

# Neurobrucellosis with Hearing Loss and Cerebellar Involvement / A Case Report

## İŞİTME KAYBI VE SEREBELLER TUTULUMLU NÖROBRUSELLOZ / OLGU SUNUMU

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### Summary

Brucellosis is a disease of ubiquitous distribution. Direct CNS infection is quite rare. Cranial nerves commonly involved are optic, oculomotor, abducent, facial and vestibulocochlear nerves. Among these there seems to be a predilection for the vestibulocochlear nerve leading to sensorineural hearing loss. We report a patient presenting with acute onset of ataxia and sensorineural hearing loss shown on audiometry.

**Key Words:** Brucellosis, Hearing loss, Neural involvement

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### Özet

Bruselloz yaygın tutulumu olan bir hastalıktır. Doğrudan santral sinir sistemi tutulumu nadirdir. Optik, okulomotor, abducent, fasiyal ve vestibulokoklear sinirler sık tutulan kraniyal sinirlerdir. Bunların içinde sensorinöral işitme kaybına yol açacak şekilde vestibulokoklear sinirin tutulma eğilimi vardır. Burada akut ataksi ve odyometri ile tespit edilen işitme kaybı ile başvuran bir hasta sunulmaktadır.

**Anahtar Kelimeler:** Bruselloz, İşitme kaybı, Nöral tutulum

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Brucellosis is a disease caused by gram negative coccobacilli which infect humans either by direct contact with infected animals or ingestion of raw meat or unpasteurized milk from such animals. It is caused by one of the three strains: *B. mellitensis*, *B. abortus*, *B. suis* (1).

Neurobrucellosis is a neurological complication which is directly attributable to the direct effect of *Brucella* organism or its endotoxins on the meninges and brain (2). The reported incidence of neurobrucellosis varies between %0.5 to %25 of all brucellosis cases (3).

We report a patient with brucellosis having new onset of hearing loss and cerebellar findings.

### Case Report

38 year-old female presented to the emergency room with a history of fever, headache, palpitation, night sweats for one and a half month and dizziness, loss of balance to her either side, tinnitus and progressive hearing loss for 4 days. She did not complain of nausea and vomiting. Her headache began slowly from the back of the neck, increased in intensity and was responsive to drugs. During one and a

half month period, she lost 8 kg of weight. There was also sporadic spells of fever reaching 39°C. There was no history of sonic trauma or exposure to ototoxic drugs. There was a history of eating raw cheese.

On admission, the blood pressure was 95/65 mmHg, pulse rate was 108/minute and temperature was 38.5°C. Although the patient was drowsy, there was no impairment in her mental capacity. On physical examination, liver and spleen were palpable. Ear, nose and throat examination were normal. On neurological examination, funduscopy was normal. Visual acuity was normal. Meningeal signs were absent. There was no loss of muscle strength in extremities. Deep tendon reflexes were increased and flexor plantar responses were present. She was dysmetric and dysdiachokinesic in upper extremities. She had an ataxic and broad based gait. The sensory system was normal.

There was iron deficiency anemia, hyponatremia and mild elevation of liver enzymes. *Brucella* spot test was negative but Wright agglutination test (indirect Coomb's test) was positive at 1/320 titer. Lumbar puncture revealed clear CSF with normal pressure. CSF protein was 15 mg/dl and glucose was 70 mg/dl. Gram stain was negative and CSF cultures were negative for ARB, fungi, bacteria including *Brucella*. Anti-nuclear antibody was negative. Cranial MRI on 1.5 Tesla both with and without contrast didn't demonstrate any abnormality. Pure tone audiogram worsened with higher tones.

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On the basis of the history and positive serology, a diagnosis of neurobrucellosis was made and the patient was treated for six weeks with doxycycline 2x100 mg/day and rifampicin 600 mg/day. During follow up, her drowsiness, hearing loss and ataxia regressed significantly. The audiogram was normal after two months of treatment.

### Discussion

Brucellosis is a disease of ubiquitous distribution. Incidence of neurobrucellosis, receiving few attention in reviews about brucellosis, is in decline (4). Reported clinical pictures include acute, subacute or chronic meningitis, diffuse and localized encephalitis, myelitis, radiculoneuritis, compressive myelopathy and mononeuritis (5,6). Optic, oculomotor (7), abducent (8,9), facial (2), and vestibulocochlear (2,3,9) nerves are commonly involved. Among these, there seems to be a predilection for the vestibulocochlear nerve (9,10) leading sensorineural hearing loss (2,8), this is relatively late manifestation in the natural history of the disease (2). Significant hearing impairment results in 12% of brucellosis. Meningitis is the most common neurologic manifestation of brucellosis. Meningovascular brucellosis caused by panarteritis has a predilection to the skull base like tuberculosis or syphilis (2). This type of brucellosis may mimic transient ischemic attack. On rare occasions, intracranial hypertension caused by arachnoiditis in the posterior fossa leading to hydrocephalus may be the presenting feature (11).

