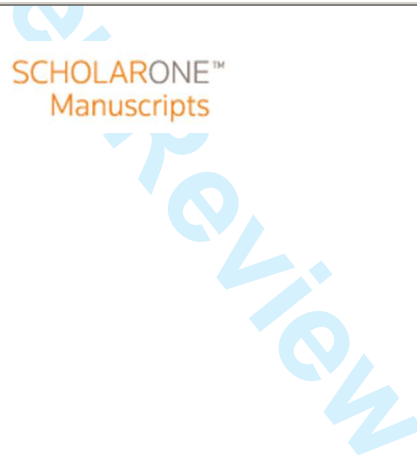




<b>Title</b>	<b>Bilateral aberrant C1/2 intradural vertebral arteries: a rare cause of cervical myelopathy</b>
<b>Author(s)</b>	<b>Tsang, ACO; Tsang, FCP; Lui, WM</b>
<b>Citation</b>	<b>ANZ Journal of Surgery, 2017, v. 87 n. 3, p. 202-203</b>
<b>Issued Date</b>	<b>2017</b>
<b>URL</b>	<b><a href="http://hdl.handle.net/10722/207766">http://hdl.handle.net/10722/207766</a></b>
<b>Rights</b>	<b>This is the accepted version of the following article: ANZ Journal of Surgery, 2017, v. 87 n. 3, p. 202-203, which has been published in final form at <a href="http://onlinelibrary.wiley.com/doi/10.1111/ans.12870/abstract">http://onlinelibrary.wiley.com/doi/10.1111/ans.12870/abstract</a>; This work is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License.</b>

**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



1  
2  
3  
4 **Bilateral Aberrant C1/2 Intradural Vertebral Arteries: a**  
5  
6  
7 **Rare Cause of Cervical Myelopathy**  
8  
9

10  
11  
12 **Authors:**

13  
14  
15 Anderson Chun On Tsang,<sup>1,2</sup> MBBS, Frederick Chun Pong Tsang,<sup>2</sup> MBBS FCSHK,  
16

17  
18 Wai Man Lui,<sup>2</sup> MBBS, FCSHK  
19

20  
21  
22  
23  
24 **Affiliations:**

25  
26  
27 <sup>1</sup> Department of Surgery, Li Ka Shing Faculty of Medicine, The University of Hong  
28  
29 Kong, Hong Kong;

30  
31 <sup>2</sup> Department of Neurosurgery, Queen Mary Hospital, Hong Kong  
32  
33  
34  
35  
36  
37

38  
39  
40  
41 **Corresponding author:**

42 Dr Anderson C.O. Tsang, Division of Neurosurgery, Department of Surgery, Li Ka  
43  
44 Shing Faculty of Medicine, The University of Hong Kong, Queen Mary Hospital, 102  
45  
46 Pokfulam Road, Hong Kong. Tel: (852) 22553368; Fax: (852) 28184350; Email:  
47  
48 acotsang@hku.hk.  
49  
50  
51  
52  
53

54 **Running Title:**

55  
56  
57 Cervical myelopathy due to compression of aberrant vertebral arteries  
58  
59  
60

1  
2  
3  
4  
5  
6  
7 The corresponding author is not a recipient of a research scholarship.  
8

9  
10 The content of this paper is never published or presented in any medical conference.  
11

12  
13 3 figures are included in this paper.  
14

15  
16 Word count: 732 (Text)  
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

For Peer Review

1  
2  
3  
4 A 65-year-old lady presented with progressive cervical myelopathic symptoms  
5  
6  
7 for 4 years. She complained of bilateral upper limbs numbness and weakness initially,  
8  
9  
10 associated with clumsiness of fine hand movements. The motor power of both upper  
11  
12 limbs gradually deteriorated to grade 4 out of 5. Her symptoms then progressed to the  
13  
14 lower limbs with stiff and clumsy gait in recent two months. There was no sphincter  
15  
16 disturbance or neck pain. Physical examination revealed bilateral brisk tendon  
17  
18 reflexes, positive Hoffman's sign and finger escape on both hands, confirming the  
19  
20 clinical diagnosis of cervical myelopathy.  
21  
22  
23  
24  
25  
26  
27  
28  
29

