

Unilateral Hypoglossal Nerve Palsy: As the Only Presentation of Tuberculosis

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Abstract- Tuberculous spondylitis is not an uncommon disease of the spine. Near one percent of all cases of spinal tuberculosis (TB) involves craniocervical junction. Hypoglossal nerve palsy is not an uncommon neurological finding, but isolated involvement of the hypoglossal nerve is rare and limited to case reports or small case series. Here, we report a case of craniocervical junction tuberculosis presenting with unilateral hypoglossal nerve palsy. Case is a 41-year-old woman with neck and suboccipital pain since one month and unilateral right hypoglossal nerve palsy since one week. All laboratory tests were unremarkable except raised ESR level. Involvement of C1-C2 and hypoglossal canal were demonstrated by CT scan of craniocervical junction. Tissue diagnosis of TB was established by open biopsy of the craniocervical junction.

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Introduction

Craniocervical junction tuberculosis (TB) is rare and accounts for about 1% of all cases of spinal TB (1,2). TB of the craniocervical junction (CCJ) is very challenging in diagnosis and treatment. The atlas and axis involvement is more common than occipital bone. Complicating involvement of this region could be very dangerous and fatal due to cord compression secondary to atlantoaxial dislocation (2).

Hypoglossal nerve palsy is not an uncommon neurological finding, but isolated involvement of the hypoglossal nerve is rare and limited to case reports or small case series (3).

TB is an endemic disease in our geographical area and it has diverse systemic manifestations. This case report has some interesting epidemiological, neurological, and neurosurgical aspects of the nervous system presentation of TB.

In this article we report a patient with TB of the CCJ who presented with unilateral hypoglossal nerve palsy as the only manifestation of the disease.

Case Report

History and physical examination

A 41-year-old woman, who was healthy until 40 days before admission, presented with neck and suboccipital pain and stiffness. She had no complaint of fever or

malaise. There was no history of trauma. The pain was not radiating and it responded well to heat and NSAIDs. Before admission, she had two visits by general practitioners with normal lateral and AP cervical X-rays. She was treated by muscle relaxants and NSAIDs. Ten days before admission, she had normal neurological examination which was done by a neurologist, normal complete blood count (CBC), but an elevated erythrocyte sedimentation rate (ESR).

A day before admission she noted speech difficulty and tongue deviation to right on protrusion (unilateral right hypoglossal nerve palsy) (Figure 1).

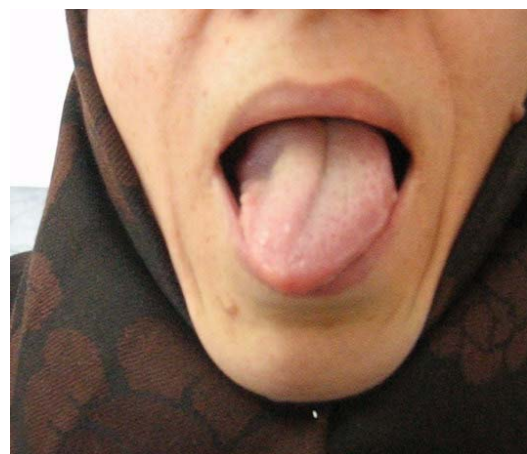


Figure 1. Deviation of the tongue to the right on protrusion and mild atrophy of the right side of the tongue (Unilateral right hypoglossal nerve palsy).

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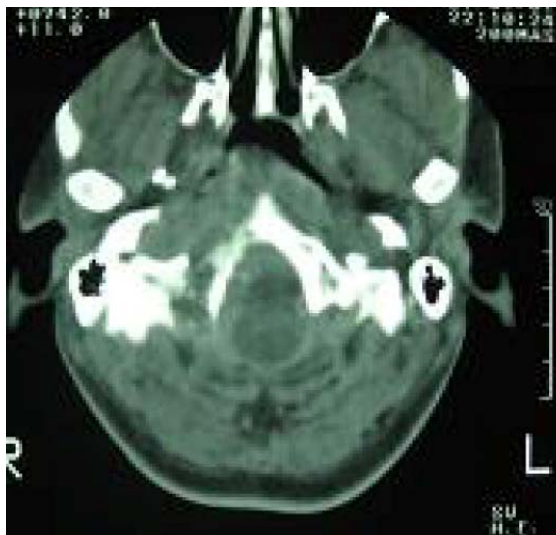


Figure 2. Lytic bone lesion in right basiocciput with soft tissue swelling bulged into the foramen magnum and nasopharynx. Note the small bone fragment near the hypoglossal canal.

