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Case Report Open Access

Sarcoidosis - A Case of "Resistant Tuberculosis"

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Abstract

Context: Sarcoidosis is a multisystem granulomatous disease of unknown aetiology. It is an under diagnosed disease in India. The close resemblance to tuberculosis and the lack of awareness contribute to the under reporting of cases.

Case report: We present a 17 year-old male who was refractory to antituberculous treatment and ultimately turned out to have sarcoidosis.

Conclusion: This case report highlights the importance of considering sarcoidosis as a differential diagnosis, in a country like India, where tuberculosis is endemic. The clinical course of this case was different from that reported in Western Literature.

Key words: Sarcoidosis; Tuberculosis

Introduction

Sarcoidosis is a multisystem granulomatous disease of unknown aetiology. It usually has a benign course, but those cases with multi system involvement have poorer prognosis. Sarcoidosis is an under diagnosed disease in India, probably due to the close resemblance to tuberculosis and the lack of awareness. But this disease is not so rare in India, as previously thought.

Case presentation

A 17-year-oldmale was admitted to our hospital because he had been suffering from loss of appetite and weight loss due to a fever of unknown origin for 1 year. He was empirically diagnosed with tuberculosis in a local hospital, and was started on antituberculous drugs even though his sputum Acid Fast Bacilli (AFB) and Mantoux were negative. Despite the antituberculous treatment for 6 months, he gradually developed lower leg weakness. He eventually had difficulty walking and getting up from a squatting position. He had a history of being treated with antituberculous drugs twice at 6 years old for pulmonary tuberculosis, followed by another course for abdominal tuberculosis. Upon admission, his height was 153 cm and weight 36 kg. He had hypotension, which improved after a hydrocortisone injection. His facial, axillary and pubic hair had decreased, with infantile external genitalia. An abdominal examination revealed hepatosplenomegaly. A neurological examination revealed normal power with bilateral lower limb spasticity, hyperrefelexia and bilateral up going plantars. Ophthalmology evaluation did not show any granulomatous foci in the eyes. There were no skin lesions.

Investigations showed Hb 10.8 g/dL, total leucocyte count 5400/mm^3 with differential count P59L46E4 ESR 86mm /1st hour Platelet count 2.1Lakhs/mm^3 PCV 27%. Urine routine examination was normal. Random blood sugar 62 mg/dL. S. Bilirubin Total/direct 0.8mg%/0.3 mg% AST/ALP /Alkaline phosphatase – 46/25/341 Total. Protein/ Albumin – 7g%/3.3 g% Prothrombin time normal Blood Urea 26 mg/dL S.Creatinine 0.6 mg/dL S. Na 138 mEq/L S.K 3.5 mEq/L HIV/HBV/HCV – Negative Peripheral Smear -Normocytic normochromic anemia, no malarial parasites seen. Bone Marrow –cellular marrow with normal marrow elements, reactive eosinophilia and plasmacytosis. Blood and urine cultures were Sterile CSF study was normal with normal protein and sugar. Rheumatoid factor 25.81 (<20) ASO titre 89.39 (<200) ANA was negative. Sputum AFB, and Mantoux were negative CRP was positive Widal test and Brucella IgM were negative. S. Calcium level was 9.3 mg/dL (8.1-10.4 mg/dl) S. ACE level 92 U/L (8–65) by spectrophotometry. Repeat value 176U/L. 25(OH) Vit D level was 9.27ng/ml S. ACTH level 6.18pg/ml (7.2–63.3) S. cortisol value was 22.65 (5-23) serum FSH value 0.61mIU/ml (0.9-15), serum LH 0.02 mIU/ml (1.7–8.6), serum Testosterone 0.32ng/ml (1.95-11.38) thyroid function tests were within normal limits. Serum ferritin (319 ng/ml) and serum ceruloplasmin (42 mg%) were within normal limits.

X-Ray chest revealed evidence of mediastinal lymphadenopathy. CT chest with contrast showed right upper paratracheal perivascular lymph nodes. Ultrasound abdomen showed hepatomegaly with hyperechoic parenchyma, enlarged spleen, portal vein. MRI Brain showed evidence of leptomeningeal enhancement along the tentorium cerebelli on both sides, with normal pituitary and hypothalamus. MRI of spine did not reveal any abnormality. Liver biopsy report was chronic active hepatitis. Upper GI endo

scopy showed granular velvety appearance of D1, D2 biopsy of which showed mild chronic inflammation, with no evidence of granuloma. Echocardiogram did not show any evidence of infective endocardiitis. Pulmonary function tests showed restrictive pattern. Taken together, he was diagnosed with sarcoidosis, restrictive lung disease, hypopituitarism, hepatosplenomegaly, and non-compressive myelopathy.

He was started on steroids and hydroxychloroquine initially. Later hydroxychloroquine was stopped and myophenoclate mofetil was added. After about two weeks he developed seizures and was started on antiepileptic drugs. But he later developed status epilepticus, went into coma and died.

Discussion

Prevalence rate of sarcoidosis of 10-40 per 100,000 have been reported from North America, Europe and Japan. The true incidence of sarcoidosis in India is not known. In Kolkata 10-12 cases/ 1000 new registrations annually (S K Gupta et al) and 61.2 / 100,000 new cases in Delhi (VP Chest Institute), have been reported. The close resemblance to tuberculosis and the lack of awareness contribute to the under reporting of cases. This boy was also treated empirically for tuberculosis several times in the past.

Sarcoidosis affects mostly people aged 20 to 40 years but occasionally affects children and older adults. The mean age of presentation in Indian studies was above 40 years, whereas in western population it affects people below the age of 40 years [1]. Sarcoidosis can affect children of any age and the prognosis is usually better than adults. In the western hemisphere, sarcoidosis is more common in females, while in India it is more common in males [2]. Moreover studies have shown that sarcoidosis occurs in all ethnic groups and communities in India. Sarcoidosis can present as acute, subacute or chronic forms. Fever and constitutional symptoms are more in Indian studies. This case was also investigated as a case of pyrexia of unknown origin. Intraabdominal lymphadenopathy has been reported from Indian studies [2]. It was probably due to the presence of intraabdominal lymph nodes, he was given antituberculous drugs in childhood. The prevalence of hypercalcemia is low in sarcoidosis in India when compared to western studies, possibly due to vitamin D deficiency which is common here. This boy also did not have hypercalcemia and his 25(OH) Vitamin D level was very low [1]. An elevated serum ACE level can be seen in other conditions like granulomatous diseases, diabetes, lymphoma ,but a value more than two times the upper limit of normal is seen only in sarcoidosis [3]. The diagnosis of sarcoidosis can be confirmed only by biopsy which should show the presence of non caseating epitheloid granuloma. But the presence of necrosis in granulomas has been reported [4]. In this case biopsy could not be done. Thoracoscopy and thoracic lymph node biopsy was planned.

Conclusion

This case report highlights the importance of considering sarcoidosis as a differential diagnosis, in a country like India, where tuberculosis is endemic. The clinical course of this case was different from that reported in Western Literature.

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