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A Rare Presentation Of A Common Disease- A Case Report

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ABSTRACT
Addison's disease (AD) or primary adrenal insufficiency was first described by Thomas Addison in patients with adrenal tuberculosis. Over the past several decades, along with the introduction of anti tuberculous treatment , the incidence of both have declined. The most common symptoms are nonspecific, therefore the diagnosis is often delayed and patients may first present with a life threatening crisis. Here we report a case of Addison's disease which on further workup was found to be due to one of the most common infections in India- Tuberculosis.

KEYWORDS- *Addison's disease, Tuberculosis, hyperpigmentation*

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I. CASE REPORT

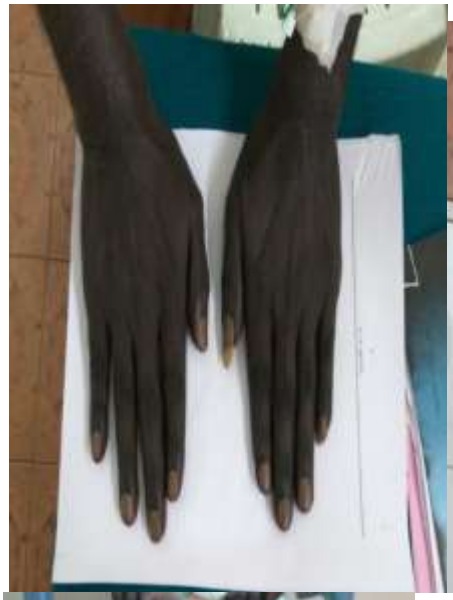
A 65 year old male patient who was a watchman by occupation was admitted with complaints of fever on and off for the past one week, Breathlessness on severe exertion for 2 weeks, generalized darkening of the skin with significant loss of weight and loss of appetite for the past 1 year. There was no history of any drug intake, comorbidities or surgeries in the past.

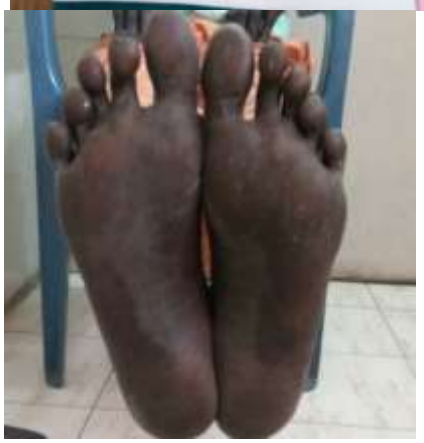
On examination, the patient had diffuse hyperpigmentation involving both hands , foot including the palmar creases. He had hyperpigmentation of buccal mucosa, mucosal surface of lips, alveolar mucosa and hard palate.

Pulse rate was 96/min. regular, normal. Blood pressure was 90/60 mmHg in right upper limb in sitting posture. Standing blood pressure was 70/40 mmHg in right upper limb, which showed that the patient had significant postural hypotension.

Cardiovascular system examination showed normal first and second heart sounds with no murmurs/other events. Respiratory system examination revealed normal bilateral vesicular breath sounds. There was no organomegaly and external genitalia was normal. Central nervous system examination was within normal limits.Fundus examination was normal

Figure 1- Picture showing hyperpigmentation of face. Legs, palms and sole.



INVESTIGATIONS-

Complete hemogram showed a total count of 4800/ microl, hemoglobin of 11.8mg/dl and platelet-3,23,000/mm3. ESR was 65mm/at the end of 1st hour; Random blood sugars were 70 mg/dl. Blood urea was 54→32 mg/dl and serum creatinine was 1 mg/ dl , indicating pre renal failure which had recovered. Serum sodium levels were 117→ 142→ 135 mEq/dl and serum potassium was 8.3→ 4.2 —> 5.1 mEq/ dl indicating hyponatremia and hyperkalemia. Liver function test and urine routine examination were normal. Chest x-ray and ECG , including viral markers(HIV I & II, VDRL, HBsAG, AntiHCV) were negative. Now to find out the causes of diffuse hyperpigmentation, we proceeded with the following investigations

1. PERIPHERAL SMEAR- normal (anemia of chronic diseases, hematological malignancies)
2. PT/APTT/INR- NORMAL
3. USG ABDOMEN- normal, to look for hemochromatosis.
4. SE.FERRITIN- 302.4 nanogram/dl (22-322)
5. VIT B12–445 pgm/ml – normal (200-900) to look for megaloblastic anemia/
6. FOLATE LEVEL- 30.9 nmol/L- normal (4.5-45.3) Folate deficiency
7. Serology for HEPATITIS A & B- Negative
8. OGD SCOPY & COLONOSCOPY- Normal-------to look for Peutz Jegher syndrome
9. USG NECK- NORMAL
10. F.T3- 2.56 pgm/gl(2.3-4.2) , F.T4- 1.06 ng/dl(0.89-1.76), TSH- 4.45 uIU/ml(0.5-6.0)-to look for hyperthyroidism