Neurobrucellosis has neither a typical clinical picture nor specific CSF or peripheral blood findings (11). Meningeal brucellosis is considered when at least one of the following criteria is present: isolation of *Brucella* from CSF; presence in CSF of anti-*Brucella* antibodies at any titre obtained with microagglutination; Coombs test can show IgG nonagglutinating immuno-globulins or Rose Bengal tests which readily detect IgM immunoglobulins; and abnormalities in CSF in the presence of confirmed brucellosis of one or more of the following variables: number of cells  $>10$  cells/mm<sup>3</sup>; protein concentration ( $>0.45$  g/l); or glucose concentration ( $<4$  g/l) or  $<40\%$  the simultaneous glycemia. It has been established that CSF ELISA is a sensitive and specific test for the rapid diagnosis of neurobrucellosis (12). A reduction in CSF brucella titer with clinical improvement in a case of meningoencephalitis has been reported (13).

The aim of treatment is to eradicate the infection and to set up a prolonged and combined therapy to prevent relapses (1). The drugs traditionally used alone or in combination are: (i) aminoglycosides such as streptomycin (3,11,12); (ii) tetracycline (11,12); (iii) trimethoprim-sulphamethoxazole (3,4,12); rifampicin (13). More recently quinolones have been shown to be effective against brucella (14) with ciprofloxacin being most commonly used. No single antibiotic is completely effective in eradicating pathogen in vivo and high relapse still ranges from 2-10

following two-drug therapy (14). Combination of streptomycin, tetracycline and sometimes rifampicin have been shown to have a lower relapse rate than individual drug regimens (3). Corticosteroids are used only in patients with arachnoiditis, cranial nerve involvement or spinal cord pathology. Prolonged therapy of three to four months may be necessary in chronic central nervous system involvement (4).

Shakir et al reported cases with bilateral sensorineural high tone hearing loss on audiometry and abnormal brainstem auditory evoked potential (BAEP) (2). They also reported one case in which the ataxia and deafness almost totally disappeared and BAEPs returned to normal. However in most cases the deafness has been irreversible (2,4).

We present this case here because her hearing loss and cerebellar signs were due to neurobrucellosis and improved with treatment. Neurobrucellosis with hearing loss is a rare diagnosis and improvement after treatment is seldom seen and published before as case reports.

### REFERENCES

1. Saleh D, Basim Y, Hassan S, and Jayent P. Neurobrucellosis: clinical characteristics, diagnosis and outcome. *Neurology* 1989;39:498-501.
2. Ratna T, Mohan K, Vel M, Okafor B. Sensorineural hearing loss in neurobrucellosis. *The Journal of Laryngology and Otology* 1993;107:1034-6.
3. Bashir R, Al Kawi MZ, Harder EJ, Jinkins J. Nervous system brucellosis: diagnosis and treatment. *Neurology* 1985;35:1576-81
4. Young EJ. Human brucellosis. *Review of Infectious Disease* 1983;5:821-42.
5. Raad S, Al-Din A, Araj G, Lulu A., Mousa A, and Saadah M. Clinical categories of neurobrucellosis. A report on 19 cases. *Brain* 1987;110:212-23.
6. Al Deeb S, Sharif H, Clark D. Imaging in spinal brucellosis. *J Neurol* 1988;325:48.
7. Mousa ARM, Koshy TS, Araj GF, Marafie AA, Muhtaseb SA, Al-Mudallal DS, Busharetulla MS. Brucella meningitis: presentation, diagnosis and treatment: a prospective study of ten cases. *Q J Med* 1986;233:873-85.
8. Lulu AR, Araj GF, Khateeb MI, Yusuf AR, Mustafa MY, Fenech FF. Human brucellosis in Kuwait: a prospective study of 400 cases. *Q J Med* 1988;249:39-54.
9. Fincham RW, Sahs AL, Joynt RJ. Protean manifestation of nervous system brucellosis. *JAMA* 1993;183:269-75.
10. CE Diaz Espejo, F Viallabos Chaves, and B Sureda Ramis. Chronic intracranial hypertension secondary to neurobrucellosis. *J Neurol* 1987;234:59-61.
11. Araj GF, Lulu AR, Saadah MA, Mousa AM, Strannegard I, Shakir RA. Rapid diagnosis of neurobrucellosis by ELISA. *J Neuroimmunol* 1986;12:173-82.
12. Lorens-Terol J, Busquets RM. Brucellosis treated with rifampicin. *Archives of Diseases in Childhood* 1980;55:486-8.
13. Al-Sibai MB, Halim MA, el-Shaker MM, Khan B, Quadri SM. Efficacy of ciprofloxacin for treatment of *Brucella melitensis* infections. *Antimicrob Agents Chemother* 1992;36:150-2.
14. Conti R, Parenti F. Rifampin therapy for brucellosis, flavobacterium meningitis and cutaneous leishmaniasis. *Rev Infect Dis* 1983;5:600-5.