Trigeminal neuralgia caused by a trigeminocerebellar artery

Case report

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This 31-year-old woman presented with typical right trigeminal neuralgia caused by a trigeminocerebellar artery, manifesting as pain uncontrollable with medical treatment. Preoperative neuroimaging studies demonstrated that the offending artery had almost encircled the right trigeminal nerve. This finding was confirmed intraoperatively, and decompression was completed. The neuralgia resolved after the surgery; the patient had slight transient hypesthesia, which fully resolved within the 1st month after surgery. The neuroimaging and intraoperative findings showed that the offending artery directly branched from the upper part of the basilar artery and, after encircling and supplying tiny branches to the nerve root, maintained its diameter and coursed toward the rostral direction of the cerebellum, which indicated that the artery supplied both the trigeminal nerve and the cerebellum. The offending artery was identified as the trigeminocerebellar artery. This case of trigeminal neuralgia caused by a trigeminocerebellar artery indicates that this variant is important for a better understanding of the vasculature of the trigeminal nerve root.

Key Words • trigeminal neuralgia • trigeminocerebellar artery • microvascular decompression • 3D magnetic resonance imaging • functional neurosurgery

Abbreviations used in this paper: AICA = anterior inferior cerebellar artery; BA = basilar artery; SCA = superior cerebellar artery.
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Midad approach. Microscopic dissection confirmed that an artery had formed loops almost encircling the nerve, compatible with the findings of the 3D fusion image (Fig. 3C and D). Careful dissection of the artery was necessary, and very small branches that pierced the nerve had to be sacrificed. The artery maintained its diameter and coursed toward the rostral direction of the cerebellum. The dissection succeeded in freeing the nerve from the artery, which was fixed with Teflon slings. Complete decompression of the nerve was confirmed (Fig. 3E). Postoperatively, the patient’s neuralgia disappeared, but she experienced transient hypesthesia of the V2 distribution, which had fully resolved at 1-month follow-up. She was discharged from our hospital with no other neurological deficit.

Discussion

The microsurgical anatomy of the trigeminal nerve root includes several arteries that contact or supply blood to the nerve root, including the SCA, AICA, and the posterior cerebellar, vertebral, and basilar arteries and branches from these arteries.3,8,9,11 All these arteries have the potential to cause trigeminal neuralgia. Nerve compression is often caused by a combination of vessels, including various veins, and the offending vessel may not be definitively identified during the operation.3 In the present case, the intraoperative finding was the extremely unusual ring-like compression of the trigeminal nerve by an artery.

The preoperative radiological studies in our case demonstrated an independent artery directly branching from the upper part of the BA; this branching vessel was considered to be the offending artery, but it was not the SCA or AICA. Therefore, the pontine artery, pontine branch from the BA, or a variant could be a candidate for the offending artery. However, the pontine branch or artery should be responsible for blood supply to the pons or the nerve root, with branching perforating arteries, but not further supply to the cerebellum.14 Intraoperative examination showed that after the artery encircled and supplied tiny branches to the nerve root, the diameter was maintained, and it coursed toward the rostral part of the cerebellum, which suggested that the artery supplied both the trigeminal nerve and the cerebellum. Therefore, none of the usual arteries could be identified as the offending artery.

The trigeminocerebellar artery has been defined as a unique branch of the BA supplying both the trigeminal nerve root and the cerebellar hemisphere, as described by Marinkovic et al. in 1996, and more cases have been demonstrated with cadaver dissection.10,15 The trigeminocerebellar artery can be divided into the pontine, trigeminal, cisternal, and cerebellar segments, and it is thought to be related to the embryonic trigeminal artery. There are insufficient data about the prevalence of the trigeminocerebellar artery, but the etiology of the artery seems to be related to the developmental process. Failure in the regression of the embryonic trigeminal artery would result in a primitive trigeminal artery, and a trigeminocerebellar artery could represent the caudal remnant of the primitive trigeminal artery.15 The trigeminal segment sometimes twists around or encircles the nerve root and occasionally tilts the root, which has potential clinical significance as a cause of trigeminal neuralgia (Fig. 3F).10,15 In our case, the offending artery clearly branched directly from the distal part of the BA, and the intraoperative findings confirmed that the artery almost encircled the trigeminal nerve and thereafter coursed toward the cerebellum. The exact distribution of the artery in the cerebellum related to the SCA, AICA, and posterior cerebellar artery was not clear because no angiographical evidence was available, but we concluded that the trigeminocerebellar artery was the correct identification for the offending artery. The trigeminocerebellar artery has previously been identified only by cadaver dissection, whereas our case has demonstrated this artery in a surgical patient.

Three-dimensional visualization of the neurovascular anatomy has improved in recent years. Preoperative visualization of the neurovascular relationship for microvascular decompression now allows both preoperative evaluation and the virtual reality for operative simulation.5,6,13 Our preoperative neuroimaging studies do not include 3D fusion imaging in every patient with trigeminal neuralgia if typical vascular contact is observed on gradient echo imaging and MR angiography. In the present case, further 3D fusion images were obtained because how and which
arteries from the distal part of the BA conflicted with the nerve root remained unclear. The present case demonstrated the high reliability of 3D fusion imaging because the preoperative 3D fusion image perfectly matched the operative findings.

The pediatric onset of trigeminal neuralgia in the present case may be an important clinical feature of the trigeminocerebellar artery. The age of 16 years is very early for the onset of trigeminal neuralgia, and pediatric onset is rare. Previously reported cases of trigeminal neuralgia in pediatric or young adult patients predominantly involved venous compression as the causative factor.\(^1\)\(^2\) In contrast, our patient’s trigeminal neuralgia was caused only by compression from an artery. Anomalous formation of the trigeminocerebellar artery in the developmental process may explain the early pathological contact with the nerve that resulted in such pediatric onset.

However, we can only speculate based on a single case, and the clinical implications of the trigeminocerebellar artery still require elucidation. The exact distribution and other functions of the trigeminocerebellar artery remain unknown because no other pathological condition, such as cerebral infarction, has been reported. Experience with similar cases is necessary to improve recognition of this artery and increase understanding of the vasculature of the trigeminal nerve root.

**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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revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Amagasaki.

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