


**Bilateral Aberrant C1/2 Intradural Vertebral Arteries: a
Rare Cause of Cervical Myelopathy**

Journal:	<i>ANZ Journal of Surgery</i>
Manuscript ID:	ANS-2014-00571.R1
Manuscript Type:	Images for Surgeons
Date Submitted by the Author:	n/a
Complete List of Authors:	Tsang, Anderson; The University of Hong Kong, Surgery Tsang, Frederick; Queen Mary Hospital, Neurosurgery Lui, Wai Man; Queen Mary Hospital, Neurosurgery
General Key Words:	Neurosurgery
Specialty Key Words:	Cervical myelopathy, Vertebral artery

 SCHOLARONE™
Manuscripts
Review

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

**Bilateral Aberrant C1/2 Intradural Vertebral Arteries: a
Rare Cause of Cervical Myelopathy**

Authors:

Anderson Chun On Tsang,^{1,2} MBBS, Frederick Chun Pong Tsang,² MBBS FCSHK,
Wai Man Lui,² MBBS, FCSHK

Affiliations:

¹ Department of Surgery, Li Ka Shing Faculty of Medicine, The University of Hong
Kong, Hong Kong;
² Department of Neurosurgery, Queen Mary Hospital, Hong Kong

Corresponding author:

Dr Anderson C.O. Tsang, Division of Neurosurgery, Department of Surgery, Li Ka
Shing Faculty of Medicine, The University of Hong Kong, Queen Mary Hospital, 102
Pokfulam Road, Hong Kong. Tel: (852) 22553368; Fax: (852) 28184350; Email:
acotsang@hku.hk.

Running Title:

Cervical myelopathy due to compression of aberrant vertebral arteries

The corresponding author is not a recipient of a research scholarship.

The content of this paper is never published or presented in any medical conference.

3 figures are included in this paper.

Word count: 732 (Text)

For Peer Review

A 65-year-old lady presented with progressive cervical myelopathic symptoms for 4 years. She complained of bilateral upper limbs numbness and weakness initially, associated with clumsiness of fine hand movements. The motor power of both upper limbs gradually deteriorated to grade 4 out of 5. Her symptoms then progressed to the lower limbs with stiff and clumsy gait in recent two months. There was no sphincter disturbance or neck pain. Physical examination revealed bilateral brisk tendon reflexes, positive Hoffman's sign and finger escape on both hands, confirming the clinical diagnosis of cervical myelopathy.

MRI of the cervical spine showed prominent vessel loops compressing the cervical cord bilaterally at the cranio-cervical junction. (Fig. 1) The bony alignment was normal and there were no significant degenerative changes or other cord-compressing lesions. CT-angiogram of the head and neck region confirmed the culprit vessels were the co-dominant vertebral arteries (VA) with an aberrant course. Normally, both VA exit the foramen transversarium of C1, then curve medially along the superior aspect of the posterior arch of C1, before entering the posterior ligaments and the dura at the foramen magnum to continue the intracranial course. In our patient, both vertebral arteries pierced the dura between the axis and the atlas, after exiting the foramen transversarium of C2 without going through the C1 foramen transversarium.

This gave rise to an aberrant intraspinal intradural course from the level of the atlas, causing compressive cervical myelopathy. (Fig. 2)

Limited suboccipital craniectomy with C1 laminectomy was performed for decompression. The spinal cord showed indentations by both VA, (Fig. 3) which were mobilized free from the cord and anchored. The thecal sac was enlarged for better decompression. The somatosensory evoked potential of all four limbs improved intra-operatively.

Patient's symptoms improved spontaneously after the operation. Post-operative MRI showed partial re-expansion of the previously compressed C1 cord, with both VAs now displaced laterally and not in contact with the cord. At 6 months after decompression, she can walk independently, with full motor power in all four limbs. The numbness and tightness of the extremities also resolved except over the left arm, where the symptoms first appeared. She was able to return to work without limitations

Anatomical variations of the distal VAs are well documented but rarely symptomatic. Only four symptomatic cases of VA entering dura between C1/2 levels had been reported in the literature.¹⁻⁴ This variation is due to persistence of the spinal branch of

1
2
3
4
5
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

the embryological Type 2 Proatlantal artery, which normally regresses during VA development. Such variation could result in duplication or aberrant course of distal VA, and is first recognized by Lasjaunias et al.⁵ In case of duplication, the aberrant C1/2 entrance of VA may co-exist with a normal foramen of magnum entering branch. In our patient, the C1/2 branch was dominant while the typical course of VA was absent, resulting in an aberrant course of distal VA. Other anatomical variations that can occur in the distal VA include C1 or C2 origin of the Posterior inferior cerebellar artery.⁶

Our patient became symptomatic at 60 years of age, similar to previous reported cases when symptom onset was from 50 to 70 years old.¹⁻⁴ It is unclear why such congenital anatomical variation should become symptomatic only in the late adulthood. We surmise it may be due to chronic pulsatile stimulation of the spinal cord by the VAs, coupled with a progressive narrowing of spinal canal secondary to degeneration, leading to myelopathy eventually.

