Bilateral blunt cerebrovascular injury resulting in direct carotid-cavernous fistulae: A case report and review of the literature

Stephanie V. Avila, Brooke Van Noy, Michael Karsy, Matthew Alexander, John D. Rolston

Departments of Neurobiology and Anatomy, ¹Neurosurgery, Clinical Neurosciences Center, ²Radiology, University of Utah, Salt Lake City, Utah, USA

E-mail: Stephanie V. Avila - stephanie.avila@neuro.utah.edu; Brooke Van Noy - brookenhansen@gmail.com; Michael Karsy - Michael.karsy@hsc.utah.edu; Matthew Alexander - Matthew.alexander@hsc.utah.edu; *John D. Rolston - john.rolston@hsc.utah.edu

*Corresponding author
²Contributed equally to manuscript

Received: 26 June 18  Accepted: 10 September 18  Published: 19 November 18

Abstract

Background: Bilateral blunt cerebrovascular injury (BCVI) has been documented in 32 patients in the English-language literature and bilateral carotid-cavernous fistula (CCF) have been reported in only 1 patient. Here, we present a case of severe, unexpected bilateral BCVI with bilateral direct CCF and review the literature of BCVI, particularly cases of bilateral injury.

Case Description: A 65-year-old woman with episodic bradycardia presented after a motor vehicle accident. On arrival, she had a Glasgow Coma Scale of 3T and progressive dilation of her right pupil. Computed tomography imaging showed a 1.8-cm right epidural hematoma (EDH) with 6 mm of right-to-left shift. No acute skull-base fracture or injury in the area of the carotid canal was noted. The patient was treated with 3% hypertonic saline and mannitol before being taken to the operating room for emergent decompression of the hematoma. Although the patient initially presented with an EDH, significant intraoperative hemorrhage was identified during surgical evacuation and later confirmed as bilateral direct CCFs during angiographic evaluation. Because of the patient's devastating injuries, life-extending measures were not continued and the patient died.

Conclusions: A review of the literature indicates that bilateral CCFs are rare, having been reported only once previously. As this case demonstrates, CCFs may occur in high-energy injuries and should be considered even if the patient does not meet traditional screening criteria.

Key Words: Blunt cerebrovascular injury, carotid-cavernous fistula, dissection

INTRODUCTION

Blunt cerebrovascular injury (BCVI) is a potentially morbid result of traumatic brain injury that can result in dissection of blood vessels.¹²,³⁵ Dissections involve injury to the endothelial, intimal, or adventitial layers of the blood vessel,⁴ and carotid dissections can be associated with development of carotid-cavernous fistula (CCF). High cervical injuries and significant force of injury are risk factors for traumatic CCF.⁵⁻⁷ BCVIs occur in 1% of...
patients with blunt trauma, and CCFs are seen in 0.2% of traumatic brain injury and 4% of patients with sustained basilar skull fractures.\[14\] Bilateral injuries represent a subset of BCVI, accounting for at most 10% of highly selected case series;\[18\] however, traumatic bilateral CCF has only been reported once before.\[16\]

The use of computerized tomography angiography has enabled rapid evaluation of patients with BCVI; however, a high index of clinical suspicion is required because BCVI and CCF can be missed. For instance, the antiplatelet treatment compared with anticoagulation treatment for cervical artery dissection (CADISS) trial, which evaluated BCVI treatment modalities, demonstrated difficulty in diagnosing BCVI in up to 20% of cases.\[7\] These results and various guidelines suggest challenges in identifying BCVI even with advanced imaging techniques.\[7,12,40\]

We present a case of severe, unexpected bilateral BCVI with bilateral direct CCF and provide a review of the literature describing the diagnosis, management, and outcomes of bilateral BCVI.

**CLINICAL PRESENTATION**

A 65-year-old woman with unknown past medical history presented after a motor vehicle rollover in which she was restrained with a seatbelt. The patient was intubated at the scene, stabilized at an outside hospital, and transferred to our hospital. She exhibited episodic bradycardia en route but retained reactive pupils. On arrival, the patient’s Glasgow Coma Scale (GCS) score was 3T with progressive dilation of her right pupil. Computed tomography (CT) imaging showed a 1.8-cm right epidural hematoma (EDH) with 6-mm right-to-left shift but no acute skull-base fracture or injury in the area of the carotid canal [Figure 1a, b]. Other injuries included extensive osseous and soft tissue injuries in the thorax. The patient was treated with 3% hypertonic saline and mannitol before undergoing emergent hematoma decompression.

