Pseudotumoral Adrenal Tuberculosis

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Abstract
tuberculosis is rare nowadays, however, it should be kept in mind in the differential diagnosis of adrenal insufficiency notably in developing countries or in immunocompromised patients. We report here a case of adrenal insufficiency secondary to tuberculosis with bilateral adrenal masses. A 45 years old man was admitted to our department for investigation of a primary adrenal failure. His past medical history was significant for pleural tuberculosis eighteen years ago. He reported several months' history of generalized weakness and unintentional weight loss. On physical examination, he presented diffuse hyperpigmentation. On hormonal assessment, there was low basal cortisol level and a high level of corticotrophin. Computed tomography scan showed bilaterally enlarged adrenal glands. The tuberculin skin test was positive. With the background of tuberculosis, adrenal insufficiency diagnosed by laboratory test and positive tuberculin skin test, bilateral enlargement of adrenal glands was considered most consistent with tuberculosis. Tuberculosis remains a classic cause of adrenal insufficiency with adrenal masses.

Keywords Tuberculosis; Adrenal insufficiency; Bilateral adrenal masses

Background
When Thomas Addison described autopsy findings of six patients with adrenal tuberculosis, in 1855, adrenal tuberculosis was the main cause of adrenal insufficiency. Although it is less frequent nowadays, tuberculosis remains one of the causes of adrenal insufficiency especially in the developing world. Therefore, it should be kept in mind when evaluating a patient with adrenal insufficiency.

1 Observation
A 45 years old man was admitted to our department for investigation of a primary adrenal failure with bilateral adrenal masses. He worked as a secretary. His past medical history was significant for pleural tuberculosis eighteen years ago. His father had also pleural tuberculosis. The patient reported several months' history of generalized weakness and unintentional weight loss.

On physical examination, he presented diffuse hyperpigmentation which predominated on exposed regions. His blood pressure was at 110/60 mm Hg and his pulse at 74/minute. The remaining clinical exam was unremarkable, notably, there were not pulmonary signs.

On hormonal assessment, there was low basal cortisol level at 39.4 nmol/l (N: 154-638) and a high level of corticotrophin (ACTH) at 602 pg/ml (N: 7.2-63.3). The remaining laboratory findings revealed mild hypochromic microcytic anemia.

Computed tomography (CT) scan showed bilaterally enlarged adrenal glands. The left adrenal mass measured 42x20x16.7 mm with calcifications and low enhancement on contrast enhanced CT scan (Figure 1). The right adrenal gland was also enlarged with moderate enhancement on contrast enhanced CT scan (Figure 2).

Tumor markers and endoscopic studies (oesophagogastroduodenoscopy and colonoscopy) were normal. CT scan of the chest revealed pleural nodules and pulmonary infiltrate regarding previous tuberculosis.

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human deficiency virus was negative. The tuberculin skin test was positive (diameter at 15 mm). Acid-fast bacilli test and mycobacterium cultures were negative.

With the background of tuberculosis, adrenal insufficiency diagnosed by laboratory test and positive tuberculin skin test, bilateral enlargement of adrenal glands was considered most consistent with tuberculosis in our patient.

The patient was put on anti-tuberculosis treatment for six months along with hydrocortisone supplementation. He was on four-drug anti-tuberculosis therapy (isoniazid, rifampicin, pyrazinamide and ethambutol) for two months, then on two-drug anti-tuberculosis therapy (isoniazid and rifampicin) for four months.

2 Discussion

Addison’s disease described by Thomas Addison in 1855, when he reported the autopsy findings of six patients with adrenal tuberculosis is a rare condition with an estimated incidence of 0.8 cases per 100,000 and a prevalence of 4 to 11 cases per 100,000. Nevertheless, it is associated with significant morbidity and mortality rates if unrecognized, but once the diagnosis is made; its treatment has spectacular effects (Arlt and Allolio, 2003).

Adrenal tuberculosis was once considered the most common cause of primary adrenal insufficiency. Nowadays, autoimmune adrenalitis is the most common cause of adrenal insufficiency. However, tuberculosis remains a common cause particularly in developing countries and in immunocompromised persons, it represents 20 to 30% of cases of Addison’s diseases in some countries (Laway et al., 2013).

Clinical manifestations may take years to become apparent, and asymptomatic infection is not uncommon. Autopsies studies found 6% cases of adrenal involvement in patients with active tuberculosis (Lamand and Lo, 2001).

Destruction of more than 90% of the adrenal cortex by the infectious process is needed before adrenal failure appears (Kelestimur, 2004). Both adrenal glands are involved, because Mycobacterium tuberculosis spreads to the adrenal glands hematogenously at the moment of primoinfection.

Clinical features of adrenal insufficiency relate to the rate of onset and the severity of adrenal deficiency. In many cases, the disease has an insidious onset and a diagnosis is made only when the patient presents with an acute crisis during an intercurrent illness. Acute adrenal insufficiency is a medical emergency manifesting as hypotension and acute circulatory failure and may sometimes lead to the death of the patient (Arlt and Allolio, 2003).

