

# ***Cervical Cord Compression by Aberrant Vertebral Artery Presenting with Neuralgic Pain: A Case Report***

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## **Abstract**

The vertebral artery (VA) is often affected by anatomical variations; however, it is usually asymptomatic and is found incidentally. Herein, we report a case of cervical cord compression caused by bilateral aberrant VAs. A 65-year-old woman presented with paroxysmal lancinating pain in the neck that later extended to the shoulders and upper arms bilaterally. Magnetic resonance imaging and computed tomographic angiography revealed an aberrant course of both VAs entering the spinal canal between the atlas and axis and compressing the cervical cord at the atlas level. Microvascular decompression was performed with transposition of the VAs, and the pain resolved immediately after the surgery. A certain number of anatomical variants of the distal VA can be explained by size variations and connection of the lateral spinal artery (LSA). Considering that an aberrant VA may correspond to an enlarged LSA, optimal transposition should be performed to prevent neurological complications.

Keywords: cervical spinal cord compression, microvascular decompression, aberrant vertebral artery, lateral spinal artery

## **Introduction**

The V3 segment of the vertebral artery (VA) usually enters the intradural space through the superior surface of the C1 posterior arch. However, the VA may occasionally penetrate the dura between the atlas and axis and run along the spinal cord, which is known as a persistent first intersegmental artery,<sup>1,2)</sup> C2 segmental-type VA,<sup>3)</sup> anomalous VA,<sup>4,5)</sup> or aberrant VA.<sup>6)</sup> Symptomatic spinal cord compression due to an aberrant VA is very rare. Herein, we describe a case that was successfully treated with microvascular decompression (MVD) and discuss the surgical treatment based on the developmental anatomy of the distal VA.

## **Case Report**

A 65-year-old woman experienced paroxysmal lancinating pain in the neck for 4 years. This neuralgic pain gradually extended to the shoulders and then to the upper arms bilaterally and worsened on the left side. The pain occurred intermittently, even during sleep, irrespective of the

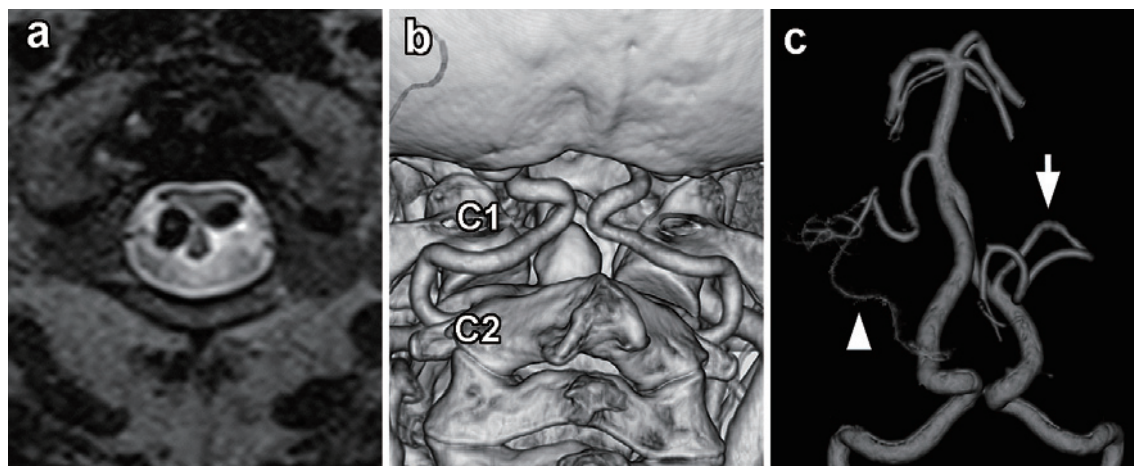
neck motion. Analgesic medications, including nonsteroidal anti-inflammatory drugs, pregabalin, baclofen, and carbamazepine, failed to relieve her symptoms. The patient had a history of hypertension that was adequately controlled with antihypertensive medications. No neurological abnormalities were found. Magnetic resonance imaging revealed bilateral signal-void areas in the posterolateral region of the spinal cord at the atlas level. The spinal cord was severely compressed and deformed by both VAs (Fig. 1a). Three-dimensional computed tomography angiography revealed an aberrant course of both VAs, which entered the spinal canal between the atlas and the axis (Fig. 1b, c). Based on these findings, we speculated that cervical cord compression by aberrant VAs was responsible for her symptoms.

