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A case of isolated hypoglossal nerve palsy in a 14-year-old girl. Could *Mycoplasma Pneumoniae* be the cause?

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Abstract

Hypoglossal Nerve Palsy (HNP) is an uncommon neurological abnormality that can provoke characteristic clinical signs, including unilateral atrophy of the tongue musculature. We present the case of a healthy 14-year-old girl who presented with acute onset of dysarthria, tongue fasciculations, and right-sided tongue weakness. An extended work out was performed with no evidence of any pathology except positive serology tests for *Mycoplasma pneumoniae* (*M.pneumoniae*). She was treated with oral steroids. Five months after the onset of symptoms, the patient recovered completely. Herein, we report the case of a reversible isolated unilateral hypoglossal nerve palsy in a teenager presumably post-infectious due to *M.pneumonia.*

Keywords

Hypoglossal nerve palsy; *Mycoplasma pneumoniae*; Children.

Introduction

Hypoglossal nerve palsy is a rare clinical condition that can provoke characteristic clinical signs, including hemi-atrophy, fasciculations and deviation of the tongue. It is commonly presented with other cranial nerve palsies. The most common cause is a tumor, predominantly malignant, as described in nearly half of the palsies, thus emphasizing the need to exclude malignancies [1], followed by trauma, stroke, aneurysm or carotid artery dissection, vascular abnormalities and infection. 3% of the cases have been diagnosed as idiopathic. Isolated HNP has only been reported in few cases post influenza vaccination or common cold as well as in infectious mononucleosis [2].

The majority of isolated hypoglossal nerve palsy cases reported in the literature represent adult patients and only a small number of pediatric case reports are published [3-9].

To the best of our knowledge this is the first case report of unilateral hypoglossal nerve palsy in a teenage girl, with high suspicion of being the result of *M.pneumonia* infection, as the evidence points to.

Case Report

A Caucasian 14-year-old girl presented at the emergency department complaining about speech impairment and mild dysphagia for the last 48 hours. She had no recent history of upper respiratory tract infection, surgery or trauma. Three weeks prior she was admitted to a pediatric department, with intense headache and neck pain. An emergency brain CT was performed but no clinical and laboratory cause was detected.

Upon initial physical examination, neurological examination revealed right-sided deviation of her tongue upon protrusion, right hemiatrophy of the tongue and fasciculations, clinical image compatible with isolated right hypoglossal nerve palsy (Figure 1 a,b). Her sense of taste was normal. The rest of neurological examination was unremarkable. Magnetic Resonance Imaging (MRI-MRA scan) of the brain, neck and cervical spine (pre/post contrast) was performed with no evidence of malignancy, vascular malformation, carotid artery dissection or demyelination. Chest X-ray and abdomen ultrasonography were normal. Detailed routine hematological and biochemical blood investigations, (including CBC, ESR, CRP, glucose, electrolytes, renal and hepatic markers) and complete hemostatic screening, were also normal. Mantoux skin test was negative. Viral serology (HSV, CMV, EBV, Adenovirus, Enterovirus), vasculitic profile (rheumatoid factor, ANA, anti-DNA, C3, C4), serum immunoglobulin levels (IgG, IgM, IgA) were in normal range. A lumbar puncture was also performed without any pathological findings (WCC: 2, Glu: 57 mg/dl, Prot: 14 mg/dl), Gram stain, PCR for CMV, HSV, EBV and bacterial culture). She received oral prednisone for 10 days initially with a dose of 1mg/kg for 5 days and continuing with tapering at 0,5 mg/kg for another 5 days, with mild improvement in speech and dysphagia. IgM serum antibodies for *M. pneumoniae* were found positive while IgG were negative. Four weeks later, antibodies for *M. pneumonia* were repeated, revealing both IgM (+) and IgG (+), indicating the possibility of recent Mycoplasma pneumoniae infection. However, no antibiotics were given, since the patient didn't have any symptoms indicating respiratory tract infection. The patient had a frequent follow up every month in the outpatient department. She improved gradually and showed complete resolution within five months (Figure 2 a,b).

Herein we report the case of a reversible isolated unilateral hypoglossal nerve palsy in a 14-year-old girl, presumably secondary (postinfectious) to asymptomatic *M.pneumoniae* infection.

Discussion

Hypoglossal nerve involvement in association with other cranial nerves is common, however isolated unilateral hypoglossal nerve palsy is a rare finding and represents a diagnostic challenge.

