



# Catatonic State as a Rare Presentation of Neurobrucellosis: A Case Report

Vajiheh Aghamollai<sup>1</sup>, Zahra Ahmadinejad<sup>2</sup>, Fatemeh Mohammadian<sup>1</sup> and Zahra Mirsepassi<sup>1\*</sup>

<sup>1</sup>Department of Psychiatry, Tehran University of Medical Sciences, Tehran, Iran

<sup>2</sup>Department of Infectious Diseases, Imam Khomeini Complex Hospital, Tehran University of Medical Sciences, Tehran, Iran

\*Corresponding author: Department of Psychiatry, Tehran University of Medical Sciences, Tehran, Iran. Tel/Fax: +98-2155412222, Email: z-mirsepassi@sina.tums.ac.ir

Received 2019 June 30; Revised 2019 July 14; Accepted 2019 September 01.

## Abstract

**Introduction:** Brucellosis is a zoonotic endemic disease, which is transmitted to the human through occupational exposure or ingestion of contaminated dairy products. Neurobrucellosis has been detected in 5% - 10% of patients with diverse manifestations.

**Case Presentation:** In this study, an atypical case of neurobrucellosis is reported. The patient was a 28-year-old man suffering from psychiatric symptoms and catatonic states. The CSF analysis was unremarkable but the diagnosis was made according to Enzyme-linked Immune Sorbent Assay, other serology tests, and brain MRI findings.

**Conclusions:** It should be mentioned, one of the differential diagnoses in patients with unexplained neurological and psychiatric symptoms in endemic areas is neurobrucellosis and a multidisciplinary approach to these patients is essential.

**Keywords:** Brucellosis, Neurobrucellosis, Psychiatry, Catatonia

## 1. Introduction

Brucellosis is a zoonotic endemic disease in Iran and some other countries (1-4). This infection is transmitted to the human through occupational exposure or consumption of contaminated dairy products (1). A wide range of clinical presentation is seen in brucellosis (1, 5, 6). Neurological presentation and CNS involvement is a rare complication and is detected in 5% - 10% of patients with diverse manifestations (7-9) and can occur in any stage of disease (5).

The most common presentation of neurobrucellosis is meningitis and meningoencephalitis (10). Other manifestations are peripheral neuropathy, radiculopathy, myelitis and cranial nerve involvement (5, 8). Some literature proposed unexplained psychiatric features such as depression, amnesia, psychosis, agitation, personality disorder, and euphoria in neurobrucellosis (5, 11).

This study aimed at reporting an atypical chronic-relapsing form of neurobrucellosis presented with recurrent and refractory psychiatric symptoms and catatonia, which responded dramatically to anti-Brucellosis treatment.

## 2. Case Presentation

A 28-year-old man was admitted to Roozbeh Hospital, a referral psychiatric hospital in Tehran. He was single, 8th grade educated and unemployed. There was a 10-year occupation experience as a slaughterer in his previous history. The current presentation was mutism, negativism, and refusal of food and water. For the first time, he was admitted to a psychiatric hospital three years ago due to irritability, depressed mood, and psychotic symptoms, which were somewhat resolved by symptomatic therapy but he relapsed several times and was admitted again.

The prominent presentation in all admissions was a catatonic state treated by electroconvulsive therapy (ECT), but relapses sometimes occurred later.

In the current admission, the prominent presentation was a catatonic state, so treating by ECT was recommended. He received ECT (6 sessions) and olanzapine (20-milligram daily), which resulted in a partial response. Although we observed a preliminary mild improvement, his condition deteriorated again after 2 weeks. He demonstrated irritability, disorganized behavior, physical and verbal aggression and staring attacks, which lasted only a few seconds. In the neurological consultation, he was hypersensitive and agitated. There was marked impairment in some cognitive domains, including attention, orientation, and con-

centration. Because of staring attacks and a history of convulsion in his brother, an electroencephalogram (EEG) was performed and owing to posterior epileptiform activity, he was treated by carbamazepine (600 mg per day), after which the attacks subsided.

The patient obtained 8 out of 30 in Montreal Cognitive Assessment (MoCA) test with a significant impairment in executive function, attention, and recall domain. Other neurological examinations were unremarkable, except generalized hyperreflexia.