30 MRI of the cervical spine showed prominent vessel loops compressing the  
31  
32 cervical cord bilaterally at the cranio-cervical junction. (Fig. 1) The bony alignment  
33  
34 was normal and there were no significant degenerative changes or other  
35  
36 cord-compressing lesions. CT-angiogram of the head and neck region confirmed the  
37  
38 culprit vessels were the co-dominant vertebral arteries (VA) with an aberrant course.  
39  
40  
41 Normally, both VA exit the foramen transversarium of C1, then curve medially along  
42  
43 the superior aspect of the posterior arch of C1, before entering the posterior ligaments  
44  
45 and the dura at the foramen magnum to continue the intracranial course. In our patient,  
46  
47 both vertebral arteries pierced the dura between the axis and the atlas, after exiting the  
48  
49 foramen transversarium of C2 without going through the C1 foramen transversarium.  
50  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

1  
2  
3  
4 This gave rise to an aberrant intraspinal intradural course from the level of the atlas,  
5  
6  
7 causing compressive cervical myelopathy. (Fig. 2)  
8  
9

10  
11  
12 Limited suboccipital craniectomy with C1 laminectomy was performed for  
13  
14 decompression. The spinal cord showed indentations by both VA, (Fig. 3) which were  
15  
16 mobilized free from the cord and anchored. The thecal sac was enlarged for better  
17  
18 decompression. The somatosensory evoked potential of all four limbs improved  
19  
20  
21 intra-operatively.  
22  
23  
24  
25  
26  
27  
28  
29

30 Patient's symptoms improved spontaneously after the operation. Post-operative  
31  
32 MRI showed partial re-expansion of the previously compressed C1 cord, with both  
33  
34 VAs now displaced laterally and not in contact with the cord. At 6 months after  
35  
36 decompression, she can walk independently, with full motor power in all four limbs.  
37  
38  
39 The numbness and tightness of the extremities also resolved except over the left arm,  
40  
41  
42 where the symptoms first appeared. She was able to return to work without limitations  
43  
44  
45  
46  
47  
48  
49

50 Anatomical variations of the distal VAs are well documented but rarely symptomatic.  
51  
52  
53 Only four symptomatic cases of VA entering dura between C1/2 levels had been  
54  
55  
56 reported in the literature.<sup>1-4</sup> This variation is due to persistence of the spinal branch of  
57  
58  
59  
60

1  
2  
3  
4 the embryological Type 2 Proatlantal artery, which normally regresses during VA  
5  
6  
7 development. Such variation could result in duplication or aberrant course of distal  
8  
9  
10 VA, and is first recognized by Lasjaunias et al.<sup>5</sup> In case of duplication, the aberrant  
11  
12  
13 C1/2 entrance of VA may co-exist with a normal foramen of magnum entering branch.  
14  
15  
16 In our patient, the C1/2 branch was dominant while the typical course of VA was  
17  
18  
19 absent, resulting in an aberrant course of distal VA. Other anatomical variations that  
20  
21  
22 can occur in the distal VA include C1 or C2 origin of the Posterior inferior cerebellar  
23  
24  
25 artery.<sup>6</sup>  
26  
27  
28  
29

30 Our patient became symptomatic at 60 years of age, similar to previous reported cases  
31  
32  
33 when symptom onset was from 50 to 70 years old.<sup>1-4</sup> It is unclear why such  
34  
35  
36 congenital anatomical variation should become symptomatic only in the late  
37  
38  
39 adulthood. We surmise it may be due to chronic pulsatile stimulation of the spinal  
40  
41  
42 cord by the VAs, coupled with a progressive narrowing of spinal canal secondary to  
43  
44  
45 degeneration, leading to myelopathy eventually.  
46  
47  
48  
49

50 Microsurgical release of the compressing VA together with decompressive  
51  
52  
53 laminectomy is an effective treatment. We used autologous fascia graft harvested from  
54  
55  
56 the cervical muscles as a sling to secure both VAs away from the cord surface.  
57  
58  
59  
60

1  
2  
3  
4 Synthetic materials such as Teflon sponge has also been used to separate the VA from  
5  
6 the cord.<sup>2,3</sup> Although that served to buffer the pulsatile stimulation from the vessel, the  
7  
8 cord was still physically in contact and compressed by the VA and sponge. We  
9  
10 believed anchoring the VA and releasing it from contact of the cord would provide  
11  
12 more lasting symptomatic relief. Improvements in the intraoperative somato-sensory  
13  
14 evoked potential monitoring and expansion of the previously compressed cord in the  
15  
16 post-operative MRI confirmed satisfactory decompression.  
17  
18  
19  
20  
21  
22  
23  
24  
25  
26