In view of hyponatremia, hyperkalemia, postural hypotension and diffuse hyper pigmentation., we did

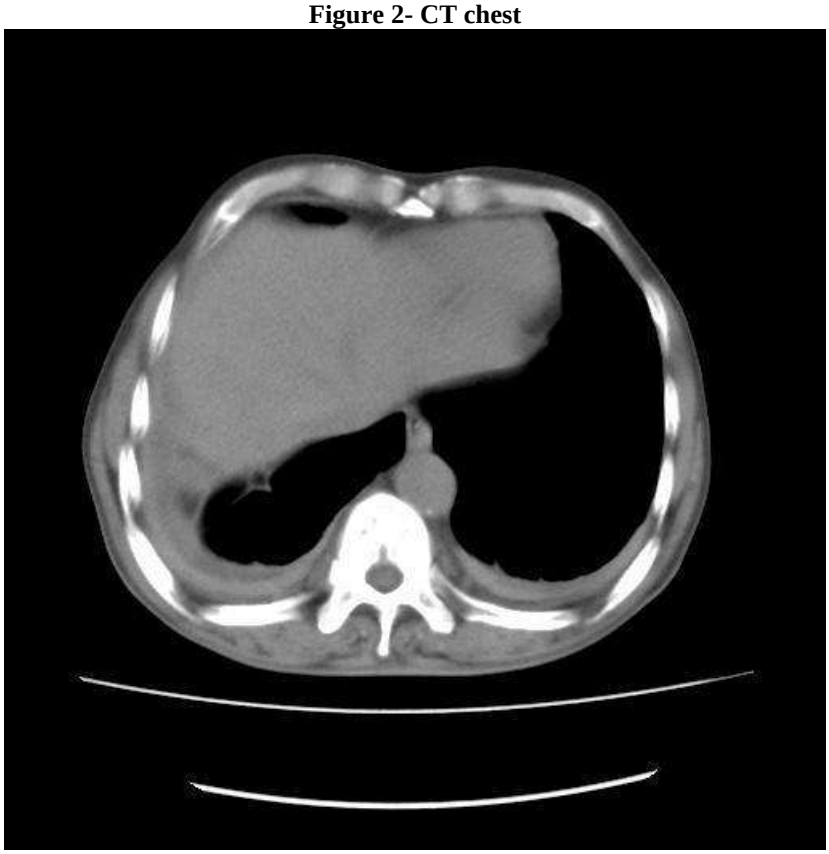
1. SE.ACTH- 927.0 pgm/ml(< 46)---elevated
2. SE. CORTISOL- 2.01 microgram/dl (4.3-22.4)---reduced
3. SHORT COSYNTROPIN TEST WAS POSITIVE(a rise of cortisol levels >20micro gm/dl after 30 min of injection of ACTH)

4. SE.RENIN/ALDOSTERONE RATIO ELEVATED

Thus a primary adrenal insufficiency was confirmed. To find out the cause for it, we proceeded with the following. ANA- 0.2 (<1- NEGATIVE),Adrenal autoantibodies, antiThyroperoxidase antibodies were negative. Non contrast CT brain 2 normal. Serum. Uric acid was 6.0 mg/dl,serum . LDH was 18 IU/l and Serum calcium was 10.1 mg/dl, were done to look for any underlying malignancy.

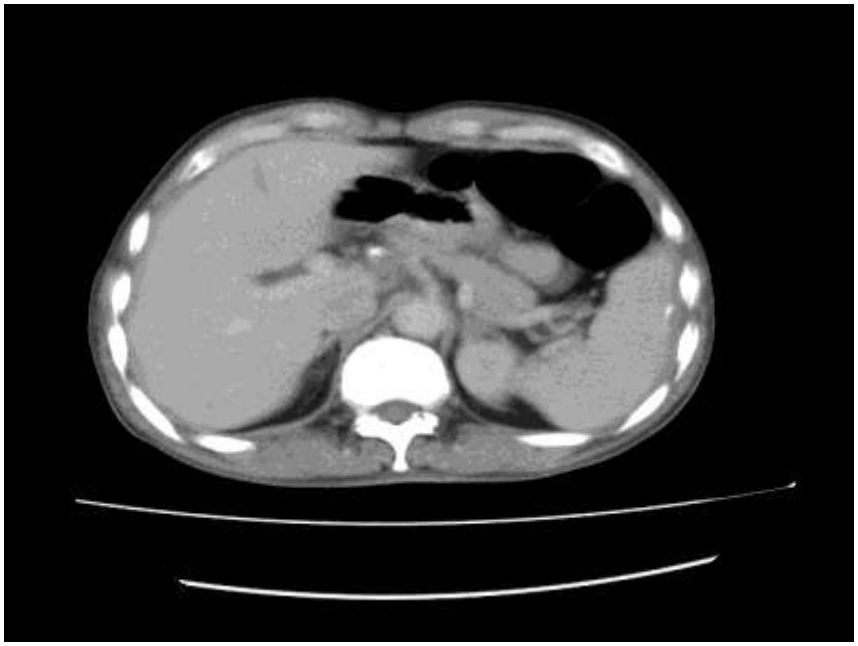
We further proceeded with a CT chest with contrast which showed bilateral pleural effusion with minimal right pleural effusion.

