Resolution of intractable retching following mobilization of a dolichoectatic vertebral artery: case report of a unique brainstem–cranial nerve compression syndrome

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The authors present the case of a 53-year-old man who was referred with disabling retching provoked by left arm abduction. At the time of his initial evaluation, a cervical MRI study was available for review and revealed an anatomical variation of the ipsilateral juxtamedullary vertebrobasilar junction. After brain imaging revealed contact of the medulla by a dolichoectatic vertebral artery at the dorsal root entry zone of the glossopharyngeal and vagus nerves, the patient was successfully treated by microvascular decompression of the brainstem and cranial nerves. This case demonstrates how a dolichoectatic vertebral artery—a common anatomical variation that typically has no clinical consequence—should be considered in cases of cranial nerve dysfunction.

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Eight months later, the video barium swallow was repeated due to persistent and increasingly disabling episodes of gagging and retching. Once again, the initial assessment was unexpectedly normal; however, at the patient’s suggestion, it was repeated as he abducted his left arm. During this maneuver, a gag reflex was provoked and outpouching and deviation of the lower cervical esophagus to the left of midline was observed (Video 2).

**VIDEO 2.** Video clip depicting the disruption of contrast flow within the esophagus as the left shoulder is abducted during a video swallow. Copyright Mount Nittany Medical Center. Published with permission. Click here to view.

A review of the cervical spine MR scans obtained prior to our consultation revealed findings of multilevel spondylosis of moderate severity, but without significant spinal cord or nerve root compression. However, the sagittal T2-weighted images depicted an ectatic vertebrobasilar junction contacting the left side of the medulla oblongata (Fig. 1). To further characterize the anatomical features of the posterior fossa vasculature, a CT angiogram was performed, followed by a 3D reconstruction of the vertebral and posterior inferior cerebellar arteries (Fig. 2). To then define the anatomical relationships between the vessels, the brainstem and cranial nerves, axial 3D FIESTA (fast imaging employing steady state acquisition) MR sequences were obtained. These images revealed ectasia of the vertebrobasilar junction contacting the left lateral aspect of the medulla without imaging evidence of compression of either the glossopharyngeal or vagus nerves (Fig. 3). DICOM images of the patient were acquired preoperatively and used to produce 3D reconstructions using OsiriX imaging software (http://www.osirix-viewer.com). A 3D reconstruction of the skull base and cervical spine was created using the 3D volume rendering feature within the OsiriX software. Portions of the occipital bone and posterior cervical spine were selectively removed to improve visualization of the bony anatomy along with the enhancing intracranial vascular structures. A 64-bit color look-up table (CLUT) was then used to optimize the visibility of bony and enhancing vascular structures. Outlines of these anatomical structures were traced from the 3D reconstructions and rendered by a medical illustrator in Photoshop. This 3D reconstruction of the posterior fossa vasculature depicted the ectatic vertebrobasilar junction forming to the left of midline with irregular tortuous loops of each posterior inferior cerebellar artery (PICA) (Fig. 4).

We speculated that arm elevation and neck flexion or extension could provoke changes in the position of either vertebral artery or the left PICA at the site of brainstem contact and then subsequently alter the function of adjacent cranial nerves or brainstem nuclei that might then precipitate retching. When considering accepted anatomical and physiological principles of the retching phenomenon, we hypothesized that dysfunction of the glossopharyngeal and vagus nerves were implicated in this case. With this in mind, an operative plan was designed to perform a microvascular decompression of the glossopharyngeal and vagus nerves.

**Operative Treatment**

A left-sided suboccipital craniotomy was performed with the patient in the lateral oblique position and with electrophysiological monitoring of cranial nerve function. The junction of the transverse and sigmoid sinuses was exposed, and the dura was incised and tacked up with tenting sutures. Under microscopic visualization, the arachnoid of the dorsal cerebellopontine cistern was incised, the cerebospinal fluid was aspirated, and the superior petrosal vein was exposed, coagulated and then incised. The cerebellar cortex was protected with cottonoids, and the cerebellar hemisphere was allowed to retract away from the petrous bone, exposing the cerebellopontine cleft. The lateral pontomedullary membrane was incised, and arachnoid membranes within the cerebellopontine and cerebel-lomedullary cisterns were sectioned. The ectatic vertebral artery was found to lie against the medulla ventral to the root entry zone of the glossopharyngeal and vagus nerves and loop near, but without contacting, the dorsal inferior aspect of the facial and vestibular nerves. The arachnoid attachments were sectioned between the superior cerebellar artery and the trigeminal nerve, the anterior inferior cerebellar artery and the facial and vestibular nerves, and the left PICA and the glossopharyngeal, vagus, and hypoglossal nerves. This allowed the left PICA and vertebral artery to be mobilized away from the brainstem without any subsequent traction upon the facial and vestibular nerves. The glossopharyngeal and vagus nerves were then cushioned from arterial pulsations with a thin layer of shredded Teflon.

