Microvascular decompression of a C-2 segmental-type vertebral artery producing trigeminal hypesthesia

Case report

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The authors report a case of trigeminal hypesthesia caused by compression of the spinal cord by a C-2 segmental-type vertebral artery (VA) that was successfully treated with microvascular decompression. Aberrant intradural VA loops have been reported as causes of brainstem compression, some of which improved with microvascular decompression. A 52-year-old man presented with progressive complaints of headache, dizziness, left facial numbness, and left upper-extremity paresthesia that worsened when turning his head to the right. Magnetic resonance imaging of the cervical spine showed the left VA passing intradurally between the axis and atlas, foregoing the C-1 foramen transversarium, and impinging on the spinal cord. The patient underwent left C-1 and C-2 hemilaminectomies followed by microvascular decompression of an aberrant VA loop compressing the spinal cord. The patient subsequently reported complete resolution of symptoms.

Key Words • vertebral artery • microvascular decompression • trigeminal hypesthesia • vascular disorders

Tortuous extradural vertebral artery (VA) loops have been reported as causes of cervical radiculopathy, some of which have benefited from anterior or posterior cervical decompression.1,6,7,9,13,18,23,25 Similarly, tortuous intradural VA loops have been reported as causes of brainstem compression, some of which improved with microvascular decompression.10,12,17,19,20,29,30 Vertebral arteries with an aberrant, not tortuous, segment are more rare. Cervical myelopathy with or without radiculopathy resulting from anomalous VA anatomy has been described.3,8,21,22,24,26,27,31-34 We present a case of the successful surgical treatment of symptomatic high cervical cord compression caused by an aberrant VA segment. Interestingly, the predominant symptom of this high cervical cord compression was facial hypesthesia.

Case Report

History and Examination. A 52-year-old male police officer, formerly a Navy SEAL, with a medical history of hypertension and hyperlipidemia, presented with progressive complaints of headache, dizziness, left facial numbness, and left upper-extremity paresthesia that worsened when he turned his head to the right. Findings from previous evaluations for stroke, neoplasm, and benign positional vertigo had been negative. According to the report, MRI of the cervical spine showed cervical stenosis. On examination, the patient was alert and fully oriented. Cranial nerves were intact, except for decreased sensation to touch in the left V1, V2, and V3 distributions. Motor strength was full throughout. Sensation was diminished diffusely to light touch and pin prick in the left upper extremity without discrete dermatomal distribution. Reflexes were normoactive without evidence of Hoffman, clonus, or Babinski signs. Magnetic resonance imaging of the cervical spine showed the left VA passing intradurally between the axis and atlas, foregoing the C-1 foramen transversarium, and impinging on the spinal cord (Fig. 1). Findings of a 4-vessel cerebral angiogram were negative for arteriovenous dural fistula or other vascular abnormalities but confirmed an aberrant course of the left VA, with premature entry intradurally between C-1 and C-2 (Fig. 2). Provocative movements and positioning of the head failed to demonstrate significant movement or kinking of the artery.

This article contains some figures that are displayed in color online but in black-and-white in the print edition.
Operation. A conservative course emphasizing blood pressure control and analgesia was undertaken unsuccessfully. After discussing the risks and benefits, the patient was taken to surgery for a vascular decompression of the cervical spinal cord. The patient was placed prone, and left C-1 and C-2 hemilaminectomies were performed. Under the operative microscope, the dura was opened and the aberrant, pulsatile VA was found compressing the cervical cord (Fig. 3). A microvascular decompression was accomplished by displacing the VA and cushioning it using Teflon. This decompression was then complemented with an expansion duraplasty using a dural substitute. A watertight seal was confirmed with Valsalva maneuvers during inspection of the suture line under the microscope. Fat and dural sealant were used to supplement the repair (Video 1).

**Video 1.** Intraoperative video showing durotomy, microvascular decompression, and expansion duraplasty. Copyright Edward A. M. Duckworth. Published with permission. Click here to view with Media Player. Click here to view with Quicktime.

Postoperative Course. On postoperative Day 1, the patient was neurologically intact and reported improved symptoms. On postoperative Day 3, with continued symptom improvement, he was discharged home without evidence of new neurological deficit or CSF leak. At the 2-week follow-up, the patient reported complete resolution of symptoms. Six months later, he was back at work as a police officer and otherwise doing well.