Figure 2- CT chest



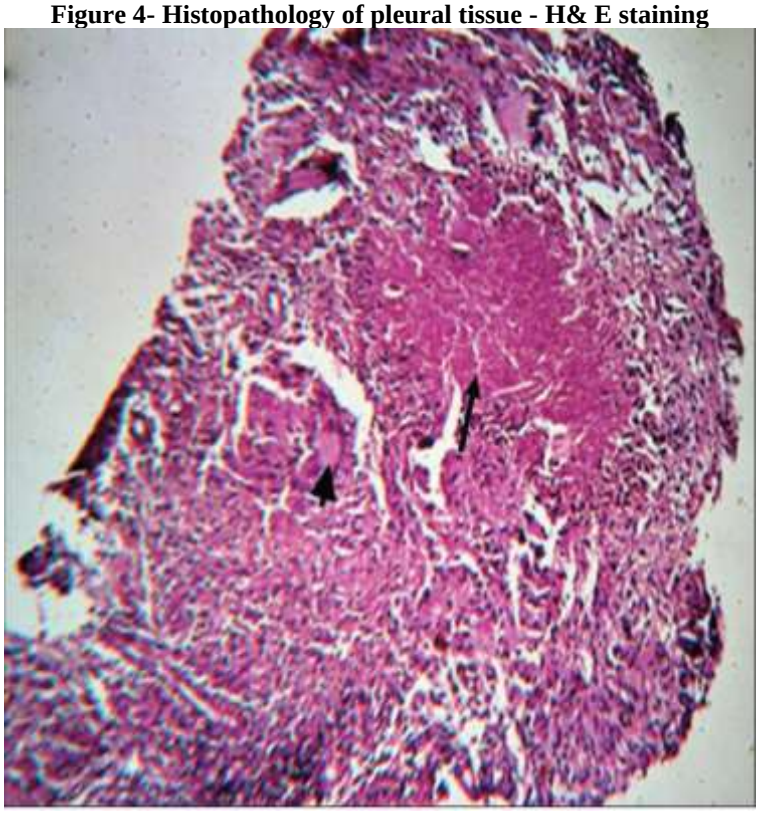
Mantoux test was strongly positive 30mm. We proceeded with Contrast enhanced CT abdomen with adrenal cuts., showed hypertrophy with nodular thickening of (L) adrenal gland, minimal nodular thickening with cystic changes noted in right adrenal gland. There were few enlarged para aortic, retro caval and calcified periportal lymph nodes **suggestive of Tuberculous etiology**

Figure 3- CECT Abdomen with Adrenal Cuts



CT GUIDED BIOPSY OF PLEURAL THICKENING WAS DONE, which showed granulomas with caseous necrosis and Langerhans cells suggestive of TUBERCULOSIS. FNAC from the abdominal lymph node(with Geneexpert and TB culture) was not done.

Figure 4- Histopathology of pleural tissue - H& E staining



THE FINAL DIAGNOSIS

ADRENAL TUBERCULOSIS DUE TO DISSEMINATED TUBERCULOSIS RESULTING IN PRIMARY ADRENAL INSUFFICIENCY-ADDISON'S DISEASE.

TREATMENT GIVEN

1. IV fluids to correct the pre renal failure
2. Inj. Hydrocortisone 100mg iv three times daily followed by oral prednisolone

3. T. fludrocortisone 0.1 mg per day
4. Category 1 Anti Tuberculosis treatment
5. Supportive measures

Patient was symptomatically better, his appetite and breathlessness improved. Patient was started on ATT with steroids, and was kept under follow up. Further biopsy from adrenal gland was deferred in view of his response to treatment and risk of hemorrhage.