On the other hand, the patient may present with features of chronic adrenal insufficiency such as weakness, tiredness, weight loss, nausea, intermittent vomiting, abdominal pain, diarrhea or constipation, general malaise, muscle cramps, arthralgia, and postural hypotension (Arlt and Allolio, 2003).
Physical examination may find cutaneous and mucosal pigmentation, weight loss and hypotension.

Routine laboratory findings include hyponatremia, hyperkalemia, azotemia, hypoglycemia and hypercalcemia.

Specific biochemical tests include low basal levels of cortisol and elevated ACTH level. Rapid ACTH stimulation test is also usually used to establish the diagnosis of adrenal insufficiency (Dorin et al., 2003).

Imaging studies done at an early stage, frequently demonstrates enlargement of the adrenals with a pseudo-tumoral aspect on CT scan. At a later stage, the enlarged tuberculous adrenals decreased in size because of fibrosis and calcification.

In Guo’s study, 75% of cases had mass-like adrenal enlargement when the duration of the disease was less than one year, whereas in longstanding disease beyond three years adrenal glands are usually smaller (Guo et al., 2007).

Peripheral rim enhancement with low attenuation in the center of the adrenals is an appealing feature of adrenal tuberculosis. This feature represents the pathologic findings of central caseous necrosis surrounded by fibrous tissue and granulomatous inflammatory tissue. This feature is however, not specific to tuberculosis and it may be seen in adrenal tumors with central necrosis (Yang et al., 2006). The peripheral rim enhancement decreases with time.

Adrenal calcification is a common sign of tuberculosis, but it is not specific, as it can also be seen in adrenal tumors in about 10%. It can be present in all stages of the disease. The overall prevalence of calcification in the study of Vita was 53% at the time of diagnosis and indicated long-standing tuberculosis (Vita et al., 1985). Wang described small calcification on CT scan in 22% patients with active tuberculosis (Wang et al., 1998).

Pathologic studies of the adrenals glands infected by tuberculous mycobacterium revealed caseous necrosis area and tuberculous granuloma at an early stage which causes destruction of the adrenal cortex leading to fibrosis and calcification (Efremidis et al., 1996).

3 Conclusion
Although tuberculosis is less frequent than before, it remains a classic cause of adrenal insufficiency, notably in developing countries. Therefore, it needs to be considered in the differential diagnosis of adrenal insufficiency or bilateral adrenal masses.

Conflicts of interests
The authors declare that no competing interests exist.

References
Arlt W., and Allolio B., 2003, Adrenal insufficiency. Lancet, 361: 1881-1893
https://doi.org/10.1016/S0140-6736(03)13492-7

Dorin R.I., Qualls C.R., and Crapo L.M., 2003, Diagnosis of adrenal insufficiency. Ann Intern Med., 139: 194-204
https://doi.org/10.7326/0003-4819-139-3-200308050-00009
PMid:12899587

Efremidis S.C., Harsoulis F., Douma S., Zafiriadou E., Zamboulis C., and Kouri A., 1996, Adrenal insufficiency with enlarged adrenals, Abdom Imag, 21: 168-171
https://doi.org/10.1007/s002619900037
PMid:8661767

Guo Y.K., Yang Z.G., Li Y., Ma E.S., Deng Y.P., Min P.Q., Yin L.L., and Chen T.W., 2007, Addison’s disease due to adrenal tuberculosis: contrast-enhanced CT features and clinical duration correlation, Eur J Radiol, 62: 126-131
https://doi.org/10.1016/j.ejrad.2006.11.025
PMid:1718208

Kelestimur F., 2004, The endocrinology of adrenal tuberculosis: The effects of tuberculosis on the hypothalamo-pituitary-adrenocortical function, J Endocrinol Invest, 27(4): 380-386
https://doi.org/10.1007/BF03351067
Lamand K.Y., and Lo C.Y., 2001, A critical examination of adrenal tuberculosis and a 28-year autopsy experience of active tuberculosis, Clin Endocrinol, 54(5): 633-639
https://doi.org/10.1046/j.1365-2265.2001.01266.x

Laway B.A., Khan I., Shah B.A., Choh N.A., Bhat M.A., and Shah Z.A., 2013, Pattern of adrenal morphology and function in pulmonary tuberculosis: response to treatment with anti-tubercular therapy, Clin Endocrinol, 79(3): 321-325
https://doi.org/10.1111/cen.12170
PMid:23414172

Vita J.A., Silverberg S.J., Goland R.S., Austin J.H., and Knowlton A.L., 1985, Clinical clues to the cause of Addison’s disease, Am J Med, 78: 461-466
https://doi.org/10.1016/0002-9343(85)90339-0

Wang Y.X., Chen C.R., He G.X., and Tang A.R., 1998, CT findings of adrenal glands in patients with tuberculous Addison’s disease, J Belge Radiol, 81: 226-228
PMid:9889955

Yang Z.G., Guo Y.K., Li Y., Min P.Q., Yu J.Q., and Ma E.S., 2006, Differentiation between tuberculosis and primary tumors in the adrenal gland: evaluation with contrast-enhanced CT, Eur Radiol, 16: 2031-2036
https://doi.org/10.1007/s00330-005-0096-y
PMid:16435135