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CASE REPORT

Bilateral vasculopexy of anomalous vertebral arteries causing cervicomedullary compression: case report and technical note

Abhidha Shah · Amit Mahore · Atul Goel

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Abstract
Introduction The authors report an extremely rare cause of cervicomedullary cord compression by anomalous ectatic vertebral arteries.
Material A 50-year-old male patient presented with a 9 month history of progressive quadriparesis. Investigations revealed that the vertebral arteries on both sides had a mirror-like course and caused a deep indentation into the high cervical cord. Bilateral vasculopexy was done using Teflon slings. The treatment resulted in rapid recovery from symptoms.
Conclusions Anomalous course of the vertebral artery can result in symptoms of high cervical cord compression. Vasculopexy can result in lasting cure from symptoms.

Keywords Anomalous vertebral arteries · Cervicomedullary junction · Compressive myelopathy · Vasculopexy

Introduction

Cervicomedullary (CM) compression due to a variety of bony and soft tissue abnormalities has been frequently identified [1, 9, 11]. We report an extremely rare cause of cervical cord compression by abnormally coursing vertebral arteries on both sides. Vertebral arteries on both sides deeply indented into the spinal cord and resulted in symptoms of myelopathy. The management issues of the case are discussed.

A. Shah · A. Mahore · A. Goel (✉)
Department of Neurosurgery, Seth G.S. Medical College and King Edward VII Memorial Hospital, Parel, Mumbai 400012, India
e-mail: atulgoel62@hotmail.com

Case report

History and examination

A 50-year-old right-handed man, recently diagnosed to have hypertension, presented with symptoms of insidious onset and gradually progressive imbalance on walking and weakness of all four limbs for about 9 months. When admitted, he was severely crippled and needed support for all personal activities. He was unable to perform fine motor activities with both his hands and was able to walk only with support. Clinical examination revealed grade 4 spastic quadriparesis, power in the right side limbs being worse. A sensory examination showed impaired proprioception and pinprick sensation bilaterally, worse on the left side. All kinaesthetic sensations were severely impaired. Romberg's sign was positive. MR imaging revealed large flow voids in the region of the craniovertebral junction that resulted in cord compression. MR and CT angiography revealed bilateral anomalous vertebral arteries which did not pass through the transverse foramen of the atlas but turned medially after exiting the axis. The arteries entered the C1–2 interlaminar space below the posterior arch of the atlas, pierced the dura and looped medially 'kissing' each other on the dorsal surface of the cervicomedullary region deeply indenting into the cord substance (Fig. 1). There was no other bone or soft tissue anomaly in the region. There was no evidence of instability.

Operation

A midline suboccipital craniectomy with C1 laminectomy and C2 partial laminectomy was performed. The extradural vertebral arteries were identified as they exited the transverse foramen of the axis. Vertebral arteries on both sides