In the diagnosis of idiopathic HNP, neuroimaging studies (CT scan and especially MRI and MR angiography) remain mandatory in order to exclude tumoral, traumatic or vascular causes in view of the great need of a specific treatment. In our patient, MRI-MRA scan of the brain, neck and cervical spine (pre/post

contrast) didn't reveal anything abnormal. No other cause was identifiable despite thorough investigation. The presence of positive serology testing for M.pneumoniae and intense headache which led her to hospitalization three weeks prior the unilateral hypoglossal palsy, indicated the possibility of recent Mycoplasma pneumoniae infection. Acute infection with M. pneumoniae was not clearly determined even by raised IgG antibodies 4 weeks later, as our patient did not show any acute illness or respiratory symptoms. A specific etiology could not be established since CSF PCR was not performed.

M. pneumoniae infection in children has repeatedly been associated with the manifestation of different neurological diseases. It is a common respiratory pathogen mostly affecting children and adolescents. It can also lead to extrapulmonary disease with neurological disorders being the most frequent (10). Central nervous system (CNS) manifestations occur in approximately 0.1 percent of all patients with M. pneumoniae infections and approximately 6 percent of hospitalized patients [11]. CNS involvement occurs most frequently in children and includes: meningoencephalitis, ADEM, transverse myelitis, cerebellar ataxia, Guillain-Barré syndrome, cerebellar infarct, peripheral neuropathy and cranial nerve palsies [8,12-14].

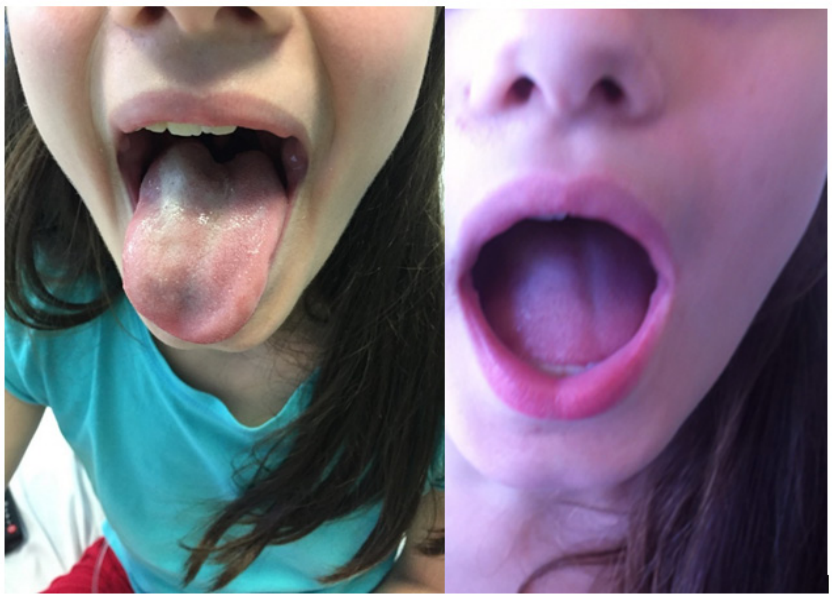

Figure 1: Right-sided deviation of the tongue on protrusion and mild atrophy of the right side of the tongue (Unilateral right hypoglossal nerve palsy).

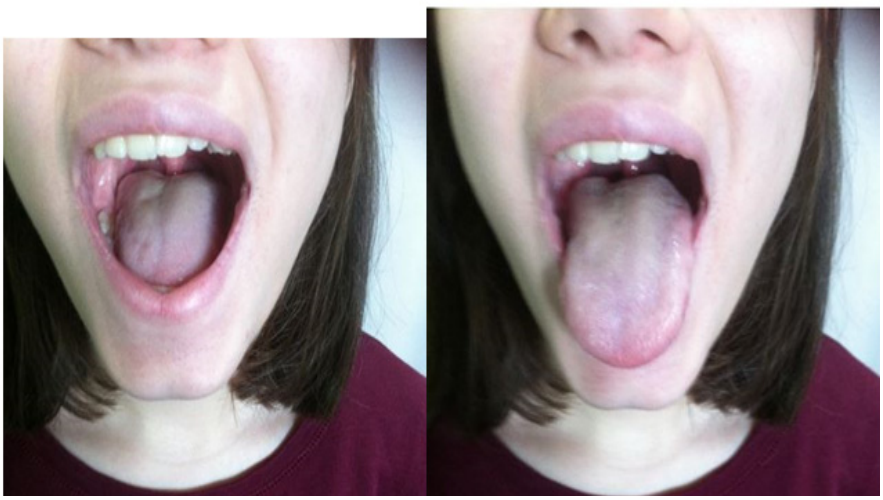

Figure 2: Complete resolution of right hypoglossal nerve palsy five months after the diagnosis.

