Journal of the Neurological Sciences 385 (2018) 144–145

Contents lists available at ScienceDirect

Journal of the Neurological Sciences

journal homepage: www.elsevier.com/locate/jns

Letter to the Editor

Spastic paraparesis and sensorineural hearing loss: keep brucellosis in mind

ARTICLE INFO

Keywords:
Brucellosis
Hearing loss
Neurobrucellosis
Spastic paraparesis
Spinal cord

Dear Editor,

We read with interest the article by Gündüz et al. [1], which was published in your journal earlier this year. The authors report a rare case of isolated spinal cord involvement with normal magnetic resonance imaging (MRI) in a patient with neurobrucellosis, and review the literature for prior similar cases. Their paper illustrates the challenges of diagnosing neurobrucellosis, and highlights the importance of keeping brucellar infection in mind when evaluating a patient with acquired spastic paraparesis, with or without abnormal imaging findings.

Brucellosis is a multisystem zoonosis with a broad spectrum of clinical manifestations that may masquerade as many other disorders and can affect any organ system [2]. Neurobrucellosis can appear at any stage of the disease and has a wide range of presentations, including meningitis, encephalitis, stroke, radiculitis, myelitis, peripheral neuropathies and neuropsychiatric symptoms [3]. Imaging findings are variable and often non-specific, and may mimic other infectious or inflammatory conditions; in addition, there is limited correlation between clinical and imaging features, and a normal imaging study can be found despite positive clinical findings [4].

Although rarely fatal in humans, brucellosis can be a severely disabling disease, and early diagnosis and treatment are crucial to avoid permanent sequelae. Its protean clinical manifestations, however, make it a diagnostic challenge, and despite being endemic in many countries, it remains under-diagnosed and under-reported [5].

We report a case of neurobrucellosis in a 29-year-old Portuguese male patient who presented with a one-year history of progressive difficulty walking. Three years earlier he had experienced a self-limited episode of headache and incoercible vomiting. Gastrointestinal studies at the time yielded negative results, but occasional vomiting persisted. Three weeks after this initial episode he had experienced an episode of monocular vision loss which completely resolved after approximately three weeks, and two years before admission he had suffered sudden and sequential hearing loss affecting both ears within one week. He had also experienced several episodes of transient paresthaesiae. No fever was reported and family history was irrelevant. The patient denied eating or drinking any unpasteurised or raw dairy products. However, he was a farmer and worked in close contact with sheep, goats and cattle; he also slaughtered the animals and handled raw meat while not wearing protective gear.

Neurological examination revealed profound sensorineural deafness, as well as spastic paraparesis, brisk tendon reflexes, bilateral ankle clonus and extensor plantar responses. No sensory deficit or sphincter abnormalities were present. Nerve conduction studies were normal. Auditory evoked potentials were absent and lower limb somatosensory evoked potentials (SEP) showed bilateral and symmetrical compromise of somatosensory pathways, with normal upper limb SEP, which in the clinical context suggested spinal cord involvement below the cervicothoracic transition. Brain MRI (Fig. 1A and B) showed diffuse leptomeningeal enhancement, mainly in the IV ventricle and basal cisterns, suggesting chronic leptomeningitis. No spinal cord changes were found, but dural enhancement was evident on spinal MRI (Fig. 1C).

Serum Rose Bengal test was positive. Cerebrospinal fluid (CSF) studies revealed lymphocytic pleocytosis (78/μL), low glucose (24 mg/dL), elevated protein (640 mg/dL) and oligoclonal IgG bands. Positive CSF *Brucella*-specific IgG and IgA antibodies were detected using an enzyme-linked immunosorbent assay. Upon further questioning, the patient revealed that several of his livestock were discovered to be infected with brucellosis in recent years, which had eventually led to his business closing.

A diagnosis of chronic neurobrucellosis was made and the patient was treated with a combination of rifampicin, doxycycline and trimethoprim-sulfamethoxazole. Periodic clinical and CSF examinations were performed and used to guide duration of treatment, which was continued up to 18 months until CSF findings resolved. There was, however, no significant clinical improvement.

Timely and accurate diagnosis of brucellosis continues to elude clinicians, with potentially devastating consequences. Acute infection frequently goes undetected, and the disease often runs an indolent course that can lead to irreversible sequelae [5].