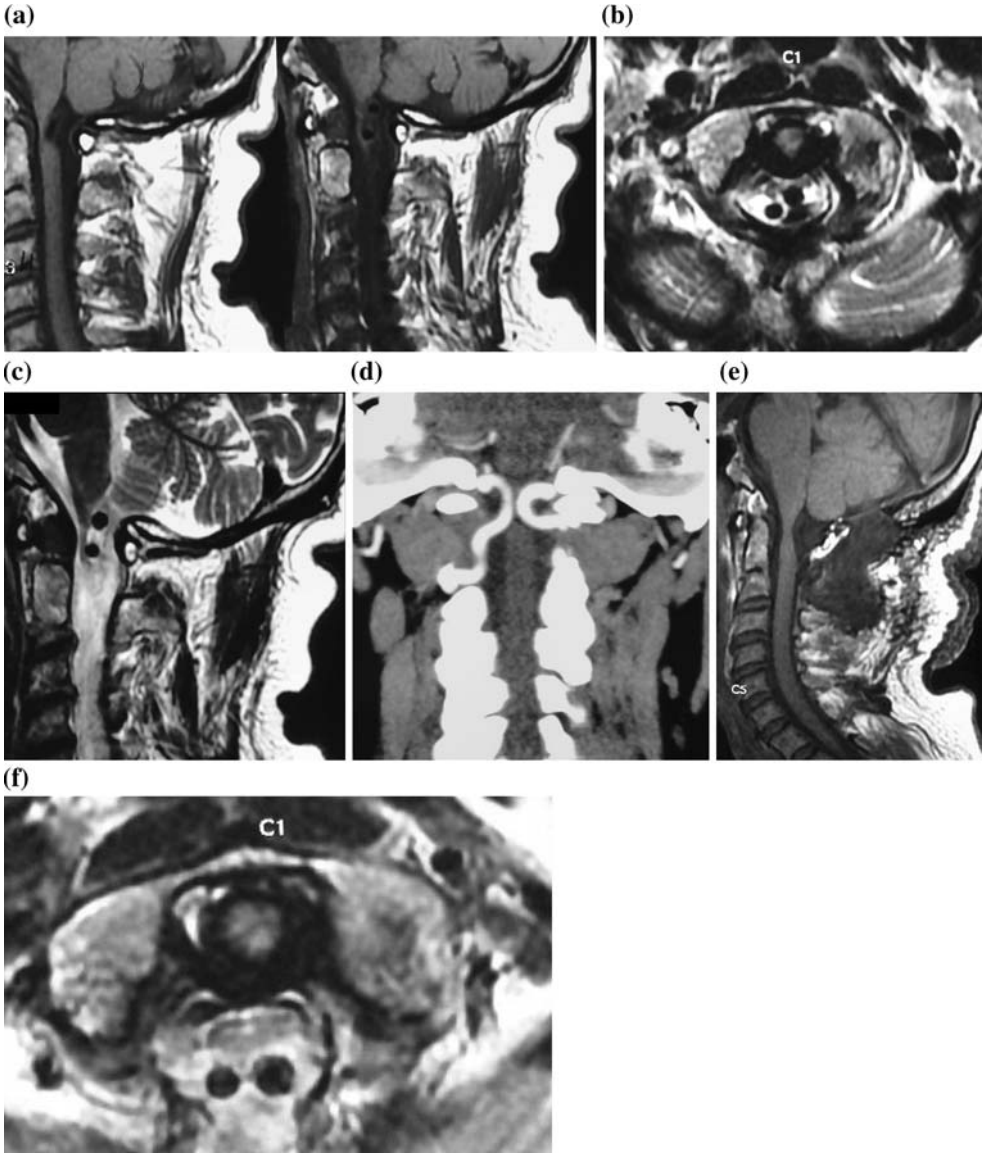

Fig. 1 **a** Pre-operative T1-weighted sagittal images of the craniovertebral junction and cervical spine showing cord compression due to anomalous vertebral artery. **b** Pre-operative T2-weighted axial image showing the abnormal intradural course of both vertebral arteries. The cord indentation caused by the left vertebral artery can be visualized. **c** T2-weighted sagittal MRI showing the cord compression by the vertebral artery. **d** CT angiography depicting the course of the vertebral arteries. The arteries entered the dura in the C1–C2 interlaminar space and then looped medially "kissing" each other on the dorsal surface of the cord. **e** Post-operative T1-weighted sagittal image showing relief of cord compression after vasculopexy of bilateral vertebral arteries. **f** Post-operative T2-weighted axial image showing the vessels distracted away from the deformed cord

were visualized coursing posteromedially between the atlas and axis over the dorsal aspect of the C2 roots. The vessels then pierced the dura mater underneath the posterior arch of the atlas. Intradurally, the vessels again looped medially 'kissing' each other over the dorsal aspect of cervicomedullary junction. The arteries traversed between the rootlets of the C2 nerve. The cervical cord at this level was indented and flattened and appeared grayish and soft. The indentation and effects on the cord appeared more severe on the left side. Microvascular techniques were used to separate the arteries from the C2 roots and from the cord. The arteries could be relatively easily lifted away from the neural tissue and the rootlets. A window was created between the neural tissue and the vessels and a Teflon sponge (size 3 mm $\times$ 4 mm $\times$ 15 mm) was passed around the arteries on each side creating a sling. Then, a non-absorbable 3-0 mersilk suture with needles on both the ends was passed through the two arms of each sling. The two ends of the suture were brought out from the dura and sutured over it. The thread was then further anchored laterally by suturing it to the adjacent muscle. This maneuver elevated the vessels away from cord without causing any

kinking or lumen compromise. The hitching also ensured that the vessels would remain separated from the cord and cervicomedullary junction underneath the dura.

Post-operative course

The patient improved in the post-operative period and made a rapid neurological recovery. At a follow-up of 14 months, the patient was able to carry out all his routine activities.