Kammer et al [15] retrospectively investigated the frequency and types of neurological involvement in 89 patients aged 0 to 18 years hospitalized with acute M. pneumoniae infection as well as their long-term outcome. In 22 of the 89 patients, neurological symptoms and/or signs were documented on admission or during the course of in-patient treatment, with encephalitis being the most frequent (6/22), aseptic meningitis (3/22), longitudinal extensive transverse myelitis (1/22), and vestibular neuritis (1/22) while 11 patients suffered nonspecific neurological symptoms and signs.

In another retrospective study of 365 children admitted to the Hospital for Sick Children over a 16-year period -between 1996 and 2013, with M.pneumoniae detected in the CSF or respiratory tract by PCR, neurologic disease was attributable to M. pneumoniae in 11.5 percent [16]. Among the 42 children with neurologic disease, encephalitis, ADEM, transverse myelitis, and cerebellar ataxia were most common. CSF PCR was positive for M. pneumoniae in 14 of the 35 children in whom it was obtained. Two distinct patterns of neurologic disease were noted: a prodrome of ≥7 days, respiratory manifestations, an immunoglobulin M (IgM) response in peripheral blood, and detection of M. pneumoniae in the respiratory tract, but not the CSF; and a prodrome of <7 days, fewer respiratory manifestations, and IgM response and detection of M. pneumoniae in the CSF only, supporting the hypothesis of two separate pathogenetic mechanisms for M. pneumoniae-associated neurologic disease, one related to direct infection of the central nervous system and one indirect, likely immunologically mediated. The pathogenesis of neurological involvement of M. pneumoniae infection is not well understood. Direct invasion of the CNS, auto immunologic effects, vascular injury, and neurotoxin involvement are discussed.

Although uncommon, CNS involvement is associated with significant morbidity and mortality [11]. In the retrospective study described above, 20 of 42 children with M. pneumoniae neurologic disease had adverse neurologic outcomes (eg, epilepsy, focal neurologic deficits, persistent headaches), but no deaths were reported [16].

In children with neurological symptoms, direct detection of the pathogen in the CNS by Polymerase Chain Reaction (PCR) is often not successful. In CSF, normal glucose, lymphocytic pleocytosis and elevated protein-not detected in our patient- is often present [11].

Our patient was treated with oral steroids as idiopathic, monosymptomatic isolated unilateral hypoglossal nerve palsy, presumably secondary to M. pneumoniae infection, an entity similar to Bell's palsy. Other examples of 'Bell's palsy of the tongue' are probably viral in origin [17]. Infections such as enterovirus, adenovirus, Epstein-Barr virus, herpes simplex virus, influenza virus and Streptococcus spp [2,3,5,6]. Infectious mononucleosis as a cause of transient and even permanent isolated hypoglossal nerve palsy has been previously reported [17].

Antibiotics were not administered as there was no pulmonary disease. However, it is not clear whether antibiotics are effective for neurological disease because of their limited penetration of the blood–brain barrier and, most importantly, the unresolved pathogenic mechanism of neurological disease.

In this report we demonstrate the case of a reversible isolated unilateral hypoglossal nerve palsy in a teenager presumably post-infectious due to M. pneumonia. We emphasize the need of an extensive diagnostic workup of potentially treatable etiologies. Neuroimaging should be part of the investigation in

order to exclude the possibility of malignancy. However, in our case we aim to demonstrate the value of recognizing self-limited, post-infectious cases of hypoglossal nerve palsies in children and to emphasize the necessity to ascertain viral serology, as well as antibodies for other infectious causes such as M. pneumonia.

Statement of Ethics

This case report was produced in accordance with institutional policies; the patient's parents gave their consent for publication of this case report and any accompanying images.

The paper is exempt from ethical committee approval, since this is a retrospective presentation of the case report

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