Case Report Neurobiology

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AN UNUSUAL PRESENTATION OF NEUROBRUCELLOSIS WITH GUILLAIN-BARRÉ SYNDROME

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ABSTRACT

Human brucellosis is an infectious, zoonotic disease that has a varied clinical spectrum of manifestations. Brucellosis is caused by the bacteria belonging to the genus Brucella. Brucellae are gram negative, aerobic coccobacilli which can affect any organ like central nervous system, joints, heart, kidney, skin, pulmonary system and genitourinary system leading to varied clinical symptoms. Neurobrucellosis is relatively rare and serious disease which includes meningoencephalitis, encephalitis, myelitis, subarachnoid hemorrhage, cranial and peripheral neuropathy, and psychiatric manifestations. We report an unusual case of neurobrucellosis presenting with manifestations of Guillain-Barré Syndrome with fever and bilateral lower motor neuron facial palsy. The patient was treated with IV Immunoglobulins and antimicrobial therapy such as doxycycline and rifampin. The patient showed marked clinical improvement. Thus timely diagnosis and effective treatment leads to good resolution of the symptoms and prognosis.

KEYWORDS : ***Neurobrucellosis, zoonotic, Guillain-Barré Syndrome***

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INTRODUCTION

Brucellosis is the commonest bacterial zoonosis and causes more than 500 000 human infections per year worldwide.[1] Brucellosis can have multisystem involvement of which neurobrucellosis is an unusual and serious complication which can even be fatal if left untreated. In adult patients, the incidence of neurological complications can range between 0-25%.immunological mechanisms are responsible for the demyelinating lesions in the cerebral white matter and spinal cord. Guillain-Barré Syndrome, occurring as a result of an acute infection such as brucellosis is quite rare.

CASE REPORT

A 44 year-old man, presented with complaints of 2-week history of fever with chills and rigors, headache and fatigue. No other history was remarkable except that he worked in a slaughterhouse. Physical examination revealed pallor,BP-90/60 mmHg and hepatosplenomegaly. Blood Cultures revealed Gram-negative non- spore forming, aerobic coccobacillus - Brucella species. *Brucella* antibody test also revealed high titers and so a diagnosis of brucellosis was made .Patient was started on Doxycycline and intramuscular Streptomycin . One week later, patient complained of vision disturbances, numbness and muscle weakness of both upper and lower limbs. He denied history of upper respiratory tract infection or diarrhoea in the recent past. On neurological examination, his Glasgow Coma Scale score was 15 out of 15. His higher mental functions and speech were normal. There was left-sided ptosis along with ophthalmoplegia with bilateral lower motor neuron type of facial palsy. Hypotonia was present in all four limbs and deep tendon reflexes were absent. Plantar reflex was flexor on both sides. Muscle power was markedly reduced ,1/5 distally and 3/5 proximally. Sensory functions were normal. Investigations done revealed a normal chest x-ray,total count, hemoglobin, platelets, and Erythrocyte sedimentation rate. liver function and hepatic function tests were normal. CT brain was normal, but magnetic resonance imaging (MRI) scan of the brain revealed leptomeningeal enhancement with associated minimal edema in the right parietal lobe. Cerebrospinal fluid analysis revealed white blood cell counts of 4 cells/cumm,glucose 4.2 mmol/L with protein of 2.47 gm/L. Cultures of blood and CSF revealed Brucella growth. *Brucella* agglutinin serology was 1: 480. The CSF was positive for Brucella serology. Nerve conduction studies and electromyogram had typical findings of demyelinating type of sensorimotor polyneuropathy. A diagnosis of GBS was made secondary to acute brucellosis infection. The patient was treated in the intensive care unit, where IV Immunoglobulins and antimicrobial therapy were administered. He gradually improved and after 4 weeks, he was able to walk with the help of support. There was almost complete resolution of his facial and ocular weakness. Repeat *Brucella* titer showed a drop to 1:160. The patient was discharged with oral Doxycycline and Rifampicin with advice on regular follow up to treatment of a total of 3 months.

DISCUSSION

This patient's presentation was entirely compatible with a diagnosis of GBS secondary to acute brucellosis. The ascending type of motor paralysis with absent deep tendon reflexes, CSF albumin-cytologic dissociation along with electrophysiological findings are classical features of GBS. The diagnosis of brucellosis is confirmed by the markedly elevated *Brucella* titers .Cranial nerve palsies have been reported in 19% of patients with GBS.[2-3]Both axonal and demyelinating forms of GBS have been documented with brucellosis.[4] The earliest description of *Brucella*-related GBS in Saudi Arabia, was recorded in 1996 in a 9-year-old girl who suffered from protracted paroxysms of severe hypertension before she developed the classical signs of GBS.[5] Molecular mimicry has been stated as the immunological basis for the development of GBS. Cross-reactive immunological responses as a result of this molecular mimicry between GM1 gangliosidesand *Brucella* lipopolysaccharides which leads to the cross-reactive response may account for the acute motor axonal polyneuropathy. In neurobrucellosis imaging findings may be found in four types: normal, meningeal contrast enhancement, white matter changes, and vascular changes.[6] According to Kochlaret al.[7], the criteria necessary for definite diagnosis of neurobrucellosis are

i. neurological dysfunction not explained by other neurologic diseases,
ii. abnormal CSF indicating lymphocytic pleocytosis and increased protein,
iii. positive CSF culture for Brucella organisms or positive BrucellaIgG agglutination titer in the blood and CSF, and
iv. response to specific chemotherapy with a significant drop in the CSF lymphocyte count and protein concentration.

Dual- or triple-combination therapy with Doxycycline, Rifampicin, Trimethoprim-sulfamethoxazole, Streptomycin, or Ceftriaxone for 3–6 months has been recommended [8]. This case report highlights the importance of considering an infectious cause such as acute brucellosis in patients with acute descending type of motor paralysis consistent with diagnosis of GBS.

CONCLUSION

This case is being presented for the unusual presentation of Guillain-Barré Syndrome in brucellosis and to signify the importance of its complications.

CONFLICT OF INTEREST

Conflict if interest declared none.

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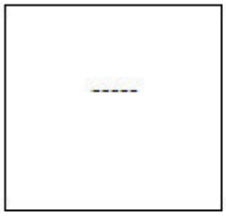

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