Discussion

Vascular loops and ectatic vertebro-basilar arteries are common causes of trigeminal neuralgia, hemifacial spasm, glossopharyngeal neuralgia, vertigo and/or tinnitus and spasmodic torticollis [8, 12]. Although rare, primary and secondary reductions of posterior cranial fossa volume, tumors both infratentorial and supratentorial have been reported to cause vascular compression syndromes [2, 4–6, 8]. Microvascular decompression of the affected cranial nerve has been the accepted modality of treatment in such cases. Even in cases with local or remote tumors, it has been speculated that vascular compression at the root entry zone ultimately causes the pain. Resection of the tumor with or without directly manipulating the vessel loop has been associated with lasting relief of symptoms. Ectatic and anomalous vertebral arterial loops resulting in cord compression have been identified only rarely [7, 10, 13–21]. We recently studied, on cadavers, the normal anatomy of vertebral artery in the region of craniovertebral junction [3]. The vertebral arteries run a vertical course from the transverse foramina of C3–C6. After its exit from the transverse foramen of the C3 vertebra, the V1-segment of the artery courses posterosuperiorly and forms a loop within the groove on the inferior surface of the superior articular facet, turns back inferiorly and then exits from the transverse foramina of the C2 vertebra. The extent of extension of the vertebral artery loop within the C2 facet varies. The vertebral artery exits from the transverse process of the C2 vertebra and takes an initial lateral bend and then traverses superiorly to enter the transverse foramen of C1. In its third segment, after exiting the transverse process foramen of C1, the vertebral artery takes an approximately 90° posterior bend and turns medially to engage in the groove on the superior surface of the posterior arch of the atlas, where turning around the superior facet of the atlas it bends anteriorly to enter the spinal canal in front of the posterior atlanto-occipital membrane. In its fourth segment, the artery pierces the dura mater and arachnoid between the occipital bone and atlas and enters the cranial cavity through the foramen magnum. From the reported literature, it appears that there is approximately 2–3% incidence of vertebral artery anomalies in the region of craniovertebral junction. Although rare, vertebral artery course and its dural entry below the posterior arch of atlas exists [3, 17]. In our case, the vertebral artery on both sides entered into the intradural compartment underneath the posterior arch of atlas almost symmetrically. It appeared that due to abnormal course, the extradural course of the vertebral artery was probably shortened and the extra length of the vertebral artery was present intradurally. This excess length of the artery formed a loop on the dorsal surface of the cord. Continuous pulsation of the large arteries over long period of time was apparently the cause of myelopathy.

Anomalous vertebral arteries in most instances do not cause any symptoms. From our literature survey, we look at isolated 13 cases of symptomatic cord compression due to anomalous vertebral arteries, out of which 6 had unilateral compression and 7 had bilateral compression [16, 17]. Whilst two patients had evidence of myelopathy, rest of the cases had symptoms that could be related to nerve root compression and manifested as neck pain, arm pain or torticollis [16, 17, 21]. Intermittent stimulation of the dorsal root entry zone of the cervical nerve with pulsations of the vertebral artery can explain the neck pain and occipital neuralgia. Compression of the Lissauer's tract might be the mechanism of the shoulder and arm pain [16]. Takahashi et al. [15] reported a case where the radiological and clinical features were remarkably similar to our presented case. It was suggested by Suzuki et al. [14] that the vertebral arteries entering the spinal canal at a lower level, may lead to cord compression as the intraspinal subarachnoid space at that level may be narrow. Hasegawa et al. [10] reported a patient with an abnormal course of the vertebral artery, platybasia and hypoplasia of the atlas. They postulated that the canal stenosis associated with hypoplasia of the atlas contributed to the cord compression by the vertebral artery.

MRI and 3D CT angiography are the investigations of choice to diagnose such patients. The reconstructed CT image demonstrated the course of the vertebral arteries and its bony relations obviating the need for a four vessel angiography. Treatment options for patients with anomalous vertebral arteries with root or cord compression include vessel transposition, duroplasty, detachment of roots from the vertebral arteries or the placement of inert material between the vessel and the cord. Since the vertebral arteries are large and adequate decompression may not be achieved with mere insertion of prosthetic material between the vessel and neural tissue, transposition of the vertebral arteries and vasculopexy maybe the preferred technique. Various materials like Gore-tex, silicon tapes and nylon threads have been used to lift the arteries from the cord. All the previously reported patients with bilateral

vertebral artery compression have been treated with vertebral artery transposition and duroplasty [13–21]. In one patient, an additional prosthesis was also placed between the arteries and the cord [13]. All the patients improved following surgery except the one who developed a right hemiparesis post-operatively following a frontal infarct [20]. In our patient, a Teflon sponge was inserted in the space between the vertebral artery and the cord. Since the artery was in contact with a long segment of the cord, a wider sponge was used. After the elevation, the arteries were maintained in this position using the silk sutures that passed through the arms of the Teflon sling and hitching them to the outer surface of the dura. This was further anchored to the adjoining muscles. This method allowed primary closure of the dura except for a small patch in the region of the dural hitching which was repaired with a local pedicled muscle flap. Although duroplasty and laminectomy could have been 'sufficient' and probably safer for indirect decompression of the spinal cord, the ease with which the arteries could be dissected free and elevated off the spinal cord favored an aggressive surgical strategy. Following bilateral vasculopexy, the cervicomedullary junction was relieved of compression resulting in lasting relief of the symptoms of the patient.

Conclusion

Anomalous course of vertebral arteries may be rare cause of cervicomedullary compression. Vigilant pre-operative investigations and planning may provide lasting relief from symptoms.

Conflict of interest None of the authors has any potential conflict of interest.

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