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CASE REPORT

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Isolated Hypoglossal Nerve Palsy Secondary to Basilar Artery Dolichoectasia

Baziler Arter Dolikoektazisine Bağlı İzole Hipoglossal Sinir Felci

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ABSTRACT

Hypoglossal nerve palsy is commonly associated with metastatic tumors, trauma, and vascular anomalies, with vertebrobasilar artery dolichoectasia being an exceptionally rare etiology. This case report describes a 57-year-old male presenting with isolated hypoglossal nerve palsy attributed to basilar artery dolichoectasia, confirmed through digital subtraction angiography, which revealed significant vascular dilation and tortuosity. The patient was treated with a regimen of antihypertensive and antiplatelet agents, resulting in substantial clinical improvement over a six-month follow-up period. This report highlights the clinical significance of vertebrobasilar artery dolichoectasia as a rare but critical factor in isolated cranial nerve palsy, addressing the diagnostic complexities and therapeutic considerations involved. The case underscores the importance of advanced neuroimaging in identifying subtle vascular anomalies and demonstrates the benefit of a multidisciplinary approach in managing such rare presentations.

Keywords: Hypoglossal nerve, paralysis, basilar artery, dolichoectasia.

ÖZ

Hipoglossal sinir felci genellikle metastatik tümörler, travma ve vasküler anomalilerle ilişkilidir; baziler arter dolikoektazisi ise bu durumun son derece nadir bir nedeni olarak karşımıza çıkmaktadır. Bu olgu sunumunda, dijital substraksiyon anjiyografi ile doğrulanan belirgin vasküler dilatasyon ve tortiozite gösteren baziler arter dolikoektazisine bağlı olarak izole hipoglossal sinir felci gelişen 57 yaşındaki bir erkek hasta sunulmaktadır. Hasta, antihipertansif ve antiagregan ajanlarla tedavi edilmiş ve altı aylık takip sürecinde belirgin bir klinik iyileşme gözlenmiştir. Bu olgu sunumu, baziler arter dolikoektazisinin nadir ancak klinik açıdan kritik bir etiyolojik faktör olarak önemini vurgulamakta, tanısal zorluklar ve tedavi yaklaşımlarını ele almaktadır. Aynı zamanda vasküler anomalileri saptamada ileri nörogörüntüleme yöntemlerinin önemini ve bu tür nadir olguların yönetiminde multidisipliner bir yaklaşımın faydasını göstermektedir.

Anahtar Kelimeler: Hipoglossal sinir, paralizi, baziler arter, dolikoektazi.

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CASE PRESENTATION

Hypoglossal nerve palsy is most frequently attributed to metastatic tumors (49%), trauma (12%) and stroke (6%). Other etiologies include idiopathic factors, demyelinating diseases, motor neuron disorders, arterial dissections, aneurysms, vertebrobasilar artery ectasia/dolichoectasia (VBD), neoplasms, metastases (commonly from renal, prostate, and breast cancers), jugular vein thrombosis, botulism, iatrogenic factors, radiotherapy, and infections.[1] Among these, VBD represents a particularly rare etiology. Dolichoectasia is a dilative arteriopathy characterized by the elongation, dilation, and tortuosity of arteries, predominantly affecting the intracranial vertebral and basilar arteries. Although the pathophysiological mechanisms remain incompletely understood, one hypothesis suggests that VBD arises from early fragmentation and degeneration of the internal elastic lamina, leading to the progressive loss of elastic tissue in the intima-media layer and subsequent smooth muscle atrophy. An alternative theory implicates atherosclerotic changes, especially in hypertensive patients, as a contributing factor to this vascular anomaly.[2]

The diagnosis of VBD requires the observation of vertebrobasilar arteries dilated beyond 4.5 mm in diameter, with elongation and displacement of adjacent structures, such as the suprasellar cistern. VBD prevalence in the general population ranges from 0.06% to 5.8%, with most cases remaining asymptomatic and incidentally discovered during imaging performed for unrelated reasons, particularly in hypertensive individuals over 40 years old. Symptomatic cases may present with clinical manifestations due to cerebral ischemia, hemorrhage or compressive effects on surrounding structures, including the brainstem, third ventricle or cranial nerve roots.[3]

A review of the literature reveals a limited number of cases documenting isolated hypoglossal nerve palsy as a result of basilar artery dolichoectasia, underscoring the rarity of this condition. Herein, we present a case of isolated unilateral hypoglossal nerve palsy secondary to VBD, aiming to contribute to the scarce body of knowledge regarding this unusual presentation. Given the paucity of reports, this case emphasizes the importance of considering VBD in the differential diagnosis of hypoglossal nerve palsy, particularly in patients with concurrent vascular risk factors.