**Postoperative Course**

In the recovery room, the patient was able to abduct his left arm without retching. On postoperative Day 2, he developed intermittent episodes of asymptomatic atrial...
flutter. A cardiac echo ordered by the consulting cardiologist revealed mild concentric left ventricular hypertrophy, a mildly dilated left atrium, and atrial flutter without significant valvular abnormalities. The atrial flutter resolved after the administration of metoprolol, 25 mg by mouth every 6 hours. The patient was discharged to home on the 5th postoperative day.

At the 3-month postoperative follow-up visit, he reported infrequent episodes of tingling in his throat with a sensation of imminent retching. At 8 months after surgery, after several hours doing yard work including raking leaves, the patient had one episode of retching with arm abduction. He restricted his activities of daily living to simple household tasks for several days and was then able to resume an active lifestyle without any recurrent episodes. One year following surgery, the retching episodes have resolved. He travels throughout the country by commercial airlines to inspect construction sites, can perform aerobic exercises, and using a pedometer while on vacation, recorded walking 11.8 miles 1 day without experiencing any episodes of retching.

**Discussion**

The imaging findings of the dolichoectatic vertebral artery contacting the lateral aspect of the medulla raised a clinical suspicion that brainstem or cranial nerve compression might be implicated in this case. The neurophysiological regulation of vomiting is complex and incompletely understood. In a review, Babic and Browning describe that the nucleus tractus solitarius (NTS) receives
afferent inputs from the trigeminal, vestibular, glossopharyngeal, and vagus nerves and then coordinates the emetic response through efferent pathways to the ventral respiratory group, nucleus ambiguus, and dorsal motor nucleus of the vagus nerve within the medullary reticular formation of the ventrolateral medulla. They go on to assert that these structures initiate swallowing and salivation mechanisms within the oropharynx and coordinate the respiratory, cardiovascular, and gastrointestinal systems involved in vomiting. The act of retching is mediated by afferent inputs from the glossopharyngeal nerve arising from the pharynx or posterior one-third of the tongue; the nerve fibers proceed to the inferior glossopharyngeal ganglion and subsequently to the NTS and spinal trigeminal nucleus. This pathway synapses with the nucleus ambiguus in the lateral medulla to form an efferent arc to the glossopharyngeal and vagus nerves. Pharyngeal branches then innervate the pharyngeal muscles of the larynx and pharynx, the striated muscles of the upper esophagus, the uvula, levator veli palatine, and palatoglossus muscle to complete the reflex.

While retching can resemble the act of vomiting, it does not result in the expulsion of gastric contents. As demonstrated in our patient (Video 2), the anatomical alterations of his esophagus that were provoked by left arm motion did not lead to vomiting. Following a thorough review of the radiographic literature, no video images could be identified capturing the act of retching during the performance of a barium swallow. Furthermore, the depiction of unilateral esophageal outpouring with arm abduction during the performance of a barium swallow has not been previously reported. We submit that this observation was a consequence of altered central nervous impulses transmitted by the left glossopharyngeal and vagus nerves to the left-sided pharyngeal musculature.

Compression of cranial nerves within the posterior fossa by tumors or vascular anomalies can lead to combinations of pain, sensory loss, and muscle weakness throughout the face and within the oropharynx. In 1934, Dandy proposed that trigeminal neuralgia was caused by compression of the trigeminal nerve by adjacent arteries, with the superior cerebellar artery being involved in nearly one-third of his 215 operative cases. In the modern era, Rhoton’s manuscripts detailing the microsurgical anatomy and operative approaches within the posterior cranial fossa, along with Jannetta’s reports of the microvascular decompression technique of cranial nerves, have pioneered surgical therapies for disabling neurological conditions such as trigeminal neuralgia, hemifacial spasm, and glossopharyngeal neuralgia. In the microvascular decompression literature, the most common cause of cranial nerve compression is tortuous branches of the vertebrobasilar vasculature. Reports have even documented the use of surgical techniques to mobilize and reposition dolichoectatic vertebral arteries, along with their branches, in patients with dysphagia, dysphonia, and weakness of the extremities—deficits that were attributed to brainstem compression.

