



Case Report

Endovascular management of iatrogenic vertebro-vertebral fistula: Black Swan event in C2 pedicle screw

Jitender Chaturvedi¹, P. Venkata Sudhakar², Mohit Gupta¹, Nishant Goyal¹, Shiv Kumar Mudgal³, Priyanka Gupta⁴, Sandeep Burathoki⁵

Departments of ¹Neurosurgery and ²Orthopedic Surgery, All India Institute of Medical Sciences, Rishikesh, Uttarakhand, ³College of Nursing, All India Institute of Medical Sciences, Deoghar, Jharkhand, Departments of ⁴Neuroanesthesia and ⁵Neurointervention, All India Institute of Medical Sciences, Rishikesh, Uttarakhand, India.

E-mail: *Jitender Chaturvedi - drjittu28@gmail.com; P. Venkata Sudhakar - torpedo89@gmail.com; Mohit Gupta - drmohitguptams@yahoo.com; Nishant Goyal - drnishantgoyal@gmail.com; Shiv Kumar Mudgal - peehupari05@gmail.com; Priyanka Gupta - drpriyankagupta84@gmail.com; Sandeep Burathoki - skburathoki@yahoo.co.in



*Corresponding author:

Jitender Chaturvedi,
Department of Neurosurgery,
All India Institute of
Medical Sciences, Rishikesh,
Uttarakhand, India.

drjittu28@gmail.com

Received : 17 March 2022

Accepted : 13 April 2022

Published : 06 May 2022

DOI

10.25259/SNI_261_2022

Quick Response Code:



ABSTRACT

Background: Vertebro-vertebral fistulas (VVF) are rare. Anatomically, they consist of an arteriovenous fistula, a direct pathological communication between vertebral veins (including the epidural vertebral venous plexus) and extradural vertebral artery. The various etiologies include trauma, iatrogenic, or spontaneous (e.g., NF-1 or Ehlers Danlos Syndrome). The clinical presentation may include acute/delayed onset of radiculopathy and/or myelopathy. They may further be characterized by the delayed onset hearing loss to tinnitus and/or the sensation of *water in the ear*.

Case Description: We report successful endovascular management for iatrogenic VVF in a 37-year-old female who was diagnosed with an odontoid fracture (Anderson type IIC). She underwent a posterior C1 lateral masses to C2 pedicle/laminar screw fixation. An intraoperative vertebro-vertebral fistulas (VVF) was recognized during the procedure and it was managed successfully with percutaneous transarterial endovascular coiling.

Conclusion: Iatrogenic VVF should immediately be suspected when the implant trajectory goes slightly off track during a C1-2 fixation. Immediate postoperative DSA and MRI are advisable, irrespective of whether the patient is symptomatic. These lesions are best managed with endovascular coiling with or without detachable balloons.

Keywords: Endovascular coiling, Iatrogenic, Odontoid fracture, Vertebro-vertebral fistula, Black Swan

INTRODUCTION

Vertebro-vertebral arteriovenous fistulas are rare. They consist of a high flow shunt between the extracranial vertebral artery, its muscular or radicular branches, and the adjacent vertebral venous plexuses. The etiology of these vascular lesions can be traumatic, iatrogenic, or spontaneous.^[2,7,8] These lesions present as cerebral ischemia, vertebrobasilar insufficiency, cervical myelopathy and/or radiculopathy, pulsatile tinnitus, or a neck mass.^[1,3,4] Both surgical and endovascular techniques have been utilized to manage VVF. Here, we report a patient who underwent a C1-2 fixation for Type II C2 fracture and developed an iatrogenic VVF successfully treated with endovascular coiling.

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CASE REPORT

Clinical presentation with quadriplegia

A 37-year-old female presented with a progressive quadriplegia and hypoxia following a road traffic accident. Following emergent intubation, she was found to have 0/5 motor function on the right and 2/5 function on the left in the C5-T1 distributions and 2/5 function in the right lower extremity, i.e., L2-S1 myotomes.

Computed tomography (CT)/MR presentation of odontoid Type II fracture

The CT studies showed an anteriorly displaced odontoid fracture through the base of dens (Anderson D'Alonzo type IIC) on CT scan [Figure 1]. The MR revealed a retroflexed odontoid narrowing the foramen magnum and upper cervical canal with compression of the cord at the cervicomedullary junction.

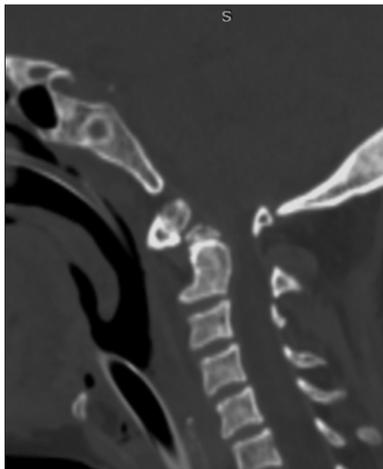


Figure 1: Preoperative sagittal CT scan showing the C2 vertebral body fracture II C.

