

# Hearing Rehabilitation in Neurobrucellosis by Cochlear Implantation: Case Report

## Nörobruselloziste Kohlear İmplantasyon ile İşitme Rehabilitasyonu

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### ABSTRACT

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Brucellosis is an endemic zoonotic disease which is seen especially in mediterranean countries. Neural involvement of brucellosis occur in 5% of patients so called neurobrucellosis. Hearing loss as a presenting symptom in neurobrucellosis is unusual. In this article we present a case of neurobrucellosis with bilateral progressive sensorineural hearing loss that was treated by cochlear implantation succesfully. Cochlear implantation in neurobrucellosis is discussed with the literature on the subject.

#### *Keywords*

*Cochlear implantation; brucella; deafness*

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### ÖZET

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Brusella endemik zoonotik bir hastalıktır ve genellikle akdeniz ülkelerinde görülür. Brusellada nörolojik tutulum yaklaşık %5 hastada görülür ve bu duruma nörobrusellozis adı verilir. Nörobruselloziste başlangıç şikayeti olarak işitme kaybı görülmesi nadir bir durumdur. Bu çalışmada bilateral ilerleyici sensörinöral işitme kaybı olan ve kohlear implantasyon ile başarılı bir şekilde tedavi edilen bir nörobrusellozis hastası sunuldu. Nörobrusellozis hastalarında kohlear implantasyon literatür bilgileri eşliğinde tartışıldı.

#### *Anahtar Sözcükler*

*Kohlear implantasyon; brusella; sağırılık*

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## INTRODUCTION

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**B**rucellosis is a zoonotic disease which is caused by microorganisms belonging to genus brucella. The disease is endemic in middle east and mediterranean countries.<sup>1</sup> Brucellosis is still the most common zoonotic disease worldwide by 2014. The incidence rate of brucellosis in Turkey ranges from 16 to 30 per 100 000.<sup>2</sup> The most frequent symptoms are arthralgia (73.7%) and fever (72.2%). Antibiotics including doxycycline and streptomycin appeared to be the most effective therapy. Neurological findings occur in less than 5% of the patients, called as neurobrucellosis (NB), with meningitis being the most common clinical manifestation.<sup>3</sup> Cranial neuropathy in NB is not rare and involvement of the vestibulocochlear nerve has been shown in previous studies.<sup>4</sup>

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## CASE REPORT

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A-54-year-old woman was referred to our department with bilateral progressive hearing loss. Two months after the hearing loss she had nausea, vomiting, double vision and disturbed balance complaints. Lumbar puncture was performed and cerebrospinal fluid (CSF) pressure was 28 cm H<sub>2</sub>O. Microscopic examination of CSF showed 52 lymphocytes/mm<sup>3</sup>, 120 mg/dL protein and 30 mg/dL glucose (blood glucose measured simultaneously was 96 mg/dL). The diagnosis of NB was made after the serum brucella agglutination titer of 1/10, CSF brucella agglutination titer of 1/20 and CSF brucella antihuman-globulin titer of 1/160. The cranial magnetic resonance imaging (MRI) showed focal hyperintense lesions in the optic radiatio, centrum semiovale and periventricular white matter which is interpreted as leukoencephalopathy. The patient was treated with a regimen of doxycycline and rifampicin. Ceftriaxone was added after 2 months of initial therapy. The treatment was stopped after the recovery of neurological symptoms and clearance of laboratory tests at 4 months. The first pure tone audiogram (PTA) showed bilateral severe sensorineural hearing loss (SNHL). Tympanogram was normal and there were no stapedius muscle reflexes on both sides. Transient evoked otoacoustic emissions were negative bilaterally. Hearing status did not improve despite the antibiotherapy. The second PTA after 4 months revealed the same result with the first test. There were no auditory brainstem re-

sponses at maximum stimulus levels in both ears. After using hearing aids for 8 months the patient is believed not to benefit from any treatment but the cochlear implant (CI). Temporal MRI and computer tomography did not show any cochlear nerve or inner ear abnormalities. The patient is then thought to be a potential candidate for CI. However, the exact mechanism of the hearing loss was unclear and the durability of the auditory cortex was still a questionmark. A promontorium stimulation test was performed and a positive result was elicited from the left ear. This test helped us to make the decision for CI to the left ear.

The cochlea was patent and a 24-Channel CI512 with Contour Advance electrode was implanted. All electrodes were inserted and activated. Neural response telemetry was performed intraoperatively and electrically evoked compound action potentials were successfully recorded for all electrodes. The patient was discharged on the sixth day without any postoperative complications. The first fitting was performed at the first month after the operation. Free field hearing thresholds were at 40 decibel. Three months postoperatively, free field hearing thresholds were at 30 dB level and the speech discrimination rate was 54%.

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## DISCUSSION

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Brucellosis is the most common zoonotic disease in the world with an incidence of 10-20/100 000 per year.<sup>1</sup> Although it has been eradicated in some countries, brucellosis is still endemic in mediterranean and middle east region. NB occurs in less than 5% of brucellosis patients. Neurological findings may be the only signs of brucellosis for some patients like our case.<sup>5</sup> Antibiotherapy including doxycycline in combination with two or more other drugs such as rifampicin or co-trimoxazole is preferred by most of the clinicians.<sup>6</sup> Clinical response and the return of CSF findings to normal values are the main criteria for the duration of treatment.

SNHL in NB is a popular clinic manifestation and have been studied before.<sup>4</sup> Hearing status should be evaluated in all forms of brucellosis because it may be the only symptom.<sup>7</sup> SNHL may be an isolated symptom however it is thought to be a part of meningeal involvement in most of the cases. Hearing loss is usually bilateral and progressive in neurobrucellosis. In a case report, Cagatay et al. reported a NB patient with neuromuscular weakness and hearing loss in which all neurologic symptoms improved except hearing loss after

antibiotherapy with methylprednisolone.<sup>8</sup> There are also some studies claiming that brucella does not appear to be associated with hearing loss.<sup>9</sup> Nevertheless in a large series of 187 NB patients, hearing loss due to the involvement of vestibulocochlear nerve was found to be 10%.<sup>10</sup> The etiology could be the loss in inner hair cells, neural tissue hypoxia, a defect in transmission from auditory neurons to cochlear nucleus or lesions in central auditory cortex. However the exact mechanism of SNHL in NB have not been clearly identified.

There is not enough data for treatment modalities for the SNHL due to NB. At this point, selecting the right patient for CI comes across as a problem. In 2009,

Guneri et al reported the first CI for NB.<sup>11</sup> They stated that promontorium test can be useful in selecting the patients for CI.

In this article, we report the second successful CI for SNHL due to NB. Because of the unclear mechanism of hearing loss in these patients, performing CI is a challenging decision. We use the promontorium stimulation test as the investigators mentioned previously. SNHL in NB has been mentioned in case reports and there is not enough data in the literature for the evaluation and treatment modalities in these patients. CI may be a good alternative for hearing rehabilitation for selected patients.

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