Microsurgical release of the compressing VA together with decompressive laminectomy is an effective treatment. We used autologous fascia graft harvested from the cervical muscles as a sling to secure both VAs away from the cord surface.

Synthetic materials such as Teflon sponge has also been used to separate the VA from the cord.^{2,3} Although that served to buffer the pulsatile stimulation from the vessel, the cord was still physically in contact and compressed by the VA and sponge. We believed anchoring the VA and releasing it from contact of the cord would provide more lasting symptomatic relief. Improvements in the intraoperative somato-sensory evoked potential monitoring and expansion of the previously compressed cord in the post-operative MRI confirmed satisfactory decompression.

In conclusion, we presented a rare case of cervical myelopathy due to bilateral vertebral arteries compression at C1 level, secondary to the persistence of intradural Type 2 proatlantal artery. Laminectomy and microvascular decompression with autologous fascia graft resulted in resolution of symptoms.

References:

1. Ball BG, Krueger BR, Piepgras DG. Anomalous vertebral artery compression of the spinal cord at the cervicomedullary junction. *Surgical neurology international* 2011;2:103.
2. Ha EJ, Lee SE, Jahng TA, Kim HJ. Cervical Compressive Myelopathy due to Anomalous Bilateral Vertebral Artery. *Journal of Korean Neurosurgical Society* 2013;54:347-9.
3. Shah A, Mahore A, Goel A. Bilateral vasculopathy of anomalous vertebral arteries causing cervicomedullary compression: case report and technical note. *European*

spine journal : official publication of the European Spine Society, the European Spinal Deformity Society, and the European Section of the Cervical Spine Research Society 2012;21 Suppl 4:S505-8.

4. Takei H, Sagae M, Chiba K, Ogino T. The long-term follow-up of surgical treatment for cervical myelopathy with severe nape and upper arm pain caused by the anomalous vertebral artery: case report. *Spine* 2008;33:E611-3.
5. Lasjaunias P, Vallee B, Person H, Ter Brugge K, Chiu M. The lateral spinal artery of the upper cervical spinal cord. Anatomy, normal variations, and angiographic aspects. *J Neurosurg* 1985;63:235-41.
6. Siclari F, Burger IM, Fasel JH, Gailloud P. Developmental anatomy of the distal vertebral artery in relationship to variants of the posterior and lateral spinal arterial systems. *AJNR Am J Neuroradiol* 2007;28:1185-90.

Figure Legends

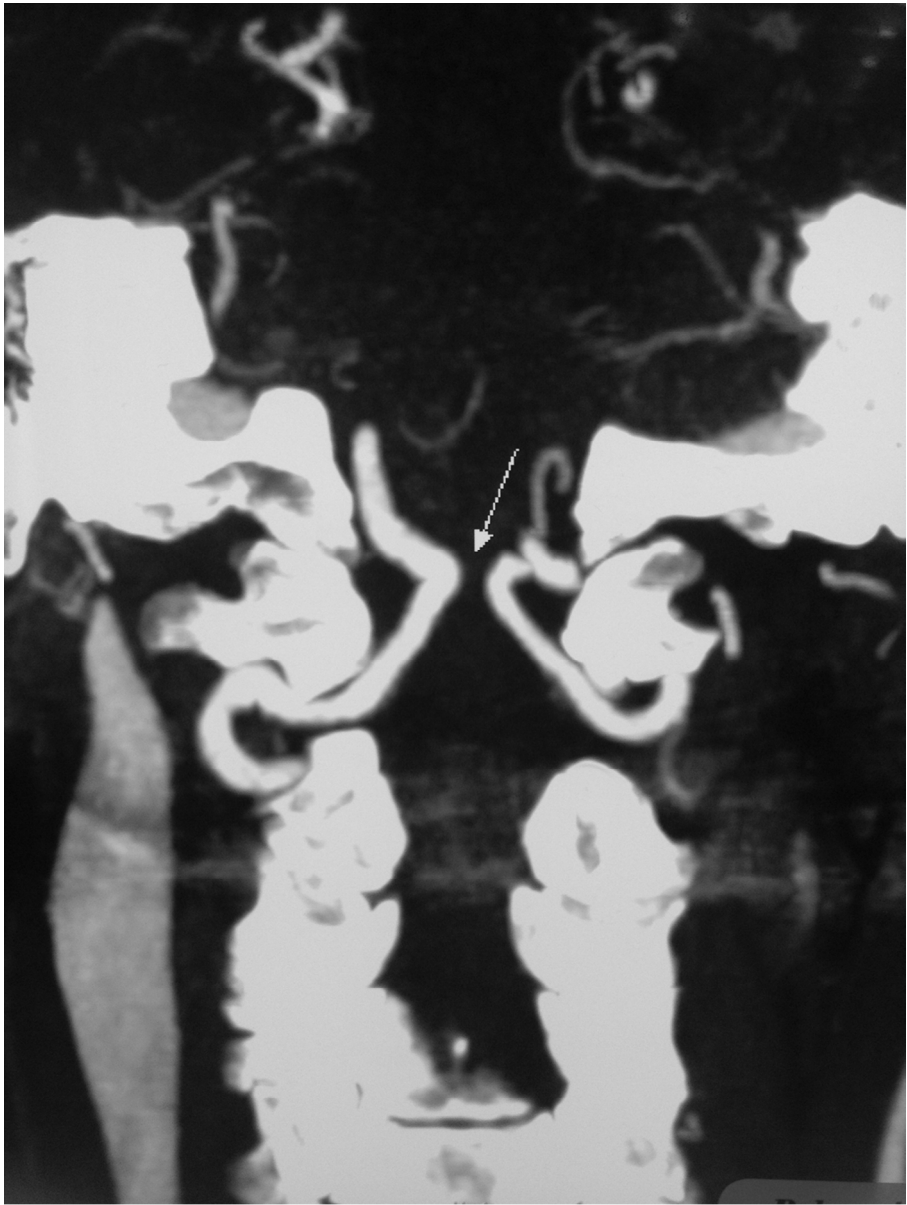
Fig. 1: Sagittal T2 MRI of the cervical spine, showing aberrant vessel loop compressing spinal cord at C1 level.