Surgery

She underwent excision of the C1 posterior arch with the placement of bilateral C1 lateral mass screws and placement of bilateral C2 pedicle screws. However, after satisfactorily placing the right-sided C2 pedicle screw into position, the left-sided pedicle resulted in unexpected brisk nonpulsatile and dark venous bleeding from the entry point; notably, it stopped immediately after the application of wax. As a rescue procedure, the final construct included bilateral C1 lateral mass screws, a right C2 pedicle screw supplemented with a right C2 laminar screws, followed by rod fixation.

Postoperative course

Immediate postoperatively, the CT CV junction [Figures 2 and 3] showed satisfactorily placed both C1 lateral mass screws and the right-sided C2-pedicle screw with right-sided C2 laminar screw. The MRI brain [Figure 4] ruled out any Infarct or hematoma. CT angiography revealed normal V1, V2, V3, and V4 segments on the right side with attenuation of the V2 segment of the left vertebral artery. A digital subtraction angiography (DSA) confirmed the left-sided vertebro-vertebral arteriovenous fistula (VVF). Specifically, the DSA showed small pseudoaneurysm sac at the superomedial aspect of the V2 segment of the left vertebral artery, with a high flow arteriovenous shunt with early filling and drainage through the cervical epidural venous plexus [Figures 5a and b]. The patient was maintained on a ventilator over the next 3–4 weeks, during which time, she demonstrated motor recovery to 3/5 function. At this point, a neurointerventionist performed a coiling of the fistula. The post procedure angiogram confirmed complete occlusion of vertebra-vertebral fistula, as seen in [Figures 5c and d]. Over a period of another month, she improved to be taken out of ventilator with power in all four limbs to the level of 3/5 on MRC grade.

DISCUSSION

Vertebro-vertebral fistula (VVF) is a pathological communication between vertebral artery or its muscular/

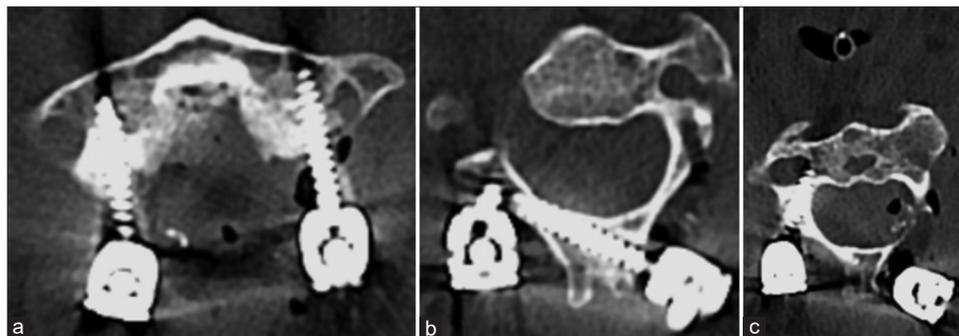


Figure 2: (a) Postoperative CT scan axial cut at the level of C1 showing bilateral lateral mass screws and (b) axial CT cuts at the level of C2 showing right laminar screw. Tract made for pedicle screw insertion on the left side is also appreciated (responsible for fistula formation). (c) Right pedicle screw at C2.

Table 1: Literature review on iatrogenic VVF.

S. No.	Authors and year	n	Level and number of Iatrogenic VVF	Etiology of VVF	Treatment
1.	Alijobebeh <i>et al.</i> , 2018	280 (Only 226 VVFs had their fistula location identified)	<u>C1-C2=11</u> C2=5 C3=3 C4=4 C5=15 C6=14 C7=3 C2-C6=2	Iatrogenic=68 (Spontaneous=136 Traumatic=76)	Endovascular constructive=11 Endovascular destructive=39 Surgery=11 None=7
2.	Briganti <i>et al.</i> , 2013	3	C2-C3=2 C3-C4=1	Traumatic, C2-6 Fracture=1 Spontaneous (Post Chiropractic Manipulation)=1 Spontaneous (NF-1)=1	All three patients underwent detachable coil occlusion of fistula and parent VA, i.e., Endovascular destructive
3.	He <i>et al.</i> , 2021	2	C4=1 C1=1	Post Acupuncture	First Patient: Endovascular destructive, i.e., Coiling using six interlocking detachable coils with complete occlusion of VA and fistula. Second Patient: Surgical ligation of VA above and below fistula Endovascular destructive.
4.	Chaturvedi <i>et al.</i> , 2022	1	<u>C1-2</u>	Postsurgical	Endovascular destructive.