MAIN POINTS

- Spinal dural AVF is one of the diagnoses that both clinicians and neuroradiologists should consider in patients with progressive paraparesis who have long-segment hyperintense lesions on spinal MR imaging.
- Spinal angiography is the gold standard for diagnosis.
- Although treatment options vary from patient to patient, the most effective methods are microsurgical occlusion and endovascular embolization.

CASE REPORT

A 57-year-old male presented to the emergency department with complaints of numbness on the right side of his face, tongue deviation and enlargement, slurred speech, headache and elevated blood pressure (174/100 mmHg), all of which had developed approximately 12 hours prior to admission. The patient had a documented history of poorly controlled hypertension, with irregular adherence to antihypertensive medication and a significant smoking history totaling 30 pack-years.

Neurological examination revealed deviation of the tongue to the right while inside the mouth (Figure 1A) and to the left when protruded (Figure 1B), along with restricted tongue movements and dysarthria. The gag reflex was intact and examinations of other cranial nerves were unremarkable. Motor, sensory and cerebellar system assessments were within normal limits, with normoactive deep tendon reflexes. Initial imaging, including diffusion-weighted magnetic resonance imaging (MRI) and non-contrast cranial computed tomography (CT), showed no abnormalities. The patient was admitted to the neurology department for comprehensive evaluation and further investigation of the underlying etiology.

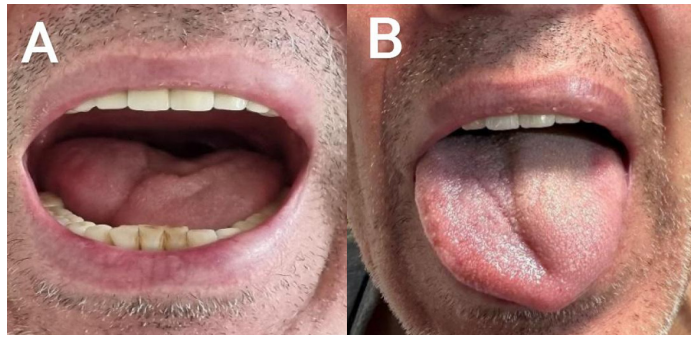

Figure 1. (A) Tongue deviation to the right observed when the tongue is at rest within the oral cavity. (B) Pronounced tongue deviation to the left upon protrusion.

Detailed anamnesis ruled out recent trauma or surgical intervention that could account for hypoglossal nerve damage. Notably, the patient reported consistently elevated blood pressure readings, generally exceeding 140/90 mmHg. To exclude potential differential diagnoses such as demyelinating diseases, space-occupying lesions or infectious etiologies, a series of contrast-enhanced MRIs encompassing the cranial, cervical, nasopharyngeal and soft tissue regions was performed, yielding no pathological findings. Electromyography of the left hypoglossal nerve confirmed acute peripheral neurogenic involvement, consistent with the clinical presentation.

Comprehensive laboratory investigations, including hematological, biochemical, infectious serological tests and tumor markers, returned unremarkable results. Rheumatologic evaluation revealed no evidence of vasculitis, and positron emission tomography imaging was similarly devoid of abnormal findings. Subsequent carotid CT angiography (CTA) of the head and neck, pursued to evaluate for vascular abnormalities, identified dissections of the bilateral internal carotid arteries and basilar artery dolichoectasia (Figure 2A and B).

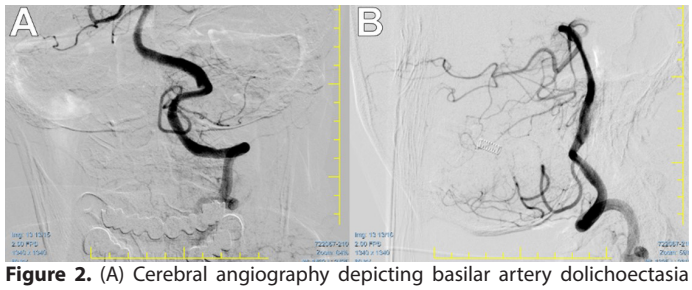

Figure 2. (A) Cerebral angiography depicting basilar artery dolichoectasia with a diameter of 8.20 mm in anterior (B) and lateral view.

