CASE REPORT

Brucellar Chorea – A Rare Manifestation of Brucellosis

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Abstract:

Brucellar chorea is a rare and unusual presentation of brucellosis. We would hereby like to report a case of Brucellar chorea. The purpose of reporting this case is to create awareness about the neuropsychiatric manifestations of brucellosis. Neurobrucellosis should be considered as one of the differential diagnoses in patients having long-standing fever with neurological manifestations, especially in endemic zones like India.

Keywords: 2-ME, Brucellosis, Chorea, SAT, Zoonosis

Introduction

Brucellosis is a zoonosis with worldwide existence. It is a systemic infection that can involve multiple organ systems and can manifest in acute, sub-acute or chronic forms. Various complications may develop during any stage of the disease involving musculoskeletal, cardiovascular, genitourinary, reticulo-endothelial and central nervous systems. Most patients of brucellosis experience non-specific Central Nervous System (CNS) symptoms like headache, irritability, insomnia, anxiety and depression. Neurobrucellosis, due to invasion of the (CNS), may manifest as meningitis and meningoencephalitis in 3 to 5 % of cases, at times complicated by cerebral abscess, cranial nerve deficits and ruptured mycotic aneurysms [1-3].We hereby report a case of brucellar chorea, which is a very rare complication of brucellosis.

Case Report:

An 11-year-old female child from rural area in Karnataka was admitted with complaints of low grade fever since six weeks, headache since three weeks, and involuntary movements of limbs and face, and altered sensorium since eight days. There was no history of sore throat, joint pains or swelling, rash, breathlessness, cough, weight loss or known exposure to drugs and toxins. There was no history of tuberculosis or with contact of tuberculosis. No other family members had similar complaints. History of direct contact with animals and raw milk ingestion was positive. She had received a course of oral antibiotics for suspected salmonella infection.

General physical examination of the patient revealed a moderately built and nourished girl. Her vital signs were normal. Bilateral cervical lymphadenopathy was present.

She was conscious, oriented but restless and emotionally unstable. Her speech was slurred. Cranial nerve functions were normal. Superficial and deep reflexes were normal. Involuntary, irregular, jerky, purposeless movements of limbs, twitching of face and lip smacking were present. Rest of the nervous system examination was unremarkable.

Cardiovascular, respiratory and per abdominal examinations were normal. With the above clinical findings, she was provisionally diagnosed

as a case of chorea with rheumatic / infectious/ metabolic / toxic as the most likely etiological differential diagnoses.

Routine blood investigations were unremarkable except Erythrocyte Sedimentation Rate (ESR), which was found to be 30 mm at the end of first hour. CT scan of the brain showed no abnormalities. Thyroid function tests, serum electrolytes including calcium and magnesium levels were within normal limits.

CSF examination revealed 200 cells / mm^3 with 95 % lymphocytes, protein of 200 mg / dl and sugar 40 mg / dl. Gram's, ZiehlNeelsen's staining and India ink preparations of the CSF were negative. Mantoux, Widal and HIV screening tests were negative. She also tested negative for C reactive protein, anti-steptolysin O and anti-DNase B, antinuclear and anti-dsDNA antibodies. MRI of the brain could not be performed due to unavailability.

As a part of an ongoing research project on Pyrexia of Unknown Origin (PUO), the serum was screened for Brucella agglutinins with Rose Bengal Plate Test (RBPT) which came positive. Serum Agglutination Test (SAT) and 2-Mercaptoethanol tests (2-ME) showed titers of 1:640 and 1:320 respectively. As serum showed significant agglutinins, CSF sample was tested for Brucella agglutinins by SAT which showed titers of 1:64. Blood culture grew *Brucella melitensis* biotype1. CSF culture for Brucella spp. was sterile. Based on CSF picture, presence of Brucella agglutinins both in the serum and CSF and positive blood culture, a diagnosis of neurobrucellosis was made. The patient was started on doxycycline 100 mg, rifampicin 450 mg orally in two divided doses. Additionally injection streptomycin 500 mg intramuscularly was given daily for initial 14 days. Doxycline and rifampicin were continued for a total period of 6 months. Follow-up blood titers for Brucella agglutinins showed significant decrease.

Discussion

Brucellosis is a neglected disease in India. Due to lack of awareness about the disease among physicians and non-availability of laboratory testing facilities, we have a partial knowledge about its prevalence. Most of the patients with systemic brucellosis present with pyrexia of unknown origin. About 3-5 % have predominant central nervous system involvement. The common neurological manifestations include meningitis, meningoencephalitis, diffuse encephalopathy, posterior fossa (ataxic or brainstem) syndromes, meningomyelitis, inflammatory peripheral neuritis / radiculitis, inflammatory demyelinating syndromes, papilledema or papillitis without other focal features. Additionally neuropsychiatric manifestations like depression, psychosis, agitation, personality disorders and euphoria can also occur [3]. In rare instances, abnormalities referable to the basal ganglia and associated systems develop, including parkinsonism, chorea, athetosis, narcolepsy, or cataplexy. These clinical manifestations are much more confusing and may suggest a wide variety of alternative diagnoses, including other infectious, degenerative, endocrine, metabolic, autoimmune, genetic and paraneoplastic conditions. At times when no evident physical abnormality is found, patient is considered to have a functional disorder or even suspected of malingering.

Because of the protean clinical manifestations of brucellosis, strong clinical suspicion, detailed history with emphasis on occupational, environmental and socio-cultural conditions is essential to aid the diagnosis. Definitive diagnosis of neurobrucellosis can be made based on following criteria: abnormal CSF picture showing protein rich lymphocytic pleocytosis, positive Brucella agglutinins in blood and CSF, positive blood or CSF cultures for Brucella organisms and response to specific chemotherapy [2]. Additionally molecular diagnostic methods like PCR can be useful.

Chorea is a very rare and unusual presentation of neurobrucellosis. It is mostly overlooked and rarely reported because of diagnostic complexity. So far, few cases of brucellar chorea have been reported in the literature [4-6].

India is predominantly an agrarian economy with live stock rearing as a common income supporting activity, owing to which people are exposed to animals directly or indirectly. Also raw milk ingestion is a common practice. Hence, the possibility of brucellosis in patients with unexplained neurological and psychiatric symptoms with rural background should not be overlooked and due importance should be given for epidemiological history.

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