We completed evacuation of the EDH without difficulty; however, significant unexpected bleeding was uncovered from the skull-base and carotid-cavernous triangle. There was worsening bleeding from deep within the Sylvian fissure, prompting us to open the dura to achieve additional decompression. Extraluminal coagulation of the middle meningeal artery and ipsilateral neck pressure reduced the bleeding, but incompletely. We packed the wound and proceeded for emergent angiography to identify the source of bleeding.

Digital subtraction angiography (DSA) showed a large dissecting pseudoaneurysm of the cavernous segment of the right internal carotid artery (ICA), with arteriovenous shunting into the cavernous sinus consistent with a direct CCF. Outflow was noted through the bilateral inferior petrosal sinuses with no visualized cortical venous reflux [Figure 1c, d]. Flow in the right anterior (ACA) and middle (MCA) cerebral artery circulation was sluggish, and there was displacement of the right MCA branches because of mass effect from residual extra-axial blood and packing material. No abnormal fistula was seen during external carotid artery (ECA) injection (not shown). The left ICA demonstrated extensive dissection and arteriovenous shunting into the cavernous sinus, consistent with a direct CCF. Outflow from the left-sided CCF was noted into the petrosal sinuses and left external jugular veins [Figure 1e, f]. The left ACA and MCA flows were diminished, but no abnormality of the ECA was seen. Endovascular treatment and open surgical carotid bypass were discussed, but the patient proved to be hemodynamically unstable for treatment, and the family chose not to pursue further life-extending measures. This case report does not require patient consent per our institution.

**DISCUSSION**

CT, magnetic resonance angiography, or DSA can be used to screen for BCVI.\[51\] Various sets of criteria have been designed to predict the need for imaging to screen for BCVI, including the Denver and Memphis scores in adults and the Utah and McGovern scores in
Table 1: Summary of literature involving bilateral, traumatic blunt cerebrovascular injury (BCVI)

| Reference          | Age/Sex (n = 33) | Presentation                                      | Vessel(s) involved | Management                      | Follow-up (months) | Outcome                                      |
|--------------------|------------------|---------------------------------------------------|--------------------|---------------------------------|--------------------|----------------------------------------------|
| Fukuda et al. (1989) | 17/M             | Polytrauma, GCS11                                  | Vertebral; carotid | Heparin, antiplatelet           | 6                  | Improved exam, improved vessel anatomy       |
| Galetta et al. (1991) | 48/M             | Visual loss, facial droop                           | Carotid            | Heparin, warfarin               | 2                  | Loss of vision, unchanged facial droop       |
| Ballard et al. (1992) | 20/F             | Facial weakness, hemiparesis, expressive aphasia, tongue deviation | Carotid            | Heparin                         | 5                  | Alive                                        |
| Morrow and Smith (1995) | 37/F             | Polytrauma, hemiparesis                            | Carotid            | Heparin, warfarin               | 0                  | NA                                           |
| Janjua et al. (1996) | 41/F             | Lower extremity paraparesis                        | Carotid            | Anticoagulant                   | NA                | NA                                           |
| Duncan et al. (2000) | 39/M             | Numbness, hemiplegia, facial weakness              | Carotid            | Heparin                         | 1                  | Hemiplegia                                   |
| Lee and Jensen (2000) | 43/M             | Headache, visual disturbances                      | Carotid            | LMWH, aspirin                   | 2                  | Returned to baseline                         |
| Babovic et al. (2000) | 37/F             | Hemiparesis                                       | Carotid            | Clopidogrel                     | 8                  | Returned to baseline                         |
| Malek et al. (2000) | 43/F             | Hemiparesis                                       | Carotid            | Angioplasty, aspirin, ticlopidine | 20                 | Returned to baseline                         |
| Busch et al. (2000) | 27/F             | Progressive coma                                   | Vertebral; carotid | Surgical reconstruction         | NA                | NA                                           |
| Pittock et al. (2001) | 40/M             | GCS12, facial weakness, hemiparesis, hemineclect  | Carotid            | Heparin                         | 12                 | Mobile with assistance                       |
| Fanelli et al. (2004) | 17/M             | GCS9, hemiplegia                                  | Carotid            | Stent, ticlopidine, heparin     | 13                 | Mild, improving weakness                     |
| Clarot et al. (2005) | 38/M             | GCS9, hemiparesis                                  | Carotid            | None                            | 0                  | Death                                        |
| Yong and Heran (2005) | 25/M             | GCS6, dilated right pupil                          | Vertebral; carotid | Heparin bridge to warfarin      | 3                  | Improved                                     |
| Chokyu et al. (2006) | 61/F             | Tetraparesis                                      | Carotid            | Stent, aspirin                  | 2                  | Alive                                        |
| de Borst et al. (2006) | 13/F             | GSC 3                                             | Carotid            | LMWH, aspirin                   | 6                  | Patency of bilateral ICA                     |
| Adaletli et al. (2007) | 10/M             | Headache, hemiparesis                             | Vertebral; carotid | None                            | 2                  | Occlusion of posterior circulation           |
| Furtner et al. (2006) | 43/M             | Pain, amaurosis fugax                              | Carotid            | Heparin, warfarin, aspirin, fluidione | 12                 | Alive                                        |
| Fang (2008)         | 50/M             | Pulsatile proptosis, chemosis, intracranial bruit, decreased visual acuity | Carotid            | Stent, anticoagulation           | 24                 | Resolution of dissecting pseudoaneurysms and CCFs |
| Srivastava et al. (2008) | 58/M             | GCS11, hemiparesis, aphasia, SAH, atlantooccipital dislocation | Carotid            | Heparin, warfarin               | NA                | NA                                           |
| Leach and Malham (2009) | 23/F             | S AH, atlantooccipital dislocation                 | Vertebral; carotid | Enoxaparin sodium, warfarin    | 0                  | Ambulating, following commands in all extremities |
| Molacek et al. (2010) | 49/F             | GCS3, seizures                                    | Carotid            | Angioplasty                     | 0                  | Returned to baseline                         |
| Keilani et al. (2010) | 52/F             | Respiratory distress                              | Vertebral; carotid | Bilateral stents, aspirin      | 3                  | Patency of bilateral ICA                     |
| Samra et al. (2011) | 24/M             | Respiratory distress, upper and lower extremity tract, GCS6 | Carotid            | Aspirin                         | NA                | NA                                           |
| Chiba et al. (2014) | 60/F             | Unconscious                                       | Carotid            | None                            | 0                  | Died                                         |
| Koleilat et al. (2014) | 23/F             | Basal ganglia infarction, mandibular fracture, temporal contusion, hemiparesis | Carotid            | Heparin, warfarin               | 32                 | Independent ambulation, persistent left side weakness, persistent non-occlusive ICA dissection |

