**Case report**
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# The insidious presentation and clinical polymorphism of neurobrucellosis: About two case reports

Nesrine Ghedamsi[1] ✉, Hajer Derbali[1], Ines Bedoui[1], Meriam Messelmani[1], Najiba Fekih-Mrissa[2], Ridha Mrissa[1]

[1]Department of Neurology, Military Hospital of Instruction of Tunis, Tunisia
[2]Molecular Biology Unit (UR17DN06), Laboratory of Hematology, Military Hospital of Instruction of Tunis, Tunisia

✉ Corresponding author:
nesrine.ghedamsi@gmail.com

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**Introduction**. Even though it is rare, neurobrucellosis is characterized by broad clinical and imaging features. The diagnosis is often late at the cost of irreversible neurological sequelae.

**Case report**. We report two cases of neurobrucellosis. The first case is a 29-year-old man with chronic headache, radiculopathy and diplopia. The second case is a 26-year-old man who presented a progressive gait disturbance and hearing loss. In both cases, magnetic resonance imaging revealed leptomeningeal gadolinium enhancement and the second case had additional bilateral hypersignal of the acoustic-facial bundle. Cerebrospinal fluid analysis showed a positive culture for Brucella and elevated titers of anti-Brucella antibodies. Both patients received combined antibiotic therapy without significant improvement.

**Conclusion**. Our cases highlight the importance of considering Brucella infection in patients with unexplained neurological symptoms in endemic regions, even in the absence of infection symptoms.

**Keywords**: neurobrucellosis, meningo-radiculary neuropathy, vestibulo-cochlear nerve impairment

Referees:
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**Les symptomes et le polymorphisme clinique insidieux de la neurobrucellose : présentation de deux études de cas**

**Introduction**. Bien que rare, la neurobrucellose se caractérise par des aspects cliniques et d'imagerie très variés. Le diagnostic est souvent tardif, causant des séquelles neurologiques irréversibles.

**Étude de cas**. Nous rapportons deux cas de neurobrucellose. Le premier est celui d'un homme de 29 ans souffrant de céphalées chroniques, de radiculopathie et de diplopie. Le second est celui d'un homme de 26 ans ayant présenté des troubles progressifs de la marche et une perte d'audition. Dans les deux cas, l'imagerie par résonance magnétique a révélé un rehaussement leptoméningé au gadolinium, et le second cas présentait un hypersignal bilatéral supplémentaire du faisceau acoustique facial. L'analyse du liquide céphalo-rachidien a révélé une culture positive pour Brucella et des titres élevés d'anticorps anti-Brucella. Les deux patients ont reçu une antibiothérapie combinée sans amélioration significative.

**Conclusion**. Ces deux cas soulignent l'importance d'envisager une infection à Brucella chez les patients présentant des symptômes neurologiques inexpliqués dans les régions endémiques, même en l'absence de symptômes infectieux.

**Mots-clés** : neurobrucellose, neuropathie méningo-radiculaire, atteinte du nerf vestibulo-cochléaire

## Abbreviations

CSF: cerebrospinal fluid
MRI: magnetic resonance imaging
CT-TAP: computed tomography of thorax abdomen pelvis

## 1. Introduction

Since its first description by David Bruce in 1887, brucellosis has been a worldwide widespread anthropozoonosis. It has an incidence of around 500 000 new cases a year [1]. Neurological complications are rare and can be confused with other neurological disorders [2]. Herein we report two cases of neurobrucellosis with different clinical presentations.

## 2. Cases presentation

The first case is a 29-year-old man who consulted for a 6-month history of frontal headache without vomiting, blurred vision nor fever. A few weeks later, he reported a progressive diplopia, radiculalgia, asthenia and weight loss. Clinical examination revealed bilateral abducens nerve paresis and brisk tendon reflexes. T1 gadolinium brain and spinal cord MRI showed leptomeningeal enhancement (Figure 1). Complete blood count and C-reactive protein levels were normal. Rose Bengal serological screening was positive. Opening pressure was normal at lumbar puncture. Cerebrospinal fluid (CSF) analysis revealed 800 cells/µL with lymphocytes predominence (95%), low glucose levels (1.5 mmol/L) and high protein levels (11 g/L). CSF culture isolated a Brucella spp. Positive CSF Brucella-specific IgG antibodies titers were significant (1/160). Electromyoneurography identified L5 lumbar radiculopathy. Associated systemic investigations including computed tomography of thorax abdomen pelvis (CT-TAP) and echocardiography were normal. He received a combined antibiotic therapy (rifampicin 600 mg twice a day, doxycycline 100 mg twice a day) for 12 months. He also received dexamethasone (0.4 mg/kg/d) with an oral relay of corticosteroids. There was no significant clinical improvement and the patient developed erectile dysfunction later.