In the brain MRI, bilateral symmetric signal changes were reported in basal ganglia.

All of these findings, including the history of recurrent episodes of nocturnal fever and sweating, the past job as a slaughterer and living in the endemic part of Iran (Arak city), compelled us to consider infectious diseases, especially brucellosis, as the main differential diagnosis and necessary lab tests were performed. Laboratory findings are summarized in Table 1.

Although Wright and 2ME tests were unremarkable, based on the high suspicion of neurobrucellosis the ELISA IgG, IgM was requested in both serum and CSF, which were positive for brucellosis.

Consultation with an infectious disease specialist was done who advised specific antibiotic therapy for neurobrucellosis.

About 2 weeks after antibiotic therapy with doxycycline (100 mg twice daily), rifampicin (600 mg daily), ceftriaxone (2 g twice daily), olanzapine (20 mg daily), and carbamazepine (600 mg daily), he was completely oriented and his irritability and aggression were reduced dramatically and the cognitive state was partially improved. He obtained 14/30 in MoCA test. The losing points were detected, especially in attention, executive function, and recall. The anti-Brucellosis treatment was continued for nine months. We followed up the patient for two years. The patient did not experience any psychotic or catatonic symptoms in this period.

### 3. Discussion

This patient was an unusual case of neurobrucellosis. He experienced multiple relapses with psychiatric presentation and catatonic state.

Neurobrucellosis has diverse manifestations such as headache, meningitis, focal neurologic deficits, coma, cognitive decline as well as psychiatric presentations, including Psychosis, depression, and behavioral changes (1, 3, 5, 6, 8, 11, 12).

One study reported depression in about 38.5% of patients without obvious neurological symptoms (13).

**Table 1.** Laboratory Blood and CSF Tests

Test	Results
<b>Serology</b>	
ESR	3
CRP	Negative
Wright	1/40
2ME	1/20
Anti-Brucella Ab (IgM)	0.5
	(< 0.8: Negative; 0.8 - 1.1: Borderline; >1.1 Positive)
Anti-Brucella Ab (IgG)	72.7
	(< 16: Negative; 16 - 22: Borderline; ≥ 22: Positive)
HIV Ab	Negative
<b>CSF</b>	
Colour	Colourless
Appearance	Clear
WBC	1
RBC	1
Protein	26
Glucose	63
Wright	Negative
2ME	Negative
ADA	16
Anti-Brucella Ab (IgM)	Negative
Anti-Brucella Ab (IgG)	Positive
ACE	3.3
MTB PCR	Negative
PCR for HIV	Negative
Cryptococcus PCR	Negative

Although psychiatric manifestations have been more commonly reported in chronic brucellosis, they can be a presenting symptom in the acute stage of illness (5).

Epileptic seizures also are rare in brucellosis. In a case series of 6 patients, 2 cases had no seizure but most of them showed grandmal seizures (8, 14).

The differential diagnoses were an inflammatory disease, an autoimmune disease or a chronic infection, including tuberculosis. So, CSF evaluation, brain MRI and serology tests were performed to rule out these diseases.

The diagnosis of neurobrucellosis is based on clinical and epidemiological evidence and should be confirmed by serologic or microbiologic tests. The widely used diagnostic laboratory tests are standard serum agglutination tests (wright, wright coombs and 2ME) and Western blot and ELISA for the detection of IgG and IgM, which is the test of

choice for complicated and chronic cases (Araj, 2010).

According to the patient's occupational history (slaughterer), living in an endemic part of Iran (Arak city), and the high suspicion to brucellosis, serology tests, including serum agglutination tests, were performed which were unremarkable, but positive ELISA IgG in serum and CSF was confirmed neurobrucellosis in our case.

Some literature suggested relapse could be diagnosed by the detection of IgG and IgA antibodies but not IgM, which was confirmed in our patient (15, 16). This was also mentioned in other studies that ELISA could be more sensitive (5), especially in relapsing cases such as our patient (17).

This case was considered a chronic relapsing form of neurobrucellosis, which is one of the most complicated features of the disease. Some risk factors for this condition have been noted, including delayed (> 10 days) or ineffective treatment (18).

