8]. The etiologies of Addison’s disease can be autoimmune disease, dysgenesis, hemorrhage, metastases, infiltration, infection, pharmaceuticals and adrenalectomy.[8, 9] Tuberculosis is the most common infected cause. Cryptococcosis, considered as an opportunistic infections, caused in Addison’s disease was rare and mostly reported in immunocompromised patients particularly those with HIV.[2] However, this view has been challenged because of dramatic increase of cryptococcosis in immunocompetent hosts.[10] In other words, cryptococcosis should be considered in immunocompetent patients with septicemia.

In present case, disseminated cryptococcosis was demonstrated by CSF and serum cryptococcal antigen titers. Addison’s Disease was proved by hormone examinations. CT scan and Puncture biopsy of left
adrenal gland indicated that cryptococcosis may be the reason of adrenal insufficiency. This patient had been in healthy and did not have HIV, tuberculosis or malignancies. Although he had a history of HBV, the recovery was verified by the negative HBsAg test. Liver cirrhosis was not revealed in CT scan as well. He was considered to be immunocompetent.

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**Table 2:** Comparison of Prior Cases including CT images

PMH: previous medical history  DM: diabetes mellitus

On comparing the CT findings in our patient with reported immunocompetent patients with adrenal cryptococcal infection, we noted several differences (Table 2). Most cases were described as the masses of bilateral adrenal glands with peripheral enhancement.[4, 6, 7] Hung et al. reported a case of adrenal cryptococcal infection without enhancements.[5] The masses was described being continued in this case, however, no imagines were shown in their report. To our knowledge, this is the first report which have follow-up CT for 3 times in 10 months in immunocompetent patient with bilateral adrenal cryptococcosis combining with Addison’s Disease. Moreover, it was significant that no changes was observed in follow-up CT images when the patient stopped using cortisol and ATCH ascending was presented. This indicates that non-enhancing adrenal masses is a stable stage with irreversible insufficiency of adrenal glands caused by cryptococcosis and antifungal therapy is useless in recovery of adrenal insufficiency. Meanwhile, the loss of volume was appeared after treatment in the cases which showed peripheral enhanced in enlarged adrenal glands.[4, 6] This confirms that peripheral enhancement indicates an active stage of adrenal cryptococcosis. In summary, the pattern of enhancement can reflect the activity of adrenal fungemia.
CONCLUSION

In conclusion, we present a case of immunocompetent patient with Addison's Disease caused by bilateral adrenal disseminated cryptococcosis. Enhanced CT scan can be valuable in activity assessment of adrenal cryptococcal infections.

REFERENCES