The second patient is a 26-year-old man who consulted for a 9-month history of urinary incontinence, dysuria, and gait disturbance. Three months later, he developed progressive limbs weakness and hearing loss. No fever was reported. Clinical examination showed a spastic paraparesis, brisk tendon reflexes with bilateral Babinski sign, horizontal nystagmus and hearing loss. T1 gadolinium brain MRI revealed bilateral enhancement of the acoustic-facial bundle associated to leptomeningeal enhancement. Spinal cord imaging was normal (Figure 2). Blood inflammation markers were normal. Serological blood screening were positive for Brucella antibodies. CSF analysis revealed a lymphocytic pleocytosis (45 cells/µL), low glucose (1 mmol/L) and high protein levels (2.25 g/L). CSF Brucella-specific antibodies were positive. CSF culture isolated Brucella spp. The patient's audiogram showed moderate bilateral sensorineural hearing disability (Figure 3). The CT-TAP and echocardiography, as well as systemic investigations, were normal. An intravenous combined antibiotic therapy (rifampicin 600 mg twice a day, trimethoprim-sulfamethoxazole 200 mg per day) associated with dexamethasone (0.4 mg/kg/d) were prescribed followed by oral relay. The antibiotic therapy was maintained for 12 months. The patient had as sequelae a paraparesis and a bladder dysfunction.

## 3. Discussion

Brucellosis is a zoonotic infection that is still endemic in many areas. In Tunisia, brucellosis has increased in prevalence since the past 20 years [3]. It is transmitted from livestock to humans by eating or drinking contaminated animal products or with a direct contact with infected animals. However, in most cases, the way of transmission is not identifiable; as is the case for our patients. The most common symptoms include: fever, general deterioration, arthralgia and myalgia [1]. Fever, though common, is not constantly observed, as it is the case of our patients. Neurological involvement in systemic brucellosis is rare and the incidence is around 0.5% to 25% [2]. It can occur at any stage of the disease, and involve both the central and peripheral nervous system. A broad clinical spectrum of neurobrucellosis is described by Petra et al. in a series of 221 patients [4]. Hearing loss is reported as a common clinical feature, present in

