Iranian Journal of Clinical Infectious Diseases
2007;2(4):203-205

CASE REPORT

Neurobrucellosis; A case report

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ABSTRACT

Background: Brucellosis with different pictures is common in our country and should be noticed in high risk patients.
Patient: A 23 years old male farmer, presented with headache, vomiting, and tremor. Disease had begun 6 months ago with right side orchitis, fever, chills, then, gradually illness, weakness, tremor, anorexia, vomiting, nonproductive cough, retrosternal pain and dysphagia, blurred vision, generalized muscular rigidity and disability in daily activities were added. In physical exam illness, weakness, fever, wet skin, tremor, generalized muscular rigidity, papillary edema were detected. He had positive Wright test (1/1250) and lymphocytic pleocytosis in CSF. Three drug regimen and steroid (1 month) were administered and he responded well to this initial therapy, however, during his 3rd month follow up, he developed severe bilateral hearing loss unresponsive to steroids and 6 months later he had a self-limiting 20 hours left sided hemiparesis.
Conclusion: Neurobrucellosis may present with parkinsonism (tremor and generalized muscular rigidity) and meningovascular involvement. This may even progress under 3 antibiotic regimen treatment.

Keywords: *Neurobrucellosis, Parkinsonism*.
(Iranian Journal of Clinical Infectious Diseases 2007;2(4):203-205).

INTRODUCTION

Brucellosis is a common zoonotic and an important occupational disease in our country (1). It presents as acute, subacute and chronic courses by involving different systems with varied manifestations. CNS involvement is important clinically and occurs in 2-12% of cases (2-4).

Brucellosis and neurobrucellosis are more common in second to forth decades (5). Involvement of entire central and peripheral nervous system and also psychologic disturbances can occur.

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Every patient with neurologic manifestations in endemic areas should be ruled out for brucellosis. For susceptible cases, travel history, occupation, and similar symptoms in other family members should be sought. Isolation of organism from blood, cerebrospinal fluid (CSF), and bone marrow; and serum antibody detection could establish the diagnosis (2,5).

PATIENT

A 23 years old farmer was admitted because of illness and weakness. Disease had begun 5 months ago by weakness, fever, chills, orchitis, and tremor for which he received 2 weeks treatment. One month later, orchitis resolved but headache, blurred

vision, epigastric pain and vomiting were added. His past medical history was unremarkable. His brother had brucellosis one year ago. His sheep experienced abortion, hematuria and death.

He was admitted by headache, bilateral blurred vision, nonproductive cough, anorexia, vomiting, retrosternal pain, odinophagia, abdominal pain, constipation and tremor in extremities. In physical examination he was conscious, very ill, afebrile with generalized muscle rigidity, cogwheel rigidity and tremor in upper extremities, epigastric tenderness, bilateral papillary edema with direct and indirect light reflex, normal visual field, visual acuity of 9/10, as well as no meningeal and cerebellar signs and no Kayser-Fleisher ring. He had normal cardiovascular exam and normal scrotum.

Paraclinical findings included normal complete blood count (CBC), urine analysis (U/A), chest-x ray, echocardiography, electroencephalography (EEG), brain CT, abdominopelvic sonography, and erythrocyte sedimentation rate (ESR)=15. Abnormal findings were: standard tube agglutination (STA)=1/1280, echographic findings of right testis postinflammatory lesions and secondary atrophic avascularity, and erosive gastritis in gastroscopy with negative rapid urease test (RUT) and helicobacter antibody.

Treatment was commenced with ceftriaxone, rifampin (due to gastric intolerance to doxycycline & sulfamid), antiacid and omeperazole. Repeated STA was negative, and 2ME was 1/640 by high dilution.

Lumbar puncture (LP) showed lymphocytic pleocytosis, negative veneral disease research laboratory (VDRL), STA=1/40, and negative smear, culture and PCR for tuberculosis.

Having improved gastritis, doxycycline and prednisolone were added to the regimen, then, at the end of the first month, prednisolone and ceftriaxone were discontinued and treatment was continued with 3 drugs (doxycycline, rifampin and cotrimoxazole). Two months later, he was discharged with good health, mild blurred vision, papillary edema and mild rigidity and tremor which all resolved after 4 months except for tremor, however, bilateral sensory-neural hearing loss appeared that was unresponsive to steroid.

Nine months after the initial symptoms, he had a TIA (transient ischemic attack) presented with left-sided self-limiting hemiparesis for 20 hours. Three drugs were used for 1 year with no side effect. All signs and symptoms were resolved except for bilateral hearing loss.

DISCUSSION

Brucellosis could present with multiple clinical manifestations in different systems and may occur in different courses (2). This may lead to delayed diagnosis, for example, peripheral neuropathy and motor weakness of a patient with brucellosis was diagnosed after 18 months (6). Our patient was visited many times with physician with no attention to brucellosis in his brother and in his sheep (death, abortion), therefore, diagnosis and treatment was delayed for 4 months. Although false negative STA is misleading (2), as occurred in our case. It is demonstrated that antiplasmic protein antibody against Brucella spp by ELISA, Western Blot of CSF in brain involvement (7), and imaging may not have correlation with clinical manifestations (8). Our patient has normal brain CT and positive STA (1/40) in CSF analysis.

Meanwhile, parkinsonism that was presented with cogwheel muscle rigidity and tremor at rest can be found in infarcts, postencephalitis, and peripheral neuropathy as in brucellosis (9), however, sporadic cases with brucellosis were also reported since 1986 (10). Rigidity and tremor in our patient resolved without intervention. Isolated rigidity has been reported in basal ganglia diseases (11) and can be the first manifestation of neurobrucellosis (12).

On the other hand, disease progression under suitable treatment was a matter of concern since he

developed 8th cranial nerve paresis with bilateral sensory-hearing loss during the 4th month and TIA during the 9th month of therapy. Koussa reported self-limiting TIA in 3 out of 15 neurobrucellosis patients (13). Early treatment results in complete papillary edema resolution (5). This patient had second nerve involvement as papillary edema and blurred vision for 3 months, and meningeal signs (headache, vomiting) resolved at the end of the first month.

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