MVD of the cord with VA transposition was performed. Suboccipital craniectomy and C1 laminectomy were performed *via* a midline skin incision. Both VAs coursed medially after entry into the spinal canal to indent the posterolateral aspect of the cervical cord (Fig. 2a), with the compression being more severe on the left side. Each VA was transposed using Teflon slings and adhered to the

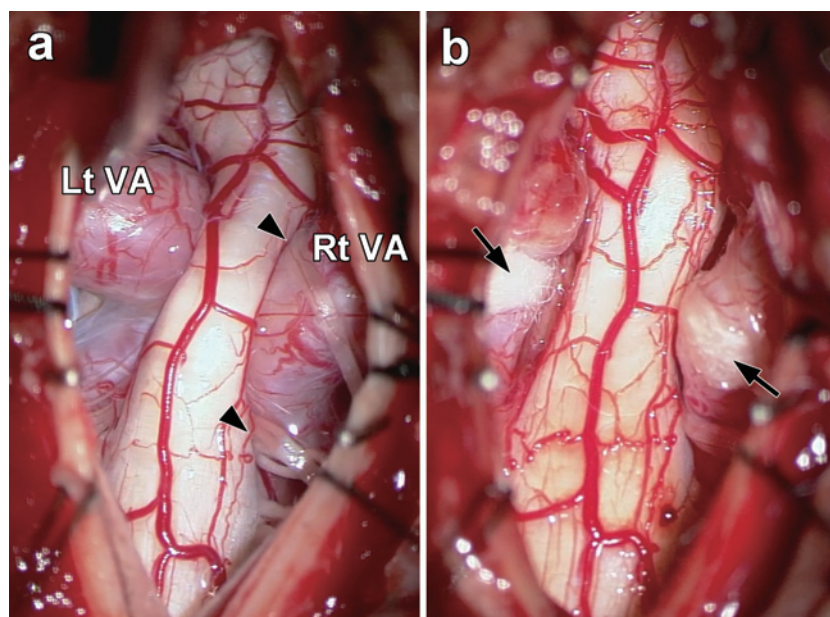
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**Fig. 1** (a) Preoperative magnetic resonance imaging showing bilateral vascular loops compressing and distorting the spinal cord. (b) 3DCTA showing that the VAs enter the spinal canal between the atlas and the axis. The C1 posterior arch is deleted with image processing. (c) 3DCTA showing an aberrant course of both VAs. The right PICA (arrow) originates from the intracranial segment of the VA, and the left PICA (arrowhead) originates from the basilar artery as a common trunk with the anterior inferior cerebellar artery. PICA, posterior inferior cerebellar artery; 3DCTA, three-dimensional computed tomographic angiogram; VA, vertebral artery



**Fig. 2** Intraoperative photographs. (a) Bilateral VAs compress the spinal cord. The C2 dorsal rootlets (arrowheads) are observed on the surface of the VA. (b) After transposition of the VAs using Teflon slings (arrows), a small space is created between the VAs and the spinal cord. VA, vertebral artery

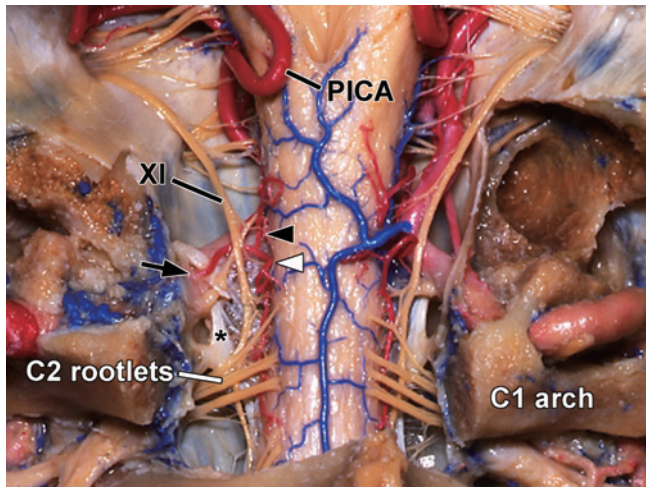
dura mater using fibrin glue. The VA provided small branches to the cervical cord; therefore, the transposition was restricted. Although the space between the VAs and the spinal cord was minimal, spinal cord decompression was maintained (Fig. 2b). The postoperative course was uneventful, and the patient remained pain-free during the 18-month follow-up period.