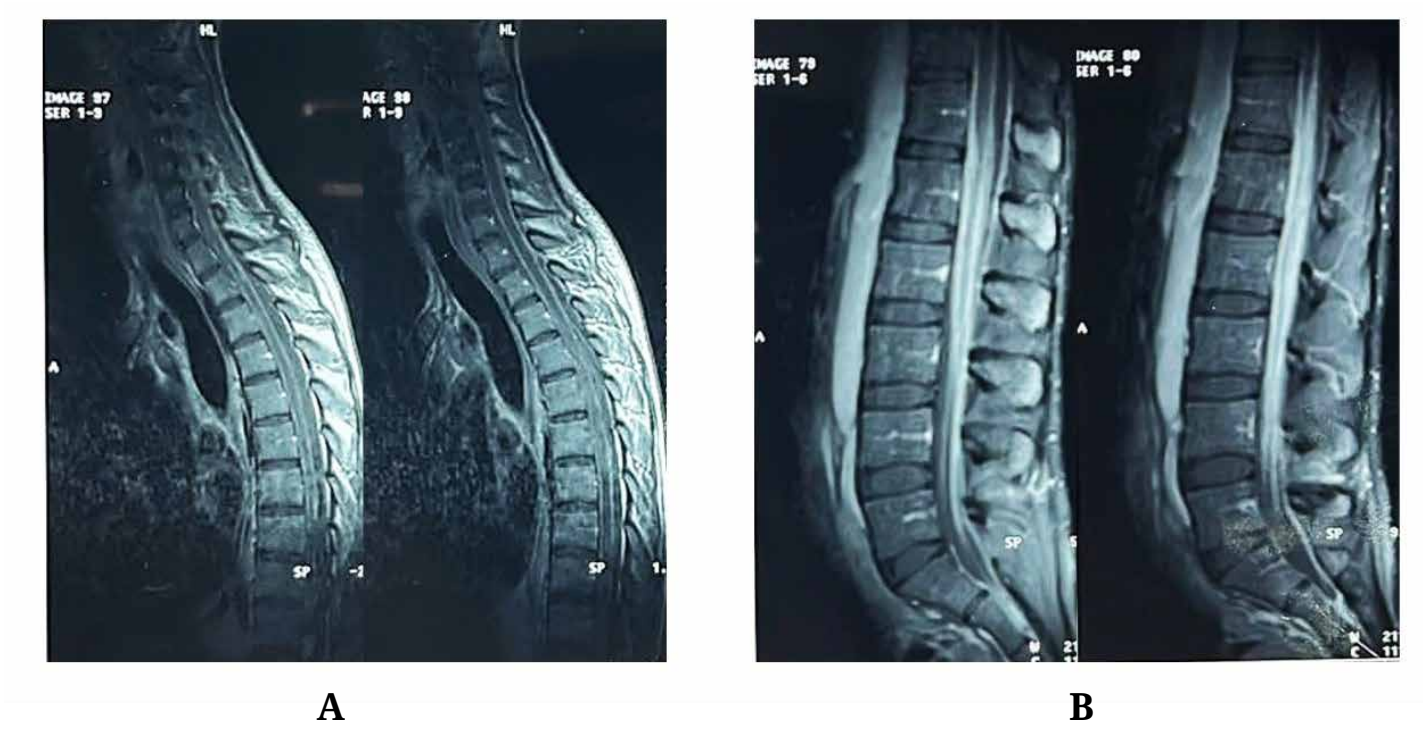


**Figure 1.** Sagittal spinal MRI on T1-weighted with Gadolinium enhancement of the first case report. A: Cervical and dorsal spine. B: Lumbar spine.

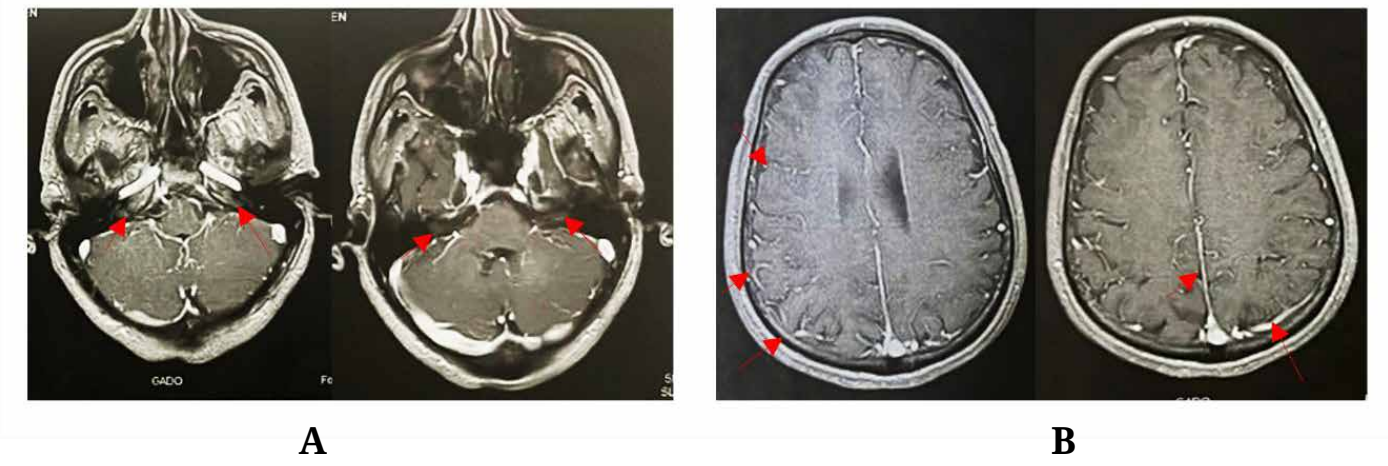


**Figure 2.** Axial brain MRI on T1-weighted with Gadolinium enhancement of the first case report. A: bilateral acoustic-facial bundle. B: leptomeningeal contrast enhancement.