II. DISCUSSION

Adrenal sufficiency is a condition in which there is destruction of the adrenal cortex and subsequent reduction in the output of adrenal hormones i.e glucocorticoids – cortisol and mineralocorticoids- aldosterone [1]. There are two types of adrenal insufficiency Primary- adrenal cortex destruction due to autoimmune etiology; Secondary—adrenal destruction due to infection, infiltration, infection, malignancies etc

Today in developed countries, primary adrenal insufficiency is a relatively rare disease [2].Betterle et al,noted a high level of autoimmune form of the disease from European studies., which was most common and ranged from 44.5- 94% of all cases, compared with Addison's disease due to tuberculosis or other causes, which ranged from 0-33.3% [1] .

The symptoms of primary adrenal insufficiency manifest when more than 90% of the adrenal glands have been destroyed [5]. For this reason, Addison's disease due to TB manifests relatively late in life, predominantly in persons aged 40-60 years.

Here is a case which is presented with generalized hyperpigmentation and classical signs of mineralocorticoid and glucocorticoid deficiency with no sex steroid deficiency. On further workup we found it to be primary adrenal insufficiency. He was not immunocompromised, there was no evidence of any other malignancy or infections , history of travel outside India or any history of organ transplantation . By ruling out all other possible causes in this age group, the etiology was narrowed down to tuberculosis. Correlating with the pleural thickening and positive biopsy from the pleural thickening, the diagnosis was narrowed down to Addison's disease due to disseminated TB.

The major CT findings in adrenal TB are bilateral enlargement of the adrenal glands with calcification. There was no peripheral contrast enhancement or central hypodensity suggesting histoplasmosis. When adrenal TB is active, enhanced CT shows enhancement of the peripheral rim of the gland showing a lower degree of attenuation. But, idiopathic Addison's disease is not characterized by either adrenal enlargement or calcification on CT.[6] The enlarged adrenals gradually shrink because of calcification and fibrosis [7].

The best way to confirm the diagnosis is by a biopsy of the gland itself, which was deferred here due to multiple reasons, especially when the size < 4 cm, the risk of internal organ damage and hemorrhage is high [9].

III. CONCLUSION

Adrenal TB is a rare endocrine disorder , in the developed nations it is usually related to autoimmune disorder but in the developing nations it is widely associated with tuberculosis. It is an important disease entity that must be identified early and treated promptly and aggressively.

This case highlights the varied and rare presentation of adrenal tuberculosis which is not much reported in literature and the fact that tuberculosis still continues to be the most common treatable infection for Addison's disease in India.

REFERENCES

[1]. Betterle C, Dal Pra C, Mantero F, Zanchetta R (2002) Autoimmune Adrenal Insufficiency And Autoimmune Polyendocrine Syndromes: Autoantibodies, Autoantigens, And Their Applicability In Diagnosis And Disease Prediction. Endocr Rev 23: 327-364.
[2]. Nomura K, Demura H, Saruta T (1994) Addison's Disease In Japan: Characteristics And Changes Revealed In A Nationwide Survey. Intern Med 33: 602-606.
[3]. Falorni A, Laureti S, De Bellis A, Zanchetta R, Tiberti C, Et Al. (2004) Italian Addison Network Study: Update Of Diagnostic Criteria For The Etiological Classification Of Primary Adrenal Insufficiency. J Clin Endocrinol Metab 89: 1598-1604
[4]. Gordon HW, Robert GD (2008) Disorders Of The Adrenal Cortex: Harrison's Principles Of Internal Medicine (17ed) The Mcgraw-Hill Companies New York.
[5]. Guo YK, Yang ZG, Li Y, Ma ES, Deng YP, Et Al. (2006) Addison's Disease Due To Adrenal Tuberculosis: Contrast-Enhanced CT Features And Clinical Duration Correlation. Eur J Radiol 62: 126-131.
[6]. Sun ZH, Nomura K, Toraya S, Ujihara M, Horiba N, Et Al. (1992) Clinical Significance Of Adrenal Computed Tomography In Addison's Disease. Endocrinol Jpn 39: 563-569.
[7]. Hauser H, Battikha JG, Wettstein P (1981) Pathology Of The Adrenal Glands. Common And Uncommon Findings In Computed Tomography. Eur J Radiol 1: 215-226.
[8]. Long Wang, Yanq J (2008) Tuberculous Addison's Disease Mimics Malignancy In FDG-PET Images. Inter Med 47: 1755-1756.
[9]. Jhala NC, Jhala D, Eloubeidi MA, Chhieng DC, Crowe DR, Et Al. (2004) Endoscopic Ultrasound-Guided Fine-Needle Aspiration Biopsy Of The Adrenal Glands: Analysis Of 24 Patients. Cancer 102: 308-314.