## Discussion

### Anatomy of the lateral spinal artery

The posterior spinal artery (PSA) supplies the posterior aspect of the spinal cord and is classically described as a pair of longitudinal arteries that runs dorsal to the dorsal rootlets. However, in the upper cervical region, the main posterior anastomotic pathway runs ventrally to the dorsal rootlets and is also termed the lateral spinal artery (LSA).<sup>7)</sup>

The LSA usually originates from the VA, either intra- or extradurally, or from the posterior inferior cerebellar artery (PICA) and divides into ascending and descending

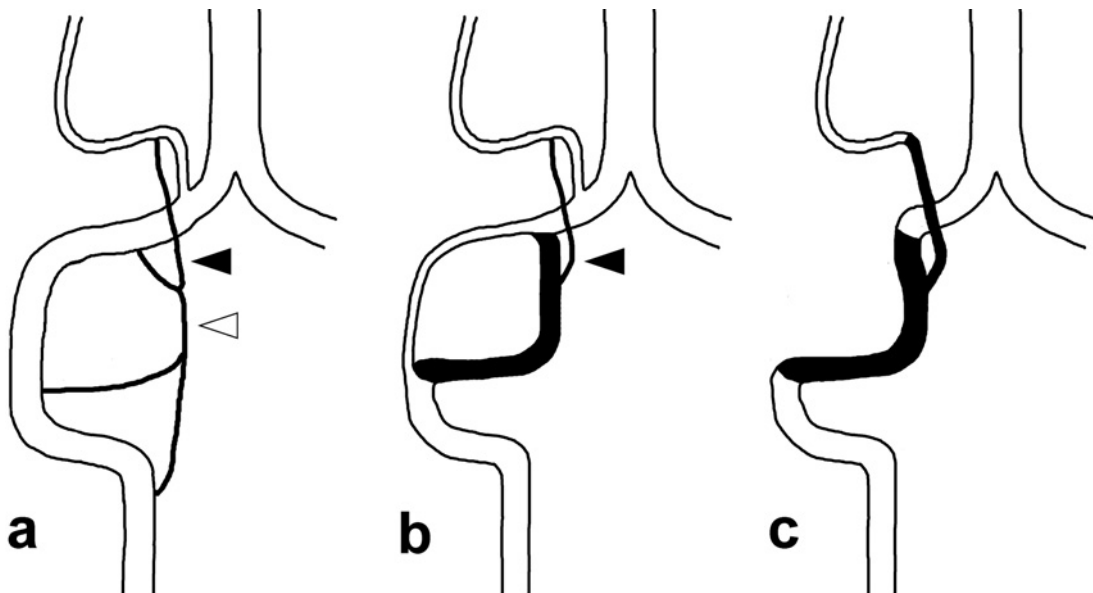


**Fig. 3** Photograph of cadaveric specimen demonstrating the normal anatomy of the LSA. The LSA (arrow) arises from the vertebral artery and divides into ascending (black arrowhead) and descending (white arrowhead) branches. The descending branch courses ventral to the dorsal rootlets and dorsal to the dentate ligament (asterisk). The photograph was provided by Dr. Masatou Kawashima. LSA, lateral spinal artery; PICA, posterior inferior cerebellar artery; XI, accessory nerve

branches (Fig. 3). The ascending branch supplies the posterior medulla oblongata and anastomoses with the PICA branches (Fig. 4a). The descending branch passes downward between the dorsal rootlets and dentate ligament and gives pial arteries to the posterior and lateral surfaces of the spinal cord. The descending branch of the LSA anastomoses with the extradural arteries arising from the VA at each metameric level and caudally at the C4 or C5 level, joining the ipsilateral PSA running dorsal to the dorsal rootlets.<sup>8)</sup>

#### Developmental anatomy of the distal VA

In the embryonic period, the VA does not develop as a proper artery possessing its own specific territory; instead, it results from longitudinal anastomoses between the cervical intersegmental arteries.<sup>9)</sup> With the transformation of anastomoses to a single lumen, primitive VA is formed. The extradural and intradural anastomoses of these metameric arteries play significant roles in the development of VA and LSA, respectively. A certain number of anatomical variations involving the distal VA can be explained by size variations and connection of the LSA. These variations include the C1 and C2 origins of the PICA, duplication of the distal VA, and aberrant course of the distal VA.<sup>7,10)</sup> The aberrant course of the distal VA is an extreme form of the VA duplication in which the limb corresponding to the normal VA is diminutive or absent, resulting in a seemingly “aberrant” VA course (Fig. 4b, c). In



