

A Neurobrucellosis Case Presenting with Bilateral Facial Nerve Palsy and Peripheral Neuropathy

Bilateral Fasiyal Sinir Paralizi ve Periferik Nöropati ile Başvuran Bir Nörobruselloz Olgusu

Sanem YILMAZ,^a
Gül SERDAROĞLU,^a
Semra ŞEN,^b
Sarenur GÖKBEN^a

^aDivision of Child Neurology,

^bDivision of Pediatric Infectious Diseases,
Ege University Faculty of Medicine,
İzmir

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Yazışma Adresi/Correspondence:
Sanem YILMAZ
Ege University Faculty of Medicine,
Division of Child Neurology, İzmir,
TÜRKİYE/TURKEY
sanem.yilmaz@ege.edu.tr

ABSTRACT Neurobrucellosis is an uncommon complication of pediatric brucellosis. Direct invasion of the central nervous system occurs in about 5% of the cases of *Brucella melitensis* infection. Meningitis or meningoencephalitis are the most common manifestations. Neurobrucellosis may rarely affect second, third, sixth, seventh and the eighth cranial nerves. Peripheral nerve involvement is also rarely seen in pediatric neurobrucellosis. Here we reported is a twelve years old girl presented with bilateral peripheral facial nerve palsy and peripheral neuropathy. This case is an unusual presentation of neurobrucellosis in pediatric population. Our report emphasizes that neurobrucellosis should be kept in mind in the cases with atypical neurologic manifestations in endemic regions.

Key Words: Brucellosis; peripheral nervous system diseases; facial paralysis; child

ÖZET Nörobruselloz çocukluk çağı brusellozunun nadir bir komplikasyonudur. Santral sinir sisteminin direkt invazyonu *Brucella melitensis* enfeksiyonlarında ancak %5 oranında görülür. Menenjit ya da meningoensefalit nörobrusellozun en sık görülme şeklidir. Nörobruselloz nadiren ikinci, üçüncü, altıncı, yedinci, sekizinci kafa çiftlerini etkiler. Periferik sinir tutulumu çocukluk çağı nörobrusellozunda nadir olarak görülen bir diğer bulgudur. Burada bilateral periferik fasiyal sinir paralizi ve periferik nöropati ile başvuran ve nörobruselloz tanısı alan oniki yaşında bir kız olgu sunulmaktadır. Bu olgu, özellikle endemik bölgelerde, atipik nörolojik bulgularla gelen olgularda nörobrusellozun akılda tutulması gerektiğini vurgulamaları açısından önemli bulunarak sunulmuştur.

Anahtar Kelimeler: Bruselloz; periferik sinir sistemi hastalıkları; fasiyal paralizi; çocuk

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Brucellosis is an infectious disease caused by gram-negative bacteria of the genus *Brucella* and it is a disease of animals. Involvement of the gastrointestinal, hepatobiliary, and skeletal systems has been reported frequently in the literature. Involvement of the nervous system is relatively uncommon and has been reported in only 3-25% of cases of systemic brucellosis.^{1,2} Direct invasion of the central nervous system occurs in about 5% of the cases of *Brucella melitensis* infection. Meningitis or meningoencephalitis are the most common manifestations. Other central nervous system manifestations of brucellosis include cerebral vasculitis, mycotic aneurysms, brain and epidural abscesses, infarcts, haemorrhage and cerebellar ataxia.^{3,4} There may be also peripheral nerve complications. These in-

clude neuropathy, radiculopathy, Guillain-Barre syndrome and a poliomyelitis-like syndrome. Neurobrucellosis can effect both the central and the peripheral nervous system. Here we present a rare case of pediatric neurobrucellosis with the involvement of both central and peripheral nervous system.

CASE REPORT

A twelve years old female patient was admitted to the hospital with backpain, weakness at the lower extremities and difficulty of closing her eyes and chewing food. She could not walk independently and had weakness at the lower exremities. She had difficulty to close her both eyes and to chew. Bilateral peripheral facial palsy was determined



FIGURE 1: The image of the case indicating bilateral facial palsy with inability to close her eyes as well as bilateral facial weakness.

(Figure 1). The other cranial nerve examinations were normal. Muscle strength was 4/5 in the upper extremities and 3/5 in the lower extremities. Deep tendon reflexes were preserved. Proprioception in the lower extremities were normal and she did not have any sensory problems. Laboratory examination showed normal complete blood count values and sedimentation rate. Serum glucose level, serum electrolytes, liver and renal enzymes were within the normal ranges as well as B12 vitamin and thyroid function tests. Craniospinal magnetic resonance imaging (MRI) was normal. The lack of any sensory deficits and the normal craniospinal MRI findings excluded the possible diagnosis of transverse myelitis. Electromyography was consistent with axonal sensorimotor polyneuropathy (Table 1). Serological tests including EBV Ig M, CMV Ig M, hepatitis A,B,C Ig M, Parvo B19 IgM, HIV and Lyme disease were negative. Serum Rose Bengal test was positive. Serum *Brucella* Wright agglutination test was 1/640 (Normal value: <1/320). 2-mercaptoethanol titer was 1/640 (normal value: <1/160). Lumbar puncture could not be performed because the parents refused to sign the informed consent form of the procedure. Based on the clinical and laboratory findings, the patient was diagnosed with neurobrucellosis and was treated with triple antibiotic therapy of rifampicin (600 mg/day)+ doxycycline (200 mg/day) for 8 weeks and gentamycin (160 mg/day) for 14 days. After two

TABLE 1: The nerve conduction study and needle electromyography findings of the case.