Fig. 2: Coronal reconstruction of CT angiogram of the neck, showing bilateral aberrant vertebral arteries entering the spinal canal between C1 and C2, with compression of the cord.

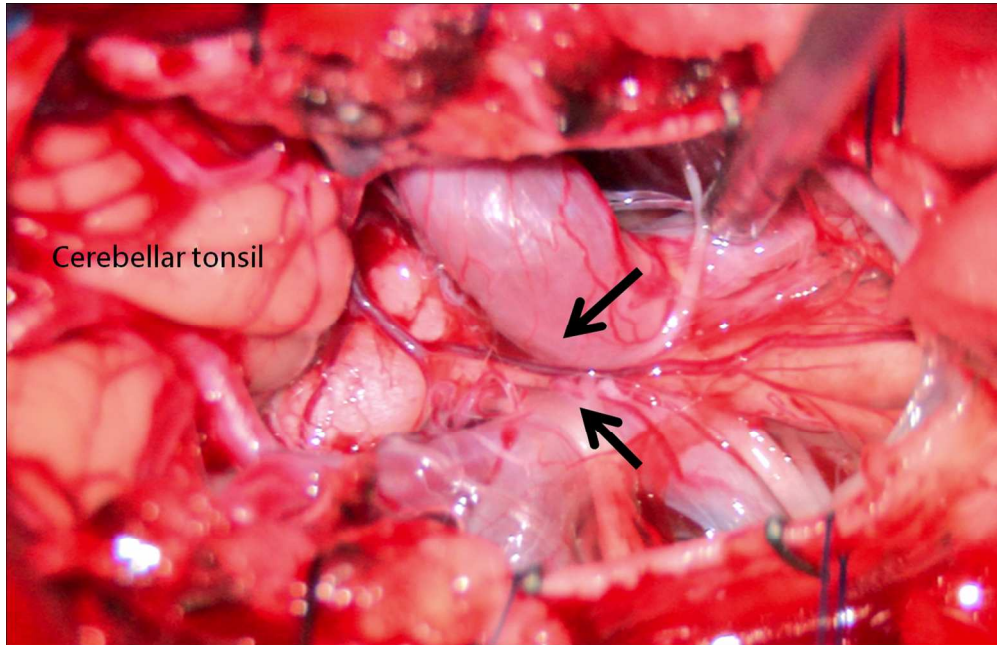
Fig. 3: Intraoperative view under operating microscope showing bilateral aberrant vertebral arteries compressing the cord.



Sagittal T2 MRI of the cervical spine, showing aberrant vessel loop compressing spinal cord at C1 level.
101x119mm (300 x 300 DPI)



Coronal reconstruction of CT angiogram of the neck, showing bilateral aberrant vertebral arteries entering the spinal canal between C1 and C2, with compression of the cord.
101x135mm (300 x 300 DPI)



Intraoperative view under operating microscope showing bilateral aberrant vertebral arteries (arrow) compressing the cord.
177x114mm (300 x 300 DPI)

Review