Our patient initially presented with non-specific findings which were followed by sequential appearance of several neurological complaints, but only spinal cord symptoms prompted neurological assessment. In this case, hearing loss, which preceded paraparesis by more than two years, was an important clue for diagnosis. Cranial neuropathies are common in neurobrucellosis; the VIII nerve is the most frequently involved, and hearing loss is a well-known complication [6]. This, along with the patient's epidemiological history, pointed to the correct diagnosis, albeit with a significant

https://doi.org/10.1016/j.jns.2017.12.036
Received 25 September 2017; Received in revised form 23 November 2017; Accepted 28 December 2017
Available online 29 December 2017
0022-510X/

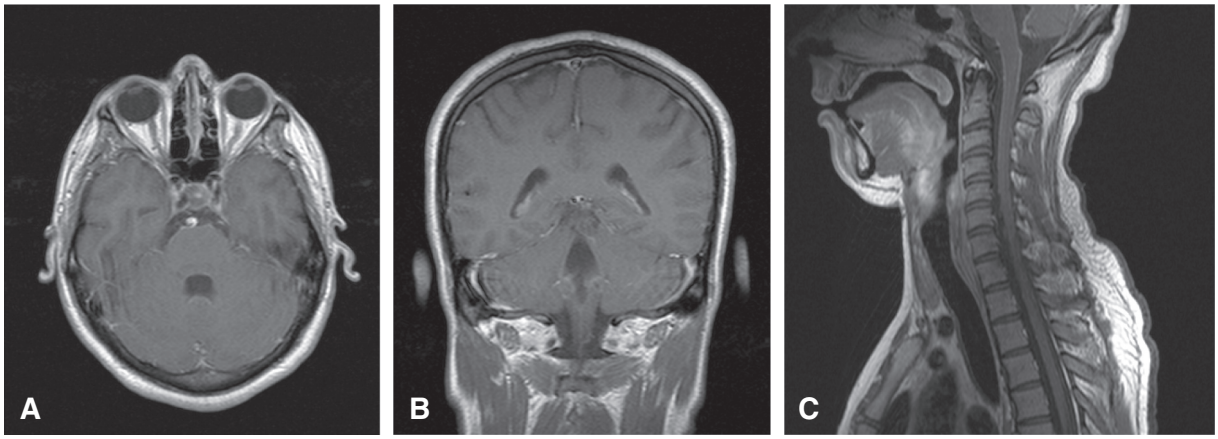

Fig. 1. Magnetic resonance images of the brain and spinal cord showing suggestive findings of chronic leptomeningitis. Axial (A), coronal (B) and sagittal (C) gadolinium-enhanced T1-weighted images showing diffuse leptomeningeal enhancement, particularly in the IV ventricle and basal cisterns, and also dural enhancement in the cervical region of the spinal cord.

delay.

This case highlights that in endemic regions, threshold for suspicion of brucellar infection should be low, and this entity should be considered in the differential diagnosis of chronic inflammatory CSF changes and unexplained neurological symptoms, even in the absence of fever, as early diagnosis and treatment are essential to avoid persistent neurologic deficits. Additional neurological findings, such as hearing loss, can provide important diagnostic clues.

Funding information

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflicts of interest

None.

References

[1] T. Gündüz, P.T. Tektürk, Z. Yapıcı, M. Kürtüncü, A. Somer, S.H. Törün, M. Eraksoy, Characteristics of isolated spinal cord involvement in neurobrucellosis with no corresponding MRI activity: a case report and review of the literature, J. Neurol. Sci. 372 (2017) 305–306, http://dx.doi.org/10.1016/j.jns.2016.11.076.
[2] G. Pappas, N. Akritidis, M. Bosilkovski, E. Tsianos, Brucellosis, N. Engl. J. Med. 352 (2005) 2325–2336, http://dx.doi.org/10.1056/NEJMra050570.
[3] T. Guven, K. Ugurlu, O. Ergonul, A.K. Celikbas, S.E. Gok, S. Comoglu, N. Baykam, B. Dokuzoguz, Neurobrucellosis: clinical and diagnostic features, Clin. Infect. Dis. 56 (2013) 1407–1412, http://dx.doi.org/10.1093/cid/cit072.
[4] M.W. Al-Sous, S. Bohlega, M.Z. Al-Kawi, J. Alwatban, D.R. McLean, Neurobrucellosis: clinical and neuroimaging correlation, Am. J. Neuroradiol. 25 (2004) 395–401.
[5] M.P. Franco, M. Mulder, R.H. Gilman, H.L. Smits, Human brucellosis, Lancet Infect. Dis. 7 (2007) 775–786, http://dx.doi.org/10.1016/S1473-3099(07)70286-4.
[6] H.C. Gul, H. Erdem, S. Bek, Overview of neurobrucellosis: a pooled analysis of 187 cases, Int. J. Infect. Dis. 13 (2009) e339–43, http://dx.doi.org/10.1016/j.ijid.2009.02.015.

Sara P. Dias*, João Sequeira, Manuel Almeida
Department of Neurology, Centro Hospitalar de Lisboa Central, Hospital dos Capuchos, Alameda Santo António dos Capuchos, 1169-050 Lisbon, Portugal
E-mail address: sara.dias@chlc.min-saude.pt

* Corresponding author.