**Fig. 4** Schematic representation of the normal anatomy and its variations of the LSA. The LSA and its corresponding segments are shown in black. (a) The descending branch of the LSA (white arrowhead) has lateral segmental connections with the VA at C2 and C3, and the ascending branch of the LSA (black arrowhead) has a cranial anastomosis with the PICA. (b) Distal VA duplication above and below the C1 arch. The black arrowhead indicates the ascending branch of the LSA. (c) If the true distal VA is absent, the variant becomes an aberrant course of the VA. When the PICA originates from the aberrant VA, the proximal segment of the PICA may correspond to the ascending branch of the LSA. LSA, lateral spinal artery; PICA, posterior inferior cerebellar artery; VA, vertebral artery



our operative findings, a normal LSA could not be observed on the surface of the spinal cord; instead, the aberrant VA ran between the dorsal rootlets and the dentate ligament and directly gave off short pial arteries, corresponding to the descending branch of the LSA. Furthermore, an ascending branch arose from the aberrant VA and gave pial arteries to the posterior medulla, corresponding to the ascending branch of the LSA. When the PICA originates from an aberrant VA, as has been reported in several cases,<sup>2,4-6)</sup> the proximal segment of the PICA may correspond to the ascending branch of the LSA (Fig. 4c). In our case, the right PICA originated from the intracranial segment of the VA, and the left PICA originated from the basilar artery as a common trunk with the anterior inferior cerebellar artery (Fig. 1c).

### Clinical presentation

Although aberrant VA is usually asymptomatic and found incidentally, it rarely manifests with clinical symptoms as a result of compression of the cervical cord and/or nerve rootlets. The symptoms include neuralgic pain, motor weakness, sensory disturbances, and urinary incontinence. Spasmodic torticollis due to compression of the accessory nerve<sup>11)</sup> and trigeminal hypoesthesia due to compression of the spinal trigeminal tract<sup>3)</sup> have also been reported. In the present case, determining the indications for surgical treatment was difficult because the patient exhibited intermittent pain without obvious neurological deficits. Objective findings are sometimes mild or not obvious despite marked deformity of the spinal cord as reported by some cases.<sup>4,5)</sup> We considered that the pain was caused by the compression of the dorsal root entry zone of the C2 nerve. The extension of pain from the neck to the shoulder and arm may have been caused by compression of the Lissauer tract, containing pain fibers, which lies in front of the dorsal root entry zone and decussates one to several segments above the level of root entry.<sup>2)</sup>

### Surgical considerations

MVD of aberrant VA has been performed using the interposition method, transposition method, or their combination. The interposition method involves insertion of a prosthesis between the vessel and nerve, whereas the transposition method involves repositioning of the vessel away from the nerve. Previous studies have demonstrated that the transposition method has a better outcome than the interposition method in patients with trigeminal neuralgia or hemifacial spasms.<sup>12,13)</sup> The interposition method is sometimes insufficient to buffer a large pulsatile force caused by the VA and further exacerbates compression by the placement of a prosthesis between the vessel and nerve. An ideal transposition is aberrant VA repositioning to free the spinal cord from contact. However, a strong rebound of the VA, branching vessels, and a crowded space may preclude complete transposition of the VA, and occa-

sionally, a small vessel branching from the aberrant VA is sacrificed to achieve complete transposition.<sup>1)</sup>

Considering that an aberrant VA corresponds to an enlarged descending branch of the LSA, the spinal cord may be congenitally attached to or even deformed by the aberrant VA as the descending branch of the LSA usually runs on the surface of the spinal cord, giving off short pial arteries. This idea may be supported by cases in which the symptoms were present only on one side despite bilateral compression of the spinal cord by the VAs.<sup>5,14)</sup> A slight deviation of the VAs caused by aging or atherosclerosis may be associated with a late onset of symptoms, as most patients develop symptoms after the middle age.<sup>15)</sup> Therefore, a small gap between the spinal cord and VA may be sufficient to relieve symptoms, and excessive transposition of the VA should not be performed to avoid ischemic complications. Our patient remained pain-free, although the cervical cord was still deformed. However, long-term follow-up is essential to monitor its durability.

### Conclusion

MVD is an effective treatment for spinal cord compression caused by VAs. Considering that an aberrant VA may correspond to an enlarged descending branch of the LSA, optimal transposition should be performed to prevent neurological complications.

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### Patient Consent

Informed consent was obtained from the patient for the publication of this case report.

### Conflicts of Interest Disclosure

All authors have no conflicts of interest.

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