

Isolated Hypoglossal Nerve Palsy Associated with Tuberculosis of the Atlantoaxial Joint: A Rare Case Report

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Abstract

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Venous thromboembolism (VTE) is the third most common cardiovascular disorder worldwide. While lower-extremity deep vein thrombosis and pulmonary embolism are typical presentations, thrombosis in atypical sites remains challenging to recognize and diagnose.

Case Description:

A 27-year-old female flight attendant with history inflammatory bowel disease (IBD), tuberculosis and combined oral contraceptive (COC) use presented with an acute abdominal pain. Initial contrast-enhanced CT revealed thrombosis of the left renal and ovarian veins. Laboratory tests showed elevated hs-CRP and D-dimer, with positive ANA but negative antiphospholipid antibodies. She was treated with intravenous heparin followed by oral rivaroxaban, though adherence was inconsistent due to episodes of heavy vaginal bleeding. Repeat CT imaging eight months later demonstrated resolution of the initial thrombi but revealed a new thrombus in the inferior vena cava extending into the right common iliac vein, accompanied by recurrent elevation of D-dimer.

This case illustrates the interplay of multiple risk factors for VTE, including IBD, prolonged immobility during long-haul travel, COC use, tuberculosis, and rifampicin therapy. These overlapping chronic and transient triggers likely contributed to recurrent thrombosis despite ongoing treatment.

Conclusions:

Recurrent VTE in unusual venous sites can occur particularly in patients with multiple risk factors. Early recognition, appropriate imaging, and anticoagulant therapy adherence are essential to preventing progression and recurrence.

Background:

Introduction

Osteoarticular tuberculosis (TB) is an uncommon form of extrapulmonary TB (EPTB) accounting for 1-6% of all TB cases and 10-15% of EPTB cases. It occurs through the hematogenous or lymphatic spread of *Mycobacterium tuberculosis* (MTB) from other parts of the body. While it is most frequently observed in the spine and hip, involvement of the cervical vertebrae is a rare manifestation that has

been reported and may result in severe neurological deficits. Due to its non-specific presentation, it is often overlooked and may cause delayed diagnosis and treatment.^{1,2}

Hypoglossal nerve palsy (HNP) is not uncommon. However, it is rare for it to present as a sole manifestation, otherwise known as isolated HNP. Its occurrence is commonly on account of an underlying cause, with intracranial and skull base neoplasms along with vertebral trauma

being responsible for up to 50% of cases.^{3,4,5} To our knowledge, it has never been reported as a manifestation of osteoarticular TB. In this report, we present a rare case of isolated HNP associated with osteoarticular TB involving the atlantoaxial joint in a 30-year-old female.

Case Description

A 30-year-old female presented with a 1-month history of neck pain. The pain was described as dull and aching, with radiation to the occipital region within the past 2 weeks. She claimed that positional changes of her head exacerbated the pain, with alleviation during rest and on supine position. It began without any preceding trauma or significant stressor. She also complained of tongue deviation of sudden onset which appeared a few days after her neck pain, which caused speech and chewing difficulties. Upon further history taking, there was unintentional weight loss. She denied other symptoms such as fever, seizures, or extremity weakness. Past medical history was unremarkable. She had never experienced these symptoms before.

Her vitals were within normal limits. On physical examination, there were bilateral enlargement of the axillary lymph nodes. Upon neurological examination, left-sided tongue deviation with fasciculation was observed. Examination of the other cranial nerves, motor and sensory functions were unremarkable. Blood work up was significant for microcytic hypochromic anemia (Hb 11.4 g/dL) and a slightly elevated ESR (26 mm/hr). Serum electrolytes, liver enzymes, and renal function tests were normal. Chest x-ray (CXR) did not show any pathological findings. Plain and contrast-enhanced head magnetic resonance imaging (MRI) revealed thickening and enhancement of the left soft tissue adjacent to the atlantoaxial joint, pathological enhancement of the left clivus, and multiple mild enlargement of the left cervical lymph nodes. Bone biopsy and MTB culture (MTBC) were not performed.