Discussion

Structural anomalies of the vertebrobasilar system can produce a variety of neurological symptoms. In cases of direct compression of neural structures by the vertebrobasilar system, local mass effect exacerbated by the pulsatile nature of arterial blood flow can produce cranial nerve or pontomedullary dysfunction, cervical myelopathy, or cervical radiculopathy. Tortuosity of the VA, in particular, has been shown to produce extradural cervical radiculopathy as well as intradural brainstem dysfunction by direct compression. Structural abnormalities of the vertebrobasilar system can produce a variety of neurological symptoms. In cases of direct compression of neural structures by the vertebrobasilar system, local mass effect exacerbated by the pulsatile nature of arterial blood flow can produce cranial nerve or pontomedullary dysfunction, cervical myelopathy, or cervical radiculopathy. Tortuosity of the VA, in particular, has been shown to produce extradural cervical radiculopathy as well as intradural brainstem dysfunction by direct compression. Such tortuosity has been observed postmortem in anatomical studies, as well as in vivo on radiography, MRI, or via traditional angiography.

Classically, Jannetta linked vascular compression of the trigeminal nerve to trigeminal neuralgia, and showed that microvascular decompression of the affected nerve could produce symptomatic relief. In these cases, trigeminal nerve dysfunction is secondary to compression by dolichoectatic or tortuous vessels of the posterior circulation. These tortuous vessels stray from natural ana-
In our case, the patient likely had trigeminal distribution hypesthesia secondary to compression of the caudal reaches of the spinal trigeminal nucleus. The spinal trigeminal nucleus spans the medulla and upper cervical cord, receiving deep/crude touch, pain, and temperature sensations from the ipsilateral face via general somatic afferent fibers. Afferent fibers to the spinal trigeminal nucleus travel principally via branches of the trigeminal nerve, but the facial, glossopharyngeal, and vagal nerves also contribute sensory modalities from parts of the mouth, ear, and meninges. Peripheral nerve fibers, most prominently those with neurons in the trigeminal ganglion, send central projections to the brainstem, entering via the pons or medulla. These projections descend to the medulla and upper cervical cord, forming the spinal trigeminal tract, synapsing with neurons in the adjacent spinal trigeminal nucleus. Second-order neuronal projections cross the midline and ascend in the contralateral brainstem as the trigeminothalamic tract to the contralateral ventral posterior medial thalamus. Third-order neurons in the thalamus then project to neurons in the sensory cortex of the postcentral gyrus. As opposed to the classic microvascular compression of first-order neurons seen in trigeminal neuralgia, our patient likely had symptoms due to vascular compression of the central projections of these first-order neurons, as well as of the second-order neurons themselves (Fig. 4).

Cervical myelopathy may be caused by VA tortuosity, which has been observed with an incidence of 2.7% in a cadaveric study, but can also result from arteries that follow an aberrant course. A review of 300 VA angiograms by Tokuda et al. found 7 cases of anomalous atlantoaxial portions of the VA. Of these anomalous angiograms, 2 were found to have VAs coursing into the spinal canal between C-1 and C-2, as was observed in our case; the term “C2 segmental type of the vertebral artery” was used to describe this anomaly. A review of the literature reveals 11 related cases, predominantly from Japan, of similar anomalies causing clinical symptoms. In these cases, the predominant symptoms were those of cervical myelopathy, caused by unilateral or bilateral premature intradural passage of the VA, or a fenestration of the VA, resulting in compression of the spinal cord. Interestingly, none of the patients in the aforementioned cases reported trigeminal distribution symptoms.

Such VA anomalies may be more often symptomatic due to the limited diameter of the upper cervical canal, which, though generous, is not as wide as the spinal canal at the foramen magnum and cannot accommodate an intruding vascular structure without cervical cord compression. In some of the reported cases, surgical intervention was undertaken. Most commonly, posterior cervical approaches with some combination of suboccipital craniectomy, cervical laminectomy, and expansion duraplasty were used. Vascular decompression was frequently used; however, an additional technique often described is VA transposition followed by tacking using suture or prostheses.

While cervical myelopathy secondary to aberrant VA compression has been described, our case seems to be the first reported instance of trigeminal hypesthesia from vascular compression of the spinal trigeminal tract and nucleus.
Vertebral artery compression of the cervical spinal cord can produce symptoms of cervical myelopathy or cervicomedullary compression. Though the literature is sparse on the topic, several case reports have linked such compression to premature or aberrant intradural entry of the VA into the spinal canal. None of the previously reported cases, however, mention trigeminal distribution dysfunction as a predominant symptom. Surgical interventions have been undertaken with good effect. Our case supports the notion that surgical decompression of VA compression of the high cervical cord provides good outcomes, particularly in the relief of trigeminal hypesthesia. It remains unclear, however, which aspect of the surgical intervention—microvascular decompression with Teflon or cervical laminectomy with expansion duraplasty—resulted in symptomatic relief, if not both.

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Disclosure
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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References

Fig. 4. Illustration demonstrating postulated compression of the spinal trigeminal tract (blue column) by a pulsatile, aberrant VA abutting the lateral spinal cord. Copyright Jovany Cruz Navarro. Published with permission.