Sensorineural Hearing Loss in Neurobrucellosis: Case Reports and Literature Review

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ABSTRACT
Brucellosis may occur at almost any anatomic location, but neurobrucellosis is an uncommon complication. Most of these cases present to physicians with symptoms of meningovascular or cranial nerve involvement. The vestibulocochlear nerve is one of the most common cranial nerves involved in neurobrucellosis. Herein, we report five patients suffering from neurobrucellosis with sensorineural hearing loss in whom no potential ototoxic cause could be found to account for the hearing loss except for brucellosis.

Key words: Sensorineural hearing loss, Neurobrucellosis

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INTRODUCTION
Brucellosis is an infection caused by bacteria of the genus Brucella. Human infection results from occupational contact with an infected animal or by ingestion of infected milk, milk products, animal, or tissues. It is reportedly common in and around the Mediterranean countries including certain areas in the Middle East[1-3].

Brucellosis may be asymptomatic with only serologic evidence of infection. Malaise, weakness, fatigue, headache, backache, myalgia, sweats, and chills are often prominent.
Localized disease may occur at almost any anatomic location, but osteomyelitis, splenic abscess, genitourinary tract infection, pulmonary disease, and endocarditis are among the most common. Neurologic complaints are uncommon and include meningoencephalitis, radiculitis and peripheral neuropathy.

Neurobrucellosis is an uncommon complication of brucellosis, the manifestations of which are protean. Most of these cases present to physicians with symptoms of meningovascular or cranial nerve involvement\(^{[1,3,4]}\). The vestibulocochlear nerve is one of the most common cranial nerves involved in neurobrucellosis\(^{[1,2]}\).

**CASE REPORTS**

**Case 1**

A 24-years-old male began to suffer from weakness, malaise, fever, and arthralgia, and five months after the appearance of the symptoms, he was admitted to a medical center and was diagnosed as brucellosis. Rifampicin and tetracycline were given for 40 days as medical therapy. There was a temporary improvement in the symptoms but they failed to resolve. A month after the end of the treatment, visual problems, hearing loss and on the last day confusion were added to the manifestations.

On admission to the infectious diseases department, his temperature was 38.5°C, heart rate 110/min, blood pressure 120/80 mmHg, and respiratory rate 18/min.

He had neck stiffness and bilateral increased deep tendon reflexes, but the neurological examination was otherwise normal. The ear, nose and throat examination was normal.

The patient’s history revealed that his mother was treated for brucellosis during the last year and that the patient had also ingested unpasteurized milk products.

His white blood cell count was 11.5 K/UL, and liver enzymes were high (SGOT: 163 U/L, SGPT: 266 U/L). Sedimentation rate was 8 mm/hour complete blood count (CBC) and blood chemistry results were otherwise normal. Serologic examination determined an agglutination titer for *Brucella* in serum of 1/256 (high). Serum O antigen of *Brucella* (1/1600) and agglutination titer for *Brucella* in cerebrospinal fluid (CSF) (1/256) were also high. Treponemal tests were negative.

On admission, audiometric examination showed bilateral mild, mixed type of hearing loss, but after three months, severe, mixed type hearing loss at all tones was present (Figure 1). The patient was diagnosed as neurobrucellosis and ceftriaxone 4 g/day

![Pure Tone Audiogram](image)

**Figure 1.** Audiometric examination of Case 1.
was given for 30 days. On the 10th day of treatment, trimethoprim-sulfamethoxazole (TMP-SMZ; 1 double-strength tablet containing 160 mg TMP and 800 mg SMZ) peroral (PO) twice daily and on the 25th day, rifampin (600 mg/PO once daily) were added to the treatment protocol. TMP-SMZ (1 double-strength tablet PO twice daily) and rifampin, 600 mg/PO once daily, were offered for six months.

**Case 2**

A 45-years-old female suffering from weakness, malaise, low grade fever, arthralgia, night sweats, and hearing loss for one year was admitted to the infectious diseases department. Her history revealed that she had ingested unpasteurized milk.

Neurologic examination and ear, nose and throat examination were normal.

CBC and biochemical test results were in normal range. Sedimentation rate was normal (12 mm/h).

Cranial magnetic resonance imaging (MRI) showed no pathologic findings. Serologic examination determined an agglutination titer for *Brucella* in serum of 1/256 (high) and an agglutination titer for *Brucella* in CSF of 1/256 (high). Treponemal tests were negative. On admission, audiometric examination showed bilateral mild, mixed type of hearing loss (Figure 2).

The patient was diagnosed as brucellosis and ceftriaxone 4 g/day was administered for 30 days. On the 10th day of treatment, TMP-SMZ (1 double-strength tablet containing 160 mg TMP and 800 mg SMZ, PO twice daily) and on the 25th day rifampin (600 mg/PO once daily) were added to the treatment protocol. TMP-SMZ (1 double-strength tablet/PO twice daily) and rifampin (600 mg/PO once daily) were offered for six months.