27 In conclusion, we presented a rare case of cervical myelopathy due to bilateral  
28  
29 vertebral arteries compression at C1 level, secondary to the persistence of intradural  
30  
31 Type 2 proatlantal artery. Laminectomy and microvascular decompression with  
32  
33 autologous fascia graft resulted in resolution of symptoms.  
34  
35  
36  
37  
38  
39  
40  
41

#### 42 **References:**

- 43  
44  
45  
46  
47 1. Ball BG, Krueger BR, Piepgras DG. Anomalous vertebral artery compression of  
48 the spinal cord at the cervicomedullary junction. *Surgical neurology international*  
49 2011;2:103.  
50  
51 2. Ha EJ, Lee SE, Jahng TA, Kim HJ. Cervical Compressive Myelopathy due to  
52 Anomalous Bilateral Vertebral Artery. *Journal of Korean Neurosurgical Society*  
53 2013;54:347-9.  
54  
55 3. Shah A, Mahore A, Goel A. Bilateral vasculopathy of anomalous vertebral arteries  
56 causing cervicomedullary compression: case report and technical note. *European*  
57  
58  
59  
60



1  
2  
3 spine journal : official publication of the European Spine Society, the European Spinal  
4 Deformity Society, and the European Section of the Cervical Spine Research Society  
5 2012;21 Suppl 4:S505-8.

6  
7 4. Takei H, Sagae M, Chiba K, Ogino T. The long-term follow-up of surgical  
8 treatment for cervical myelopathy with severe nape and upper arm pain caused by  
9 the anomalous vertebral artery: case report. Spine 2008;33:E611-3.

10  
11 5. Lasjaunias P, Vallee B, Person H, Ter Brugge K, Chiu M. The lateral spinal artery of  
12 the upper cervical spinal cord. Anatomy, normal variations, and angiographic aspects.  
13 J Neurosurg 1985;63:235-41.

14  
15 6. Siclari F, Burger IM, Fasel JH, Gailloud P. Developmental anatomy of the distal  
16 vertebral artery in relationship to variants of the posterior and lateral spinal arterial  
17 systems. AJNR Am J Neuroradiol 2007;28:1185-90.  
18  
19  
20  
21  
22

### 23 24 **Figure Legends**

25  
26  
27 Fig. 1: Sagittal T2 MRI of the cervical spine, showing aberrant vessel loop  
28  
29  
30 compressing spinal cord at C1 level.  
31

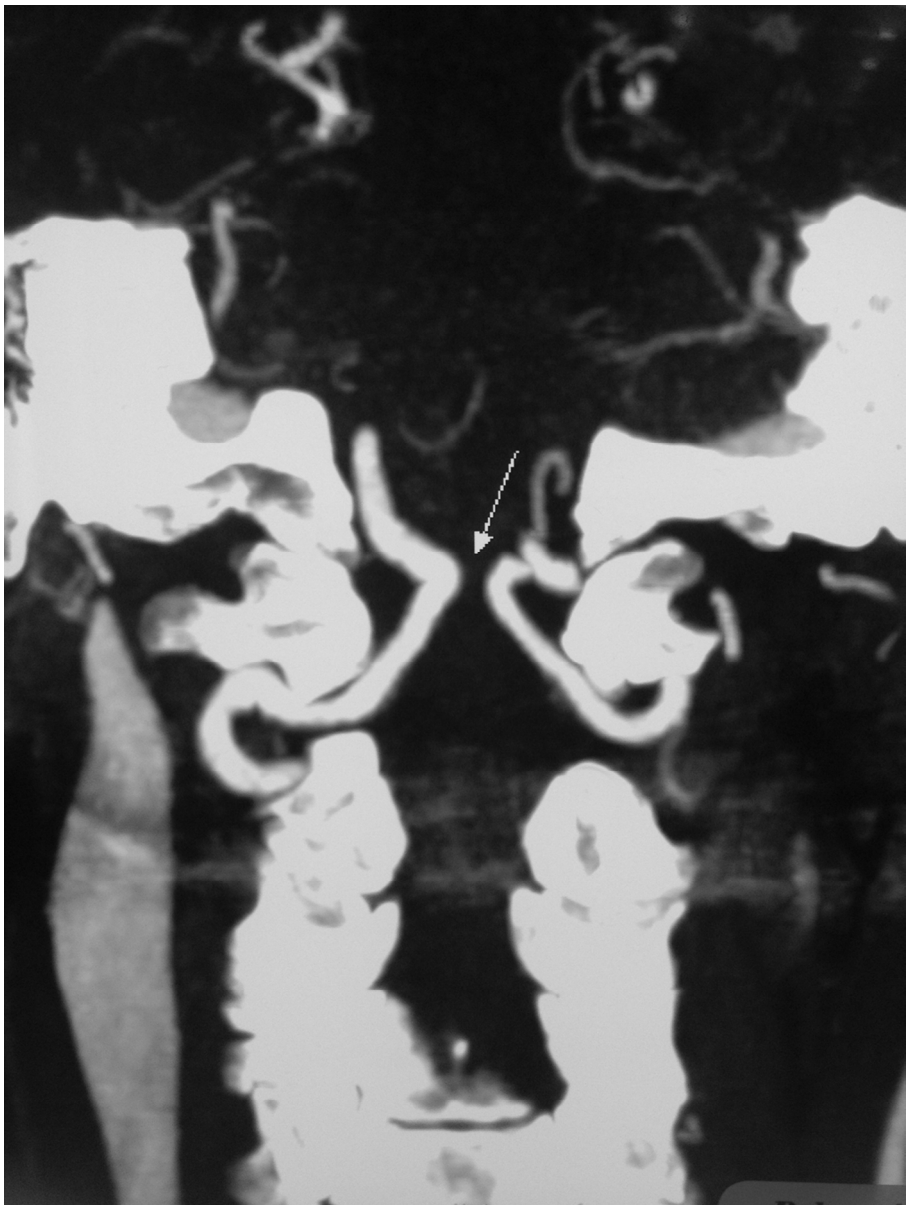
32  
33  
34  
35  
36 Fig. 2: Coronal reconstruction of CT angiogram of the neck, showing bilateral  
37  
38  
39 aberrant vertebral arteries entering the spinal canal between C1 and C2, with  
40  
41  
42 compression of the cord.  
43