On admission she had normal vital signs and was not ill or toxic. She had normal general examination except tenderness at right occipital condyle posterior to right mastoid process. Right ear otoscopy was normal. She had deviation of the tongue to the right on protrusion (right hypoglossal nerve palsy), other neurological examination were normal.

Laboratory and imaging studies

All laboratory tests including: CBC with differentiation, biochemistry, liver function tests, HIV, HBV, HCV tests, vasculitis survey, Wright test, 2ME, Coombs Wright, and VDRL (RPR) were normal. Also

chest X-ray and cervical spine X-ray were normal.

The only laboratory abnormalities were: PPD test = 8 mm and ESR = 73 mm/hr.

Cerebrospinal fluid analysis was normal. Routine smear and culture and smear and culture for TB and fungi in cerebrospinal fluid (CSF) had negative results. PCR (polymerase chain reaction) for TB in CSF was also negative. Abdominal and pelvic sonographies were normal.

Computed tomography (CT) with and without contrast of upper cervical spine and base of the skull showed: destruction of C1-C2 and condyle of the occiput in the right side. (Figures 2 and 3).



Figure 3. Axial CT Scan at the level of foramen Magnum, bone window: Lytic destructive lesion of the right occipital condyle with sequestered bone fragment adjacent to the right hypoglossal canal.

Diagnosis

Finally neurosurgical consultation was done for open biopsy and tissue diagnosis. Sample was sent for tissue culture and pathologic examination. In microscopic examination the sample had granuloma formation, and caseation necrosis suggestive of TB and acid fast bacilli were seen in the biopsy sample staining.

Treatment

After receiving tissue diagnosis, 4-drug antimycobacterial regimen was begun. Gradually occipital pain and stiffness subsided and right hypoglossal nerve palsy resolved.

At the moment (after 18 months of follow up) the patient is asymptomatic with totally normal systemic and neurological examination.

Discussion

The hypoglossal nerve is a pure motor nerve innervating intrinsic as well as extrinsic tongue muscles. The hypoglossal nerve palsy (HNP) accompanied with other neurological abnormalities such as other cranial nerve palsies or long tracts signs is not an uncommon finding (4). Keane in 1996 reported 100 cases of HNP with other neurological abnormalities, in which, malignant tumors were the most common cause (5).

However, isolated hypoglossal nerve palsy is a rare finding and in our literature review there were only a small number of case series and several case reports of it. Although there is a general agreement that the presence of hypoglossal nerve palsy usually is a sign of underlying malignancies (5,6) but there are some reports of isolated and reversible form of hypoglossal nerve palsy usually caused by post-infectious process (6).

Other etiologies for isolated hypoglossal nerve palsy are: Chiari malformation, radiotherapy, post-vaccination neuropathy, anomalous vertebral artery, metastasis, post-infectious acute disseminated encephalomyelitis, and idiopathic causes (4,6).

Craniovertebral junction TB is described as a rare condition accounting for about 0.5% of TB in general, and 6% of extra pulmonary TB (7,8). Moreover TB is still prevalent in developing countries and it is going to be seen more than before due to increasing number of immunodeficient patients (8).

In a case series reported by Bhojraj, the most common symptoms in TB of CCJ were occipital and neck pain and most of patients had constitutional

symptoms such as fever and loss of weight and appetite (2).

In other case series described by Shukla *et al.*, after neck pain as the most common symptom, spastic quadriparesis (67%), sensory deficit (54%), sphincter disturbances (25%), neck tilt (4%), and dysphagia (4%) were seen (9).

According to our literature review, we found only two report of isolated hypoglossal nerve palsy as a presentation of TB, the older one by Richards *et al.* in 1989 (10) and the recent one by Chakraborty in 2009 (11). In both of these reports involvement of CCJ by TB caused unilateral hypoglossal nerve palsy (10,11).

Our case report is one of the rarest reports of craniocervical junction tuberculosis, in which a woman with high socioeconomic level without any systemic symptoms of TB or any other site of involvement by TB, presented with neck pain and hypoglossal nerve palsy as the only neurological manifestation. The patient was treated only by medical treatment and at the moment of this report, the outcome is excellent (after 18 months of follow up). In conclusion, TB which is endemic in our country and geographical region must be considered as a differential diagnosis of neck and spinal pain. It must be remembered that TB of CCJ could be presented without any systemic symptoms. The other interesting point is that elevated ESR is a very important diagnostic test in the evaluation of patients presenting with neck and spinal pain.

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