25.8% of cases. Abducens nerve palsy was observed in 8% of patients. Polyradiculopathy and paraparesis were described as major complications of neurobrucellosis (present in 6.8% and 4.5% of patients respectively). Neurobrucellosis' diagnosis is based on clinical features, imaging and biological findings. Isolation of bacteria from blood and/or CSF is the gold standard for positive diagnosis. However, culture positivity is observed in less than 50% of cases [2,4]. Brain and/or spinal cord MRI can be normal or show inflammatory white matter signs, vascular changes, leptomeningeal, cranial nerve or radicular gadolinium enhancement, granulomatous lesions or abscesses [5]. According to the World Health Organization, therapeutic options should include doxycycline (100 mg twice a day) in association with streptomycin (1 g daily) or rifampicin at 15 mg/kg/day (600-900 mg per day). Ceftriaxone or trimethoprim-sulfamethoxazole can be used due to their good diffusion in the CSF. The duration of treatment depends on the clinical presentation and varies from 3 to 12 months depending on the CNS involvement [4]. The first patient received a combined antibiotic therapy with rifampicin and doxycycline and the second one with rifampicin and trimethoprim-sulfamethoxazole. The use of corticosteroids is reserved for complicated neurobrucellosis cases including iritis, papilledema,

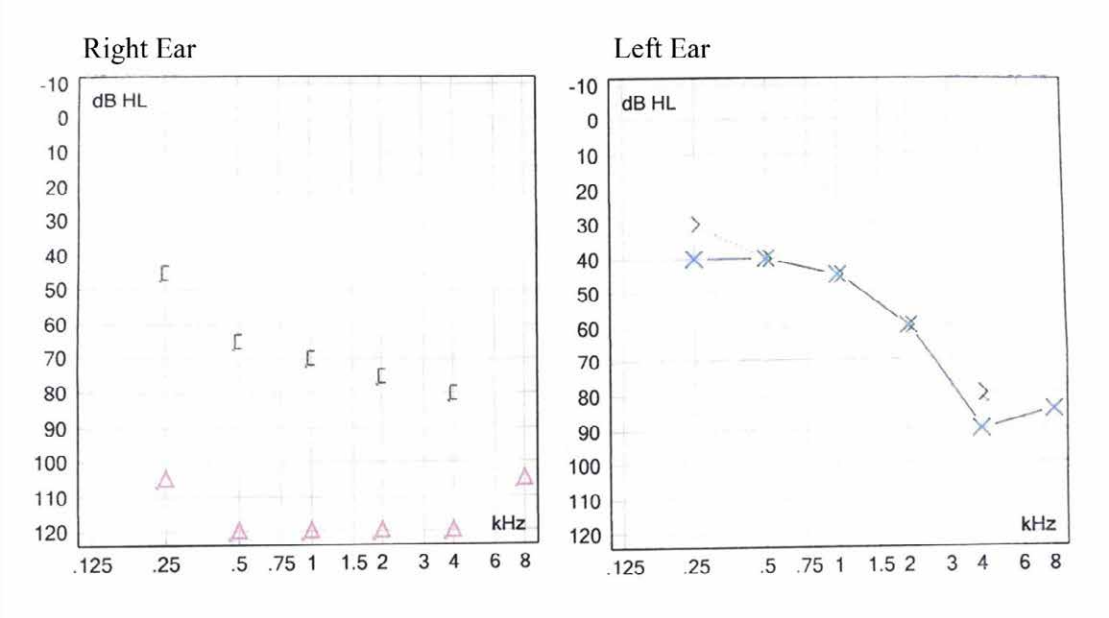


**Figure 3.** Audiogram of the second case report. Bilateral sensorineural hearing loss predominantly on the right ear.

myelopathy, polyneuropathy, and/or cranial nerve palsies [1]. Neurobrucellosis has a low rate of mortality (1%) but sequelae are unfortunately frequent and are related to the delay in diagnosis and treatment [4] as it is the case for our patients.

## 4. Conclusions

Neurobrucellosis is an infectious disease with a broad clinical spectrum that is often difficult to diagnose. Our cases highlight the possibility of cranial nerves' involvement, mainly abducens and chocleovestibular, as potential clinical presentations. Neurobrucellosis should be especially considered in endemic areas, even in the absence of fever or any obvious mode of transmission. Early diagnosis is important and may prevent irreversible sequalae.

## Statements

**Declaration of interest.** The authors declare that they have no conflict of interest.

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