Nerve conduction study	Amplitude mV (Right)	Distal latency msec (Right)	Proximal latency msec (Right)	Conduction velocity m/sec (Right)
Median sensory (1 st finger)	Not recordable			
Ulnar sensory	Not recordable			
Median motor	3	11.4	15.4	46
Ulnar motor	4	6.6	11.3	47
Median sensory (2 nd finger)	Not recordable			
Radial sensory	Not recordable			
Fibular motor	1	15.1	23.8	38
Tibialis posterior sensory	3	12.3	23.1	34
Sural nerve	Not recordable			
Needle electromyography	Denervation potentials		Reduced recruitment	
	Fibrillation	Positive waves		
M. tibialis anterior (Right)	2+	2+		+

weeks of therapy muscle strength was 5/5 both in upper and lower extremities and facial palsy fully recovered. The patient did not have any other complaints at her 1-year follow-up period.

An informed consent was taken from the parents of the patient.

DISCUSSION

Brucellosis is endemic especially in Mediterranean countries and in Turkey. The prevalence of *Brucella* seropositivity varies from 2.6% to 14.4% in Turkey.¹ Neurobrucellosis is a rare complication of brucellosis and occurs at a very low frequency in the pediatric age population.² It is known that 0.8% of pediatric *Brucella* cases affect the central nervous system.³ Neurobrucellosis is frequently observed with meningitis. Meningoencephalitis, myelitis, cranial nerve palsies, radiculopathy and neuropathy are the other neurologic complications. The clinical presentation of central nervous system varies; headache and depression, behavioral changes has been reported.^{5,6} Guven et al. reported headache, blurred vision, hearing loss and confusion significantly more common in neurobrucellosis cases than non-neurobrucellosis cases.⁶ Neurobrucellosis most frequently leads to eighth nerve palsy and can also involve second, sixth, and seventh cranial nerves. Gul et al. reported 36 cases with cranial nerve involvement in 187 neurobrucellosis cases.⁴ In their series facial nerve involvement was in only 3 cases (%1). In the literature, there are reports of several cases that involve concomitant optical, abducent, and eighth nerve paralysis; optical and abducent nerve paralysis; facial and vestibulocochlear nerve paralysis.⁵ Guven et al. reported cranial nerve involvement in 19% of 48 cases;⁶ vestibulocochlear in 5 patients, abducens in 2 patients, and facial in 2 patients. One of their patients left with a sequela of peripheral facial palsy.

Unilateral facial nerve palsy is relatively common, with an incidence of around 25 per 100.000 population. Bilateral facial nerve palsy is very rare with an incidence of 1 per 5.000.000 population.⁷ There are many causes of bilateral facial nerve palsy and the most common are idiopathic (Bell's)

palsy, Guillain-Barré syndrome, diabetes mellitus, infectious causes such as bacterial meningitis, infectious mononucleosis, sarcoidosis and human immunodeficiency virus infection. Other causes such as Lyme disease, syphilis and leprosy are documented in the literature but rarely seen.⁸ Neurobrucellosis is also a very rare cause of bilateral facial nerve palsy.

Gul et al. also reported polyneuropathy/radiculopathy in 7% of their cases.⁴ This case is notable because this is the first pediatric case with peripheral neuropathy and bilateral facial nerve palsy concurrently. Peripheral nerve involvement is also rarely seen in pediatric population. Guven et al. reported peripheral neuropathy in 6 of 48 neurobrucellosis cases over 16 years of age.⁶

Different therapy regimens based on ceftriaxone, rifampicin, doxycycline, cotrimoxazole and streptomycin were reported in literature. Guven et al. applied ceftriaxone and rifampicin for 3 weeks and continued with rifampicin and doxycycline for a total of six months.⁶ They reported that they had no recurrence in their cases. Karsen et al. used ceftriaxone+ doxycycline +rifampicin or sulfamethoxazole for 2-4 weeks followed by doxycycline +rifampicin for four weeks.⁹ All of their cases who received ceftriaxone+doxycycline and rifampicin combination for four weeks had normal cerebrospinal fluid findings after the therapy. Shakir et al. suggested the use of steroids in the cases with myelitis, cranial nerve palsy and spinal cord involvement.² Although the effectiveness of steroids has not completely been proven in neurobrucellosis, some authors apply pulse steroid for 3-5 days, then continue with oral steroids for one or two months. Our case did not use steroid therapy but had benefit from the combination of rifampicin+doxycycline and gentamycin. Her neurologic manifestations regressed in weeks.

Neurobrucellosis may appear with different clinical manifestations and neurologic symptoms. Pediatricians especially those working in endemic areas should keep in mind that neurobrucellosis can be involved in children with unexplained neurologic symptoms.

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