Contd...
The Denver screening criteria include arterial hemorrhage, cervical bruising in patients <50 years, expanding cervical hematoma, focal neurological deficit, neurological examination incongruous with CT findings, and stroke on secondary CT. Other high-risk factors that often prompt screening are Le Fort type 2 and 3 fractures, basilar skull fracture involving the carotid canal, diffuse axonal injury with GCS < 6, cervical spine fracture involving Cl–2 or the transverse foramen, cervical spine subluxation, and near-hanging with hypoxic-ischemic brain injury. In our patient, the observed neurological decline with concordant clinical and radiological findings was presumed to be due to an EDH, prompting surgical treatment without additional imaging, which might have been obtained had the patient been more stable. It is unlikely that this case would have been managed differently had the bilateral CCFs been discovered preoperatively, although this knowledge might have led to more rapid endovascular therapy after the EDH removal.

The Canadian Stroke Best Practice Recommendations for the Secondary Prevention of Stroke[30] and 2018 Guidelines for the Early Management of Patients With Acute Ischemic Stroke[31] recommended use of anticoagulation for low-grade (Biffl grade 1 and 2) injuries and combined endovascular stenting, vessel sacrifice, and surgical reconstruction for higher grade (Biffl grade 3–5) lesions. A meta-analysis of 34 clinical trials and 762 patients showed no difference between antiplatelet and anticoagulation treatment in terms of risk of death or stroke.[29] Similarly, the rate of ipsilateral stroke or death in the CADISS trial[32] did not differ significantly between antiplatelets and anticoagulants. Although the role of antiplatelet treatment for BCVI is established, anticoagulation in the setting of active intracranial bleeding is contraindicated. The presence of bilateral CCF in our case presented a treatment dilemma.