Pure psychiatric presentation in neurobrucellosis is very rare and depression is the most common psychiatric symptom (13). Based on our research and literature review, a recurrent catatonic state similar to this patient has not been reported yet.

Our patient was an atypical case of a chronic relapsing form of neurobrucellosis. He experienced episodes of neuropsychiatric symptoms and catatonic states in a chronic manner. The CSF analysis and even the serology tests, including indirect coombs, were unremarkable and only ELISA for IgM and IgG antibodies yielded the correct diagnosis. Another considerable point in the presented case was abnormal signal changes in basal ganglia in the brain MRI, which have been rarely reported in neurobrucellosis.

It should be mentioned, in the endemic areas, one of the differential diagnoses in patients with unexplained neurological or psychiatric symptoms is neurobrucellosis even when the serological studies are negative.

## Footnotes

**Authors' Contribution:** Vajihah Aghamollai, Fatemeh Mohammadian, Zahra Ahmadinejad, and Zahra Mirsepassi did the literature review. Vajihah Aghamollai, Zahra Mirsepassi, and Fatemeh Mohammadian drafted the manuscript. Vajihah Aghamollai, Fatemeh Mohammadian, and Zahra Mirsepassi collected the clinical data and presented the case. All authors read and approved the final manuscript.

**Declaration of Interest:** The authors state that there are no conflicts of interest.

**Funding/Support:** No funding was received for this research.

## References

- Bennett J, Raphael D, Blaser MJ. *Mandell Douglas and Bennett's principles and practice of infectious diseases*. 35. 8th ed. The American Journal of Tropical Medicine and Hygiene; 2015.
- Jiao LD, Chu CB, Kumar CJ, Cui J, Wang XL, Wu LY, et al. Clinical and laboratory findings of nonacute neurobrucellosis. *Chin Med J (Engl)*. 2015;**128**(13):1831-3. doi: [10.4103/0366-6999.159362](https://doi.org/10.4103/0366-6999.159362). [PubMed: [26112728](https://pubmed.ncbi.nlm.nih.gov/26112728/)]. [PubMed Central: [PMC4733728](https://pubmed.ncbi.nlm.nih.gov/PMC4733728/)].
- Gul HC, Erdem H, Bek S. Overview of neurobrucellosis: A pooled analysis of 187 cases. *Int J Infect Dis*. 2009;**13**(6):e339-43. doi: [10.1016/j.ijid.2009.02.015](https://doi.org/10.1016/j.ijid.2009.02.015). [PubMed: [19428283](https://pubmed.ncbi.nlm.nih.gov/19428283/)].
- Pappas G, Akritidis N, Bosilkovski M, Tsianos E. Brucellosis. *N Engl J Med*. 2005;**352**(22):2325-36. doi: [10.1056/NEJMra050570](https://doi.org/10.1056/NEJMra050570). [PubMed: [15930423](https://pubmed.ncbi.nlm.nih.gov/15930423/)].
- Güven T, Ugurlu K, Ergonul O, Celikbas AK, Gok SE, Comoglu S, et al. Neurobrucellosis: clinical and diagnostic features. *Clin Infect Dis*. 2013;**56**(10):1407-12. doi: [10.1093/cid/cit072](https://doi.org/10.1093/cid/cit072). [PubMed: [23446629](https://pubmed.ncbi.nlm.nih.gov/23446629/)].
- McLean DR, Russell N, Khan MY. Neurobrucellosis: Clinical and therapeutic features. *Clin Infect Dis*. 1992;**15**(4):582-90. doi: [10.1093/clind/15.4.582](https://doi.org/10.1093/clind/15.4.582). [PubMed: [1420670](https://pubmed.ncbi.nlm.nih.gov/1420670/)].