44  
45  
46  
47 Fig. 3: Intraoperative view under operating microscope showing bilateral aberrant  
48  
49  
50 vertebral arteries compressing the cord.  
51  
52  
53  
54  
55  
56  
57  
58  
59  
60

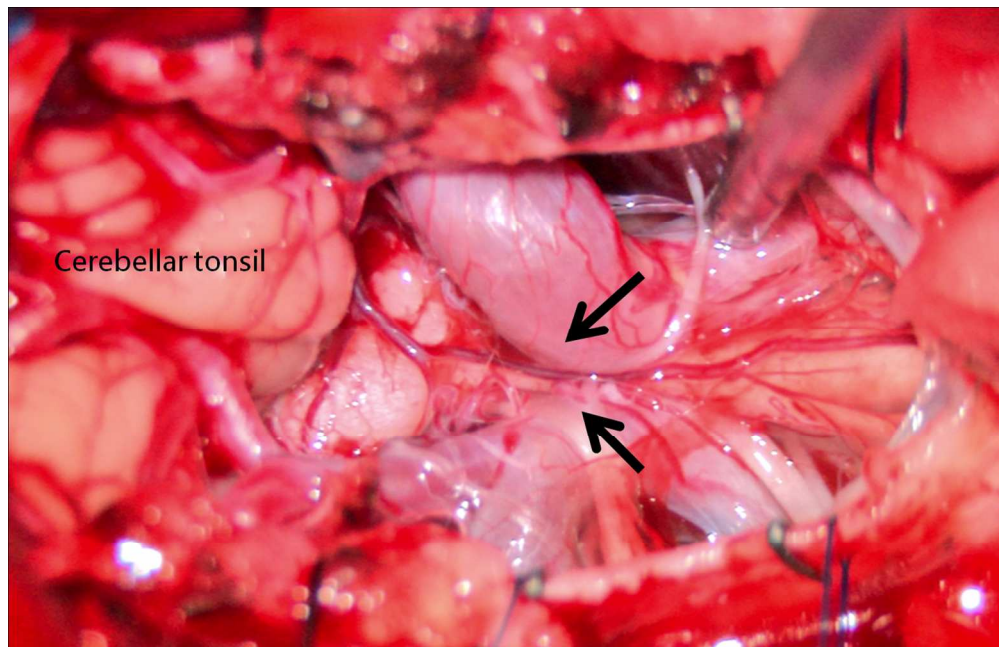


Sagittal T2 MRI of the cervical spine, showing aberrant vessel loop compressing spinal cord at C1 level.  
101x119mm (300 x 300 DPI)

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



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)



28  
29 Intraoperative view under operating microscope showing bilateral aberrant vertebral arteries (arrow)  
30 compressing the cord.  
31 177x114mm (300 x 300 DPI)

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

Review