We identified 29 studies with 32 patients who had bilateral BCVI [Table 1]. Seven patients had nonmedical intervention (stent, angioplasty, surgical reconstruction) and 4 had no treatment; antiplatelet or anticoagulant treatment was used in 25 patients (some patients had both medical and nonmedical intervention). Previous reports of bilateral BCVI indicated that cases involved either focal or polytrauma often with concomitant injuries. CCFs represent arteriovenous shunting from the ICA or ECA into the cavernous sinus. Direct CCFs involve shunting directly from the ICA into the cavernous sinus. These are usually the result of trauma, although they can occur after rupture of a cavernous segment aneurysm. Endovascular therapy is the treatment of choice for direct CCFs. Coil embolization of the recipient cavernous is now the preferred first-line treatment. In rare cases, venous occlusion cannot be achieved, and vessel sacrifice is needed.

Bilateral CCFs are rare, with only 1 prior case reported after traumatic ICA dissection. Fang[16] reported a good 2-year outcome in a case of bilateral BCVI with bilateral high-flow CCF treated with bilateral detachable balloon occlusion. In our patient, hemodynamic instability prevented endovascular treatment. Among other reported cases with bilateral BCVI but not CCF, several patients died during the hospital course, often because of other polytrauma injuries.[6,10] In addition, most prior studies demonstrated significant neurological deficit on presentation, with many patients showing little improvement despite treatment. Various treatments were also performed for these patients, including medical, endovascular, and surgical therapies.

CONCLUSION

Bilateral CCFs due to trauma are exceedingly rare, having been reported only once previously. Although the injury was fatal in our case, we believe that continued improvements in the diagnosis and treatment of BCVI, as well as awareness that CCFs may occur in high-energy injuries and should be considered even if the patient does not meet traditional screening criteria, will improve future outcomes in cases of bilateral CCF.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Adaletli I, Sirikci A, Ulus S, Yilmaz MH, Kervancioglu S, Kurugoglu S. Traumatic bilateral vertebral artery dissection at the dural entry point site in a 10-year-old boy. Pediatr Surg Int 2006;22:468-70.
2. Babovic S, Zietlow SP, Garrity JA, Kasperbauer JL, Bower TC, Bise U.
1. Janjua KJ, Goswami V, Sagar G. Whiplash injury associated with acute bilateral carotid arterial injuries: Implications of a new grading scale. J Trauma 1999;47:845-833.

2. Burlew CC, Billi WL, Moore EE, Barnett CC, Johnson JL, Bensard DD. Blunt cerebrovascular injuries: Redefining screening criteria in the era of noninvasive diagnosis. J Trauma Acute Care Surg 2012;72:330-5; discussion 336-7; quiz 539.

3. Busch T, Aleksic I, Sirbu H, Kersten J, Dalichau H. Complex traumatic dissection of right vertebral and bilateral carotid arteries: a case report and literature review. Cardiovasc Surg 2000;8:72-4.

4. Chokyu I, Tsumoto T, Miyamoto T, Yamaga H, Terada T, Itakura T. Traumatic bilateral common carotid artery dissection due to strangulation. A case report. Intern Neurol 2006;12:149-54.

5. Clarot F, Vaz E, Papin F, Proust B. Fatal and non-fatal bilateral delayed carotid artery dissection after manual strangulation. Br J Anaesth 2000;85:476-8.

6. de Borst GJ, Slieker MG, Monteiro LM, Moll FL, Braun KP. Bilateral traumatic bilateral common carotid artery dissection. J Neurosurg Pediatr 2018;21:639-49.

7. Menon R, Perry S, Norris JW, Markus HS. Treatment of cervical artery dissection. Report of three cases. J Neurosurg 2000;92:481-7.

8. Miller PR, Fabian TC, Croce MA, Cagniannos C, Williams JS, Yang M, et al. Prospective screening for blunt cerebrovascular injuries: Analysis of diagnostic modalities and outcomes. Ann Surg 2002;236:386-93; discussion 393-5.

9. Storrow AB, Smith BA. Traumatic bilateral carotid dissection with concomitant cerebral infarction. J Emerg Med 2001;20:33-8.

10. Samra NS, Ravi AK, Johnson LW, Williams M. Traumatic bilateral carotid artery dissection. J La State Med Soc 2011;163:26-8.

11. Storrow AB, Smith BA. Traumatic bilateral carotid dissection. J Emerg Med 1995;13:169-74.

12. Wein T, Lindsay MP, Cote R, Foley N, Berlinger J, Bholg S, et al. Canadian stroke best practice recommendations: Secondary prevention of stroke, sixth edition practice guidelines, update 2017. Int J Stroke 2018;3:420-43.

13. Yong RL, Heran NS. Traumatic carotid cavernous fistula with bilateral carotid artery and vertebral artery dissections. Acta Neurochir (Wien) 2005;147:1109-13; discussion 1113.