**Case 3**

A 46-years-old female began to suffer from weakness, malaise, fever, arthralgia, and hearing loss and was admitted to the infectious diseases department.

The patient’s neurologic and ear, nose and throat examinations were normal. CBC, biochemical test results and sedimentation rate were in normal range. Serologic examination determined serum agglutination titer for *Brucella* of 1/256 (high) and agglutination titer for *Brucella* in CSF of 1/256 (high).

Audiometric examination showed bilateral mild, mixed type of hearing loss (Figure 3).

The patient was diagnosed as neurobrucellosis and treated with tetracycline (2 g/day for 2 months) and streptomycin injections (1 g/day for 10 days).

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**Figure 2. Audiometric examination of Case 2.**

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Case 4 and Case 5

A 60-years-old female and a 69-years-old female diagnosed as neurobrucellosis were consulted to us from the infectious diseases department because of hearing loss. The first patient was treated five months before and the second was treated six months before for neurobrucellosis. The patients started to suffer from hearing loss during the treatment period. Their physical examinations were normal. Audiometric examination showed bilateral mild, sensorineural type of hearing loss (Figures 4, 5).

DISCUSSION

Human brucellosis was described in 1863 by Marston and the etiologic agent was isolated by Bruce[5,6]. There are four species of Brucella that cause infection in humans. The most pathogenic is Brucella melitensis, and it is the most frequent cause of brucellosis. The incubation period of brucellosis usually varies between 7 and 21 days, but may last several months[5,6].

Brucellosis may be asymptomatic with only serologic evidence of infection. Malaise, weakness, fatigue, headache, backache, myalgia, sweats, and chills are often prominent. When physical findings are present, major manifestations are splenomegaly (10-20%), lymphadenopathy (15%) and hepatomegaly (less than 10%). Involvement of the central nervous system or peripheral nervous system occurs in 2-5% of patients[2,7].

Neurobrucellosis has neither a typical clinical picture nor specific CSF findings[4,8]. Diagnosis is made according to CSF findings such as increased proteins, lymphocytosis and high Brucella agglutination titer. In few acute cases, culture for Brucella in CSF may be positive, but negative CSF culture does not rule out neurobrucellosis[3,4,8,9].

Neurobrucellosis can present as meningitis, encephalitis and myelitis[1,3,8,9]. The common pathogenic threat due to neurobrucellosis is the meningeal infection. The cranial nerve involvement in neurobrucellosis has been found as part of the varied neurological presentation[1,2,9]. The cranial nerves commonly involved are the optic, oculomotor, abducent, facial, and vestibulocochlear[1-4,8-10]. Among these, there seems to be a predilection for the vestibulocochlear nerve leading to sensorineural hearing loss[2-4,9]. According to reports, the sensorineural hearing loss may be an incidental finding.

As hearing loss usually occurs early in the course of the disease, it can help with early diagnosis in otherwise asymptomatic patients. Neurobrucellosis may cause hearing loss because of damage to the in-
ner ear and/or vestibulocochlear nerve, which manifests by a perceptive type of hearing loss, tinnitus, dizziness, and reduced response to caloric irrigation. Dizziness and tinnitus are occasionally mentioned in the literature. Specific data regarding the clinical and pathologic findings in ears affected by the *Brucella* organism are missing. Conductive type hearing loss is less common, and may be attributed to granuloma formation, scarring and development of a chronic adhesive process in the middle ear[1,2].

The relationship between brucellosis and sensorineural hearing loss has been seen in many reports. Gladysz et al. reported 147 cases of brucellosis and 52 (33%) of them were found to suffer from hearing loss due to the disease[5]. Shakir et al. reported cases with bilateral sensorineural high tone hearing loss on
audiometry and abnormal brainstem evoked potentials[2]. In most cases, the deafness has been irreversible[2,4]. Osuch et al. reported 120 patients with chronic brucellosis and 93 suffered from hearing loss that was attributed to the disease according to audiological tests[11]. Jezyna et al. reported 150 patients with chronic brucellosis and 72 of them were found to suffer from perceptive type hearing loss[12]. Muszynski et al. reviewed 105 patients suffering from chronic brucellosis, and sensorineural hearing loss related to the disease was found in 64 cases[13]. The authors believe that the inner ear damage was due to Brucella toxin penetrating into the labyrinth and thereby affecting the cochlear nerve.

CONCLUSION

Central nervous system involvement is a serious complication of brucellosis. Patients with illness of short duration usually have a rapid and favorable response to therapy. Those who present with prolonged symptoms lasting several months may respond slower to therapy and have consequent neurological sequelae.

Although the vestibulocochlear nerve is one of the commonest cranial nerves involved in neurobrucellosis, its involvement is a relatively late manifestation in the natural history of the disease. In areas where brucellosis is endemic, otologists have to be alert to the possibility neurobrucellosis in patients with sensorineural hearing loss.

REFERENCES


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