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Unique Etiology of Trigeminal Neuralgia After Acute Ischemic Stroke

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INTRODUCTION

Trigeminal neuralgia (TGN) is a common neuropathic pain syndrome with several primary and secondary causes. Classical TGN (CTGN) and Symptomatic TGN (STGN) both have been described in the literature, but not as coexistent causes. CTGN occurs due to blood vessel compression of the trigeminal nerve, and magnetic resonance imaging (MRI) or surgical visualization of blood vessel compression and nerve atrophy are needed for confirmation.[1] STGN follows the same diagnostic criteria, but has a radiographic cause other than blood vessel compression.[2] Our case suggested contributions from both, including a unique etiology for the development of CTGN via arteriogenesis after acute ischemic stroke that may require surgical intervention. Written, informed consent was obtained from the patient for publication of this case report.

CASE REPORT

A 68-year-old male presented with complaints of dizziness and right upper extremity (RUE) weakness. Exam revealed RUE ataxia, nystagmus, and dysarthria. Aortic arch and 4-vessel cerebral angiogram revealed critical right vertebral artery (VA) and posterior inferior cerebellar artery (PICA) stenoses with right VA dissection and thrombus causing a suspected right lateral medullary infarct and Wallenberg syndrome, which typically consists of contralateral upper extremity hypoesthesia to pain and temperature, hoarseness, dysphagia, nystagmus, vertigo, and cerebellar symptoms. It can cause loss of ipsilateral facial pain and temperature sensation.[3]

In our patient, stroke symptoms evolved with development of aphonia, singultus, and dysphagia, and onset of ipsilateral facial pain with hypoesthesia to temperature four days after admission. MRI showed interval conspicuity of dorsolateral medullary infarct. All symptoms improved prior to discharge.

Two weeks post-stroke, the patient began to have right sided "tooth" pain that was treated over the next several months with antibiotics, dental work, and pregabalin without relief. One year after his stroke, he was evaluated by neurosurgery for unrelenting, excruciating right facial pain primarily in the trigeminal nerve (CN-V) distribution, specifically the CN-V1/V2 distribution. Suspecting CTGN, an MRI was obtained showing a vascular loop contacting the right trigeminal nerve. Subsequently, a right retromastoid craniotomy with microvascular decompression was performed.

The operative report noted lateral petrosal veins putting pressure on the nerve and an artery contacting the nerve root entry zone. After surgery, symptoms of TGN resolved for less than two months before the pain returned. Another MRI revealed adhesion formation around the trigeminal nerve. He underwent a second retromastoid craniotomy for adhesiolysis which resulted in near complete resolution of the pain for several months. Follow-up nearly three years after revealed recurrence of moderate trigeminal nerve pain in the CN-V2/V3 distribution, controlled with acupuncture and other noninvasive modalities.

DISCUSSION

Trigeminal neuralgia (classical or symptomatic) is a common neuropathic pain syndrome affecting 10,000-15,000 new patients every year in the U.S.[4] CTGN occurs due to blood vessel compression. The most common cause of STGN is multiple sclerosis, but tumors, arteriovenous, and skull base malformations also may play a role.[1] A rarely reported cause of STGN is brainstem infarction, specifically lateral medullary infarction, which results in damage to the spinothalamic tract, nucleus ambiguous, trigeminal tract, vestibular nucleus and/or the inferior cerebellar peduncle, causing Wallenberg syndrome.[2] There are reports of TGN-like pain after dorsolateral medullary stroke that occurs in patients who initially had a loss of facial pain sensation.[5-8]

Ischemic stroke often leads to the development of collateral circulation via arteriogenesis induced by shear stress and growth factors released in an ischemic environment. It can take days to weeks for the collateral vessel to reach its final diameter, which often is associated with an increase in tortuosity and length.[9] Arteriogenesis may be an etiology for CTGN after brainstem ischemic stroke if such vessel engorgement causes trigeminal nerve compression. TGN caused by neurovascular compression likely is due to pulsations causing microtrauma to the nerve, in turn leading to demyelination and remyelination that affects action potential transmission. The most vulnerable area of the nerve to this type of trauma is the nerve root entry zone.[10] In our case, the surgeon specifically mentioned the vessel contacting the nerve root entry zone in the operative note.

Our case was unique in that it supported potentially coexistent causes of TGN. Since this patient had no symptoms of TGN prior to his ischemic stroke, one simply could attribute this to STGN, as symptoms started two to three months post-stroke. Indeed, MRI confirmation of a dorsolateral medullary infarction and physical exam findings consistent with Wallenberg syndrome suggested a symptomatic etiology. However, because two to three months also would mirror the timeframe over which collateral circulation develops, CTGN due to post-ischemic arteriogenesis compressing the previously unaffected nerve also must be considered. Surgical confirmation of the classical etiology was evidenced by nerve atrophy and compression by the lateral petrosal vein and a branch or loop of the superior cerebellar artery that had to be freed from the nerve root entry zone.

A confounding aspect of this case was that TGN returned several weeks after the first surgery despite initial relief. Following the second surgery for adhesiolysis, the patient had significant relief from right-sided facial pain and reported only mild right periorbital hyperesthesia without other sensory loss. The persistent pain, which was unaffected by either surgery, was characteristic of STGN, likely secondary to lateral medullary infarction. CN-V has three divisions, each with its

own sensory and motor distribution. STGN typically affects the area distributed by the first and second divisions of CN-V, whereas CTGN typically affects the area distributed by the second and third divisions of CN-V.[1,2]

The likelihood of facial pain is determined by the location of the infarct. Lesions to the dorsolateral medulla (as in our patient) lead to hypoesthesia and pain on the side of the lesion due to the involvement of the trigeminal descending tract and the trigeminal spinal nucleus.[2] Our patient first developed hypoesthesia to the right face four days after admission. According to Fitzek et al.[3], 50% of patients with lateral medullary strokes who initially had such hypoesthesia to temperature and pain of the ipsilateral face, developed TGN-like facial pain within 12 days to 24 months. For our patient, Wallenberg syndrome was the suspected culprit behind his persistent pain in the CN-V1 distribution. Pontine descending tractotomy might be an option for treatment of this residual pain component.[10]

This case report demonstrated a previously undescribed etiology of CTGN from arteriogenesis after VA dissection with resultant critical VA and PICA stenoses and dorsolateral medullary infarction. The proliferation and maturation of collateral circulation is well described after infarction, but collateral vessel engorgement leading to compression of the nerve root of the trigeminal nerve is an undescribed observation. Confounding this diagnosis was the more commonly referenced (though still rare) development of STGN after dorsolateral medullary infarction due to damage to several nerve tracts and nuclei in that region. Coexistent etiologies remain a distinct possibility in our case based on the intraoperative findings that clearly suggested CTGN. It is important to note this patient had no evidence of TGN prior to his stroke.

This case suggested dual causation for the development of TGN, with both classical and symptomatic components. The surgical appearance of the trigeminal nerve with atrophy secondary to vascular compression and symptomatic improvement post-surgery was evidence confirming CTGN. In addition, STGN was supported by previous case reports detailing TGN-like pain following lateral medullary stroke and by the persistent mild hyperesthesia in the CN-V1 distribution despite microvascular decompression. Furthermore, the suggested etiology of CTGN due to arteriogenesis after ischemic stroke in the vertebrobasilar circulation has not been described in the literature. We concluded that diagnosis of TGN occurring after lateral medullary infarction warrants workup to exclude a surgically correctable cause, namely the development of collateral circulation via arteriogenesis leading to CTGN.

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