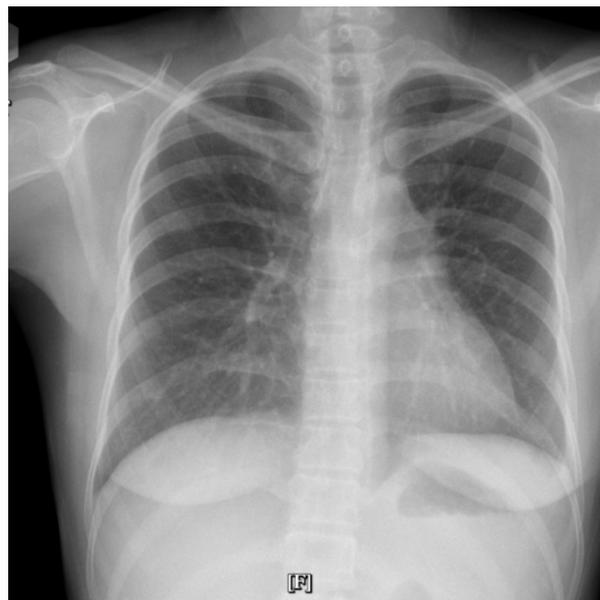


Figure 1 CXR revealing no abnormalities.

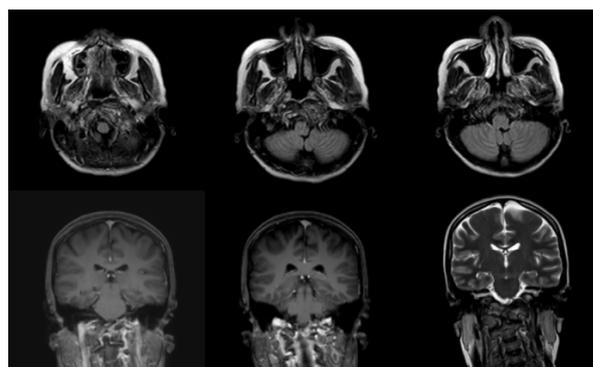


Figure 2 Contrast enhanced brain MRI revealing thickening and enhancement of the left soft tissue adjacent to the atlantoaxial joint, and pathological enhancement of the left clivus

She was diagnosed with tuberculous lymphadenitis and isolated HNP associated with osteoarticular TB of the atlantoaxial joint. She was initiated on a regimen of anti-tuberculosis treatment (ATT), comprising of rifampicin 600 mg, isoniazid 300 mg, pyrazinamide 1.600 mg, and ethambutol 1.100 mg. She was also treated with Dexamethasone 4 mg TID, tapered over a 3-week period, and Mecobalamine 500 mcg BD. Her symptoms improved within one month on ATT and continued to progress favorably. At the 6-month follow-up, her neck pain had nearly resolved and only appeared with head rotation. Tongue deviation was no longer present; however,

tongue fasciculation persisted, albeit with significant improvement.

Repeat brain MRI was conducted one year later, which demonstrated a reduction of soft tissue inflammation adjacent to the clivus and atlantoaxial joint, a decrease in the size of lymph nodes, but more prominent vertebral body destruction of the clivus and left vertebral body of C1. The patient is currently continuing the ATT regimen until the 18th month.

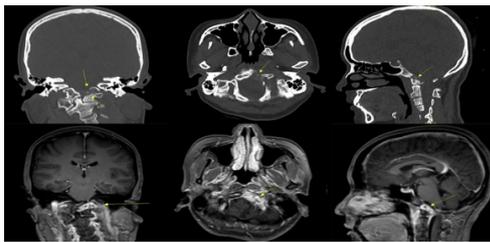


Fig 3. Contrast enhanced brain MRI bone window (row 1) and T1W (row 2) revealing reduction of soft tissue inflammation, destruction of the clivus and left C1 vertebral body upon 1 year follow-up