Vertebrobasilar dolichoectasia refers to the enlargement or dilation of a vessel. In many patients, this finding simply represents an anatomical variation of uncertain
clinical consequence; in fact, there are reports of MR findings in asymptomatic patients with documented compression of cranial nerves and the brainstem by dolichoectatic vessels. Although diagnostic criteria for vertebrobasilar dolichoectasia or tortuous vertebral arteries are not well established, one report suggested defining vertebrobasilar ectasia as an arterial diameter exceeding 4.5 mm as depicted on MR images. This is consistent with previous radiographic studies suggesting a normal basilar artery diameter of 1.86–4.53 mm on CT imaging. Modern imaging has increasingly identified asymptomatic anatomical variations, including dolichoectasia of the vertebral arteries; however, such findings are seldom described within radiographic reports. This case demonstrates how a common anatomical variation of the vertebral artery was responsible for a peculiar and disabling neurological problem and highlights the importance of the treating physician consulting with the interpreting radiologist was responsible for a peculiar and disabling neurological condition of the trigeminal and facial nerves—and to a lesser extent, the glossopharyngeal nerve—has been well documented. Reports of vascular compression of the lower cranial nerves are far fewer in number. Vascular compression of the hypoglossal nerve can lead to hemilungual spasms exacerbated by talking, chewing, or emotional stress; likewise, compression of the accessory nerve has been documented to result in spastic torticollis. Finally, compression of the vagus nerve can result in presyncopal and syncopal episodes concomitant with glossopharyngeal neuralgia or hemifacial spasm, intractable hiccups, or dysphagia. There have been additional reports of dolichoectatic vessels, most commonly the vertebral artery, that cause brainstem compression producing neurological dysfunction. These anatomical sites include: the area postrema, leading to chronic emesis; the medulla oblongata, causing respiratory failure and/or dysphagia; in addition to tinnitus, hoarseness, and ataxic gait; central sleep apnea, and diaphragmatic paralysis; the ventrolateral medulla at the root entry zone of cranial nerve IX–X, as depicted in Fig. 4, thus provoking our patient’s retching reflex. We hypothesize that left arm abduction triggered these symptoms by altering the local physical relationship between the ectatic vertebral artery and the brainstem and cranial nerves. This physical relationship could be influenced by alterations of flow of either the cerebrospinal fluid within the subarachnoid space or by blood within the vertebral artery or its branches. In either case, we submit that the shaded Teflon alters the local pulsatile forces acting upon the brainstem and cranial nerves.

A case reported by Resnick and Jannetta shares many clinical features with our case. They describe a patient who underwent a microvascular decompression of the superior vestibular nerve for treatment of disabling positional vertigo and then developed chronic intractable gaging and was found to have a hyperactive gag reflex upon examination. Upon re-exploration of the operative site performed 2 years after the original operation, they discovered that the vertebral artery and posterior inferior cerebellar artery were compressing the glossopharyngeal and vagus nerves. After a microvascular decompression was performed, these episodes of gagging resolved. The findings of their case lend support to the notion that vascular compression of the brainstem nuclei or nerves of the glossopharyngeal-vagus complex can be associated with unusual oral-pharyngeal reflexes. Their case was notable for a hyperactive gag reflex and trigger points within the left side of the patient’s mouth. While similarities exist between the case of Resnick and Jannetta and ours, important differences are to be noted. First, our patient’s episodes were exclusively provoked by left arm abduction, as seen in Video 1. Second, he had a normal gag reflex and his episodes of retching were not provoked by chewing or swallowing. Third, our patient had not undergone posterior fossa surgery. Finally, in their report, the operative description of the implicated vessels, the vertebral and the posterior inferior cerebellar arteries, were not described as having any anatomical variations such as the ectatic nature of the vertebral artery in our case. When taken together,
the factors of our case define a unique and previously un-reported case of a neurovascular compression syndrome.

Conclusions

This case suggests that vascular compression of the medulla and glossopharyngeal and vagus nerves should be considered in patients with intractable retching. While this is the second case in the literature of a patient with intractable retching successfully treated with a microvascular decompression, it represents the first case that did not immediately arise after a previous microvascular decompression, it represents the first case documenting how arm movement in those with a dolichoectatic vertebrobasilar artery can provoke brainstem–cranial nerve dysfunction leading to ipsilateral esophageal outpouring and retching.

References


Disclosures

The authors report no conflict of interest concerning the materi-
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Supplemental Information
Videos


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Conception and design: Fick, Seaman, Nelson, Swift. Acquisition of data: Fick. Analysis and interpretation of data: Fick, Seaman, Alexander. Drafting the article: Fick. Critically revising the article: all authors. Reviewed submitted version of manuscript: Fick, Seaman, Nelson. Approved the final version of the manuscript on behalf of all authors: Fick. Administrative/technical/material support: Fick, Seaman. Preparation of medical illustrations: Swift.

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