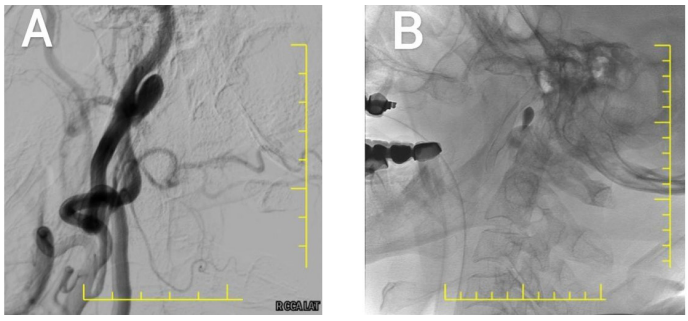

Figure 3. (A) A dissecting aneurysmal dilation of 15.9 x 8.1 mm is identified in the cervical segment of the right ICA, exhibiting a tortuous configuration. (B) The dissecting aneurysm stagnation after flow diverter stent placement.

Based on the findings, a multidisciplinary consultation was conducted with the neurosurgery and cerebrovascular teams. It was collectively determined that microvascular decompression was not indicated at this stage. Consequently, a decision was made to proceed with conservative medical management, including antihypertensive and antiplatelet therapy, to address both blood pressure control and secondary prevention. The patient was closely monitored for clinical improvement or progression.

DISCUSSION

Isolated hypoglossal nerve palsy is an uncommon clinical entity, often indicative of underlying pathological processes.[4] The hypoglossal nerve (cranial nerve XII) is a motor nerve responsible for innervating the intrinsic muscles of the tongue, including the genioglossus, hypoglossus and styloglossus muscles, thus facilitating essential functions such as tongue movement, speech and swallowing.[5]

The hypoglossal nerve originates from the medulla oblongata, with fibers exiting the skull through the hypoglossal canal. After exiting, it traverses the parapharyngeal space within the nasopharyngeal carotid sheath, where it is closely associated with the 9th, 10th and 11th cranial nerves, the internal carotid artery and the internal jugular vein. As it descends, it courses between the internal carotid artery and the internal jugular vein before turning towards the floor of the mouth near the angle of the mandible, becoming superficial beneath the posterior belly of the digastric muscle. The nerve then advances along the upper portion of the greater horn of the hyoid bone, crosses the lingual artery and continues to the sublingual region.

Throughout its extensive pathway from the medulla oblongata to the floor of the mouth, the hypoglossal nerve is susceptible to damage at multiple levels. This vulnerability often reflects a serious underlying pathology, necessitating meticulous clinical history-taking and comprehensive evaluation to facilitate accurate diagnosis.[6] The diagnosis, management and follow-up of such conditions typically require a multidisciplinary approach and advanced diagnostic modalities to explore potential skull base lesions thoroughly.

In this case, basilar artery dolichoectasia was identified as the primary etiological factor. VBD is characterized by the abnormal dilation, elongation and tortuosity of the basilar artery, which can result in various clinical manifestations due to compression of adjacent neural structures or compromised blood flow. Diagnostic criteria for VBD are based on specific morphometric measurements and anatomical displacement assessments. A basilar artery is considered ectasia when its diameter exceeds 4.5 mm. Furthermore, dolichoectasia is typically diagnosed when the artery exhibits elongation beyond the boundaries of the clivus or dorsum sella and when the basilar bifurcation is positioned above the suprasellar cistern.[7] These anatomical changes can exert significant mass effects on the surrounding neural structures, potentially leading to diverse and severe neurological symptoms. Symptomatic VBD can present with a spectrum of clinical findings, ranging from headaches and ischemic strokes to hemorrhagic events and compressive neuropathies, including cranial nerve involvement. In more severe cases, hydrocephalus may occur due to obstruction of cerebrospinal fluid pathways. The variability in presentation underscores the need for careful and systematic evaluation, particularly in cases with atypical cranial nerve deficits.