Discussion

Our patient presented with neck pain associated with unintentional weight loss and lymphadenopathy. Given the high prevalence of TB in developing countries, it should be considered as a differential diagnosis in patients presenting with such manifestations. Cervical osteoarticular involvement of TB typically presents with neck pain and stiffness, weakness in all four extremities, and often systemic symptoms such as fever, night sweats, and weight loss.^{2,6} The absence of extremity weakness, coupled with an isolated HNP, rendered our patient's symptom of neck pain non-specific and a challenge to diagnose. Definitive diagnosis of osteoarticular TB is through MTB culture or PCR of a sample obtained through biopsy.⁷ However, due to the invasive nature of an open biopsy, it was not conducted in our patient. Despite the absence of cultures and serological tests, the patient's clinical manifestations and brain MRI findings strongly suggest a TB infection with involvement of the atlantoaxial joint and clivus.

In this patient, the only neurological deficit observed was an isolated left-sided HNP, which is a rare cranial neuropathy frequently associated with an underlying condition. Due to its anatomical location and pathway, HNP normally present concomitantly with deficits of other cranial nerves, especially the glossopharyngeal (CN IX), vagus (CN X) and spinal nerves (CN XI).⁸ An isolated HNP most commonly suggests a pathology along the peripheral pathway of the hypoglossal nerve rather than central.⁹ Therefore, clinicians must have a comprehensive understanding of the hypoglossal nerve's pathway and its interactions with adjacent structures to accurately determine the location of the pathology, identify the underlying etiology, and make a precise diagnosis.

The hypoglossal nerve, also known as the twelfth cranial nerve (CN XII), innervates the tongue musculature with its nucleus arising from the lower third of the medulla. Its main afferent input is from the contralateral cerebral hemisphere. From the hypoglossal nuclei, its somatic efferent fibers descend and emerge from the brainstem, exit the skull through the hypoglossal canal, and travel along the lower cervical region.¹⁰ The initial portion of the hypoglossal nerve pathway runs in close proximity to the vagus nerve, internal carotid artery, and internal jugular vein.¹¹

Despite the lack of information regarding isolated HNP in literature, several etiologies have been associated with this phenomenon. Intracranial and skull base tumors, along with vertebral trauma, have been identified as the most common causes, responsible for approximately 50% of all cases. Other reported etiologies include trauma, infections, vascular pathologies, iatrogenic, and demyelinating disease (Multiple Sclerosis).^{12,13} On rare occasions, isolated HNPs have been reported to resolve spontaneously without an identified underlying cause, despite extensive investigation.³ Additionally, recent reports have noted isolated HNP as the only symptom of cervical artery dissection.^{4,5} Combarros *et al.* reported nine patients with isolated HNP; four were idiopathic, three were caused by neoplasm of the skull base, and two were exceptional

cases of Chiari malformation and dural arteriovenous fistula of the transverse sinus.¹⁴

In our case, the underlying causes are mechanical and infection. Due to involvement of the atlantoaxial joint by osteoarticular TB, direct compression of the left hypoglossal nerve by inflamed soft tissue led to an isolated HNP. Brain MRI was pivotal in excluding neoplastic processes and determining the exact underlying cause. Other etiologies, including trauma, were ruled out based on clinical grounds. An MR or CT angiography may be conducted in cases of suspected vascular etiology.¹⁵ To the best of our knowledge, there are no cases to date reporting a similar etiology for isolated HNP. The progression of her symptoms after initiation of ATT further supports the underlying cause of her isolated HNP, with complete recovery of tongue deviation after 6 months on ATT. Upon her one year follow-up, she did not present with any neurological deficits and continued to show clinical improvement.

Conclusion

Isolated HNP is a rarely encountered phenomenon and most frequently a consequence of an underlying pathology. Although it is commonly associated with neoplastic processes or trauma, we postulate that in this case it was induced by direct compression of the hypoglossal nerve from cervical osteoarticular TB. Therefore, it is significant that clinicians possess a comprehensive knowledge regarding the possible etiologies of isolated HNP as well as the pathway of the hypoglossal nerve.

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