Case Report

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

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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. These lesions present as cerebral ischemia, vertebrobasilar insufficiency, cervical myelopathy and/or radiculopathy, pulsatile tinnitus, or a neck mass. 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.
CASE REPORT

Clinical presentation with quadriparesis

A 37-year-old female presented with a progressive quadriparesis 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 leg, 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.

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 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.

Table 1: Literature review on iatrogenic VVF.

| S. No. | Authors and year     | n     | Level and number of iatrogenic VVF | Etiology of VVF                                                                 | Treatment                                |
|--------|----------------------|-------|------------------------------------|--------------------------------------------------------------------------------|------------------------------------------|
| 1.     | Alijobeh et al., 2018| 280   | C1-C2=11                           | Iatrogenic=68 (Spontaneous=136 Traumatic=76)                                  | Endovascular constructive=11 Endovascular destructive=39 Surgery=11 None=7         |
|        | (Only 226 VVFs had their fistula location identified) |       | C2=5                               |                                                                                 |                                          |
|        |                      |       | C3=3                               |                                                                                 |                                          |
|        |                      |       | C4=4                               |                                                                                 |                                          |
|        |                      |       | C5=15                              |                                                                                 |                                          |
|        |                      |       | C6=14                              |                                                                                 |                                          |
|        |                      |       | C7=3                               |                                                                                 |                                          |
|        |                      |       | C2-C6=2                            |                                                                                 |                                          |
| 2.     | Briganti et al., 2013| 3     | C2-C3=2                            | 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 |
|        |                      |       | C3-C4=1                            |                                                                                 |                                          |
| 3.     | He et al., 2021      | 2     | C4=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 |
|        |                      |       | C1=1                               |                                                                                 |                                          |
| 4.     | Chaturvedi et al., 2022| 1     | C1-2                               | Postsurgical                                                                     | Endovascular destructive.                |

radicular branches and venous plexus. 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 or myelopathy secondary to compression by dilated epidural venous plexus has also been reported. C2 pedicle screw placement is a common 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
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.\cite{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.

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Nil.

**Conflicts of interest**

There are no conflicts of interest.

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