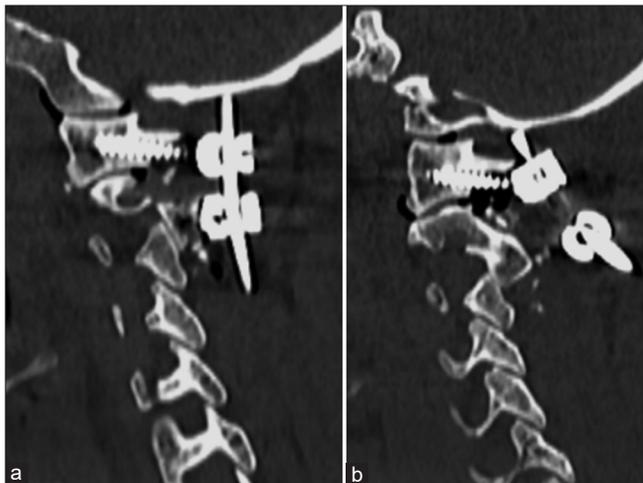


Figure 3: Sagittal CT scan showing the position of the screws within C1 vertebral body on the right (a) and left side (b), respectively.

radicular branches and venous plexus.^[8] Commonly, patients with VVF irrespective of etiology suffer from pulsatile tinnitus or sensations of “rushing” through the ear with feel of *water in ear*. Radiculopathy^[9] or myelopathy^[10] secondary to compression by dilated epidural venous plexus has also been reported. C2 pedicle screw placement is a common

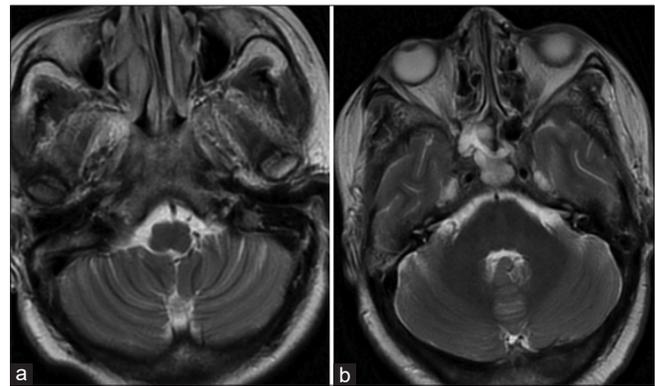


Figure 4: (a and b) Postoperative MRI scans with axial sections at the level of cerebellum showing no evidence of infarction following the injury to the left-sided vertebral artery.

neurosurgical maneuver at the CV junction for various pathologies; in this case, we had a patient undergoing a C1-C2 posterior fusion to address a Type II odontoid fracture. However, pedicle-screw breaches may occur resulting in VVF. We found 11 iatrogenic VVF occurring at the C1-2 level in the literature and have added our case to this mix, as summarized in [Table 1]. In our patient, a

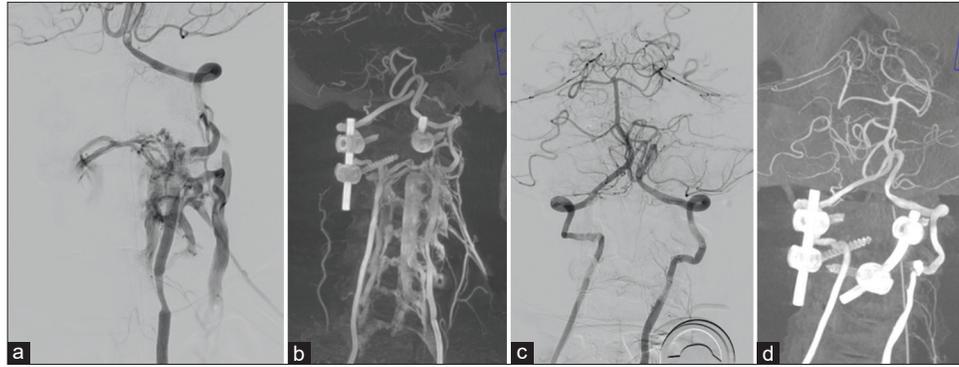


Figure 5: Left vertebral artery injection (a) and coronal reformatted image (b) showed vertebra-vertebral fistula at V2 segment with a filling of vertebral venous plexus (arrow) and intervertebral venous plexus (arrowhead). Endovascular treatment was performed using standard tri-coaxial technique, 6 F guide catheter (Envoy, Codman, US) was placed at proximal left vertebral artery. Then, microcatheter (Headway 17, Microvention, US) over microwire (Traxcess 14, Microvention, US) was selectively placed at fistulous junction. Three electro-detachable coils (Cosmos 4 mm × 12 cm, helical 3 mm × 6 cm, helical 2.5 mm × 6 cm, Microvention, US) were deployed at fistulous junction under balloon (Scepter C 4 mm × 15 mm, Microvention, US) protection of the left vertebral artery. Final angiogram showed complete occlusion of vertebra-vertebral fistula. Post coiling left vertebral artery injection (c) and reformatted image (d) showed complete closure of the fistula.

neurointerventionist successfully coiled the VVF. In fact, endovascular coiling has become the primary means of treating VVF, followed, in succession, by the application of covered stents and/or vessel reconstructions.^[3-6,10]

CONCLUSION

An iatrogenic VVF should be immediately suspected if screw placement goes off track during a C1-2 fixation. In these cases, an immediate postoperative CT, MRI, and DSA should be performed to rule out the presence of a VVF that may then be emergently coiled by a neurointerventionist.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Chaturvedi J, Sudhakar PV, Gupta M, Goyal N, Mudgal SK, Gupta P, *et al.* Endovascular management of iatrogenic vertebro-vertebral fistula: Black Swan event in C2 pedicle screw. *Surg Neurol Int* 2022;13:189.