- Ghaffarpour M, Khoshroo A, Harirchian MH, Sikaroodi H, Pourmahmoodian H, Jafari S. Imaging aspects of brucellosis with and without neurological involvement. *Acta Med Iran*. 2007;**45**(1):63-8.
- Eren S, Bayam G, Ergonul O, Celikbas A, Pazvantoglu O, Baykam N, et al. Cognitive and emotional changes in neurobrucellosis. *J Infect*. 2006;**53**(3):184-9. doi: [10.1016/j.jinf.2005.10.029](https://doi.org/10.1016/j.jinf.2005.10.029). [PubMed: [16647757](https://pubmed.ncbi.nlm.nih.gov/16647757/)].
- Yetkin MA, Bulut C, Erdinc FS, Oral B, Tulek N. Evaluation of the clinical presentations in neurobrucellosis. *Int J Infect Dis*. 2006;**10**(6):446-52. doi: [10.1016/j.ijid.2006.05.007](https://doi.org/10.1016/j.ijid.2006.05.007). [PubMed: [16914346](https://pubmed.ncbi.nlm.nih.gov/16914346/)].
- Ranjbar M, Rezaiee AA, Hashemi SH, Mehdipour S. Neurobrucellosis: Report of a rare disease in 20 Iranian patients referred to a tertiary hospital. *East Mediterr Health J*. 2009;**15**(1):143-8. [PubMed: [19469437](https://pubmed.ncbi.nlm.nih.gov/19469437/)].
- Tuncel D, Uçmak H, Gokce M, Utku U. Neurobrucellosis. *Eur J Gen Med*. 2008;**5**(4):245-8. doi: [10.29333/ejgm/82616](https://doi.org/10.29333/ejgm/82616).
- Haji-Abdolbagi M, Rasooli-Nejad M, Jafari S, Hasibi M, Soudbakhsh A. Clinical and laboratory findings in neurobrucellosis: Review of 31 cases. *Arch Iran Med*. 2008;**11**(1):21-5. [PubMed: [18154418](https://pubmed.ncbi.nlm.nih.gov/18154418/)].
- Shehata GA, Abdel-Baky L, Rashed H, Elamin H. Neuropsychiatric evaluation of patients with brucellosis. *J Neurovirol*. 2010;**16**(1):48-55. doi: [10.3109/13550280903586386](https://doi.org/10.3109/13550280903586386). [PubMed: [20151851](https://pubmed.ncbi.nlm.nih.gov/20151851/)].
- Yilmaz M, Ozaras R, Ozturk R, Mert A, Tabak F, Aktuglu Y. Epileptic seizure: An atypical presentation in an adolescent boy with neurobrucellosis. *Scand J Infect Dis*. 2002;**34**(8):623-5. doi: [10.1080/00365540210147561](https://doi.org/10.1080/00365540210147561). [PubMed: [12238582](https://pubmed.ncbi.nlm.nih.gov/12238582/)].
- Araj GF. Update on laboratory diagnosis of human brucellosis. *Int J Antimicrob Agents*. 2010;**36** Suppl 1:S12-7. doi: [10.1016/j.ijantimicag.2010.06.014](https://doi.org/10.1016/j.ijantimicag.2010.06.014). [PubMed: [20692128](https://pubmed.ncbi.nlm.nih.gov/20692128/)].
- Shoaei SD, Bidi N. Serologic evaluation of brucellosis in patients with psychiatric disorders. *Caspian J Intern Med*. 2012;**3**(4):557-8. [PubMed: [24009935](https://pubmed.ncbi.nlm.nih.gov/24009935/)]. [PubMed Central: [PMC3755865](https://pubmed.ncbi.nlm.nih.gov/PMC3755865/)].
- Ulukavak Ciftci T, Kokturk N, Demir N, Oguzulgen KI, Ekim N. [Comparison of three clinical prediction rules among patients with suspected pulmonary embolism]. *Tuberk Toraks*. 2005;**53**(3):252-8. Turkish. [PubMed: [16258884](https://pubmed.ncbi.nlm.nih.gov/16258884/)].
- Buzgan T, Karahocagil MK, Irmak H, Baran AI, Karsen H, Evirgen O, et al. Clinical manifestations and complications in 1028 cases of brucellosis: A retrospective evaluation and review of the literature. *Int J Infect Dis*. 2010;**14**(6):e469-78. doi: [10.1016/j.ijid.2009.06.031](https://doi.org/10.1016/j.ijid.2009.06.031). [PubMed: [19910232](https://pubmed.ncbi.nlm.nih.gov/19910232/)].