Isolated hypoglossal nerve palsy due to basilar artery dolichoectasia is particularly rare, as it typically arises when the hypoglossal nerve is compressed along its course through the hypoglossal canal by the ectatic artery.[8] Thus, in cases of isolated hypoglossal nerve palsy, detailed neuroimaging of the vertebrobasilar system is essential to exclude vascular abnormalities. Imaging modalities that can be utilized include sonography, MR angiography (MRA), CTA and DSA.[9]

To date, no standardized treatment protocol exists for dolichoectasia due to limited case studies. However, it is suggested that blood pressure management and ischemia-preventing therapies may benefit the prognosis, provided that the risk of hemorrhage is carefully assessed.[10]

Previous studies indicate that vertebrobasilar involvement in dolichoectasia correlates with increased neurological morbidity, although mortality is generally influenced by classic vascular risk factors rather than the specific anatomical characteristics of the vessels.[8]

In this case, the patient's hypoglossal nerve palsy was attributed to basilar artery dolichoectasia, with a diameter of 8.2 mm and a tortuous course. Although bilateral internal carotid artery dissections were also noted, they were not considered contributory to the presenting symptoms. A variety of pathophysiological mechanisms may be involved in the development of VBD. Histological studies support that damage of the internal elastic lamina and thinning of the media related to smooth muscle atrophy is at the basis of this pathology as well as prolonged systemic hypertension. Therefore dissecting aneurysms may also be present in both anterior and posterior system vessels in VBD. In our case very slowly progressing dissection without significant flow disturbances was considered since the aneurysms were asymptomatic. Uncontrolled hypertension likely contributed to endothelial dysfunction, facilitating the development of dolichoectasia and

subsequent compression of the hypoglossal nerve. After a thorough multidisciplinary consultation involving the neurosurgery team, it was determined that microvascular decompression was not appropriate at this stage. Consequently, conservative medical management was chosen, consisting of antihypertensive (telmisartan/hydrochlorothiazide 80/12.5 mg daily and lercanidipine 20 mg daily) and antiplatelet therapy (acetylsalicylic acid 100 mg daily and ticagrelor 90 mg twice daily). At the six-month follow-up, notable improvements were observed in dysarthria and tongue deviation (Figure 4A and B), with the patient reporting enhanced ease in swallowing.

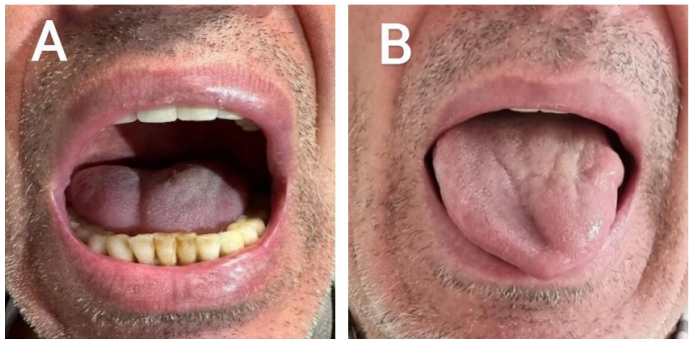

Figure 4. (A) Six-month follow-up shows reduced tongue deviation within the oral cavity (B) and during protrusion suggesting improved hypoglossal nerve function.

In conclusion, this case highlights the clinical significance of vertebrobasilar dolichoectasia as a rare yet critical differential diagnosis in patients presenting with isolated hypoglossal nerve palsy, especially in those with uncontrolled hypertension. The anatomical proximity of the hypoglossal nerve canal to the basilar artery necessitates a high degree of clinical suspicion and comprehensive evaluation, as compression-related neuropathies may arise from seemingly incidental vascular abnormalities. This case underscores the imperative for clinicians to thoroughly consider VBD in similar presentations, as the condition's insidious nature can obscure diagnosis. Meticulous clinical history-taking, a profound understanding of cranial nerve anatomy, and the strategic use of advanced neuroimaging modalities such as MRA, CTA and DSA are indispensable in detecting subtle vascular anomalies. An interdisciplinary approach, involving neurology, neurosurgery and vascular specialists, may further enhance diagnostic accuracy and facilitate the selection of appropriate therapeutic strategies, ultimately improving patient outcomes in these complex and rare case.

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