

Traumatic cervical spine subarachnoid hemorrhage with hematoma and cord compression presenting as Brown-Séquard syndrome: illustrative case

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BACKGROUND Spinal hematomas are a rare entity with broad etiologies, which stem from idiopathic, tumor-related, and vascular malformation etiologies. Less common causes include traumatic blunt nonpenetrating spinal hematomas with very few cases being reported. In the present manuscript presents a case report and review of the literature of a rare traumatic entity of a cervical subarachnoid hematoma in association with Brown-Séquard syndrome in a patient on anticoagulants. Searches were performed on PubMed and Embase for specific terms related.

OBSERVATIONS A well-documented case of an 83-year-old female taking anticoagulants with traumatic cervical subarachnoid hematoma presenting as Brown-Séquard syndrome was reported. Six similar cases were identified, scrutinized, and analyzed in the literature review.

LESSONS Traumatic blunt nonpenetrating cervical spine subarachnoid hematomas are a rare entity that can happen more specifically in anticoagulant users and in patients with arthritic changes and stenosis of the spinal canal. Rapid neurological deterioration and severe disability warrant early aggressive surgical treatment. This report has the intention to record this case in the medical literature for registry purposes.

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KEYWORDS cervical; hematoma; subarachnoid; traumatic; Brown-Sequard; laminectomy

Spinal hemorrhages are a rare phenomenon that can cause severe neurological deficit and disability. The majority of spinal hemorrhages occur within the epidural space, and rarely within the subdural or intraparenchymal compartment.¹ Spinal hemorrhages within the subarachnoid space represent only 15% of all spinal hemorrhages.² The relative rareness, conflicting nomenclature, and association with other spinal or systemic conditions make spinal subarachnoid hemorrhage (SAH) a challenging diagnosis.

Although a spinal subarachnoid hematoma designates a discrete collection of blood leading to mass effect, a spinal subarachnoid hemorrhage denotes blood within the subarachnoid space that can travel throughout the subarachnoid space. We report an interesting case of traumatic spinal subarachnoid hemorrhage with hematoma formation. The precise pathogenesis is unclear but is likely related to rupture of the vessels that travel outside of the spinal cord parenchyma within the subarachnoid space.

Spinal SAH can be caused by numerous factors, with its natural history remaining largely unknown. Common etiologies include tumor-related hemorrhages as well as vascular malformations. The majority of tumor-related hemorrhages are typically associated with ependymomas, followed by neurofibromas, astrocytomas, and lastly, meningiomas.³ Spinal SAH that occur secondary to vascular malformations are frequently caused by arteriovenous malformations (AVMs), spinal angiomas, and aneurysms.² Spinal hematomas that occur as a result of trauma are less common.^{4,5} There are only seven reported cases of traumatic cervical SAH in the literature.^{6–12} Treatment is dependent on the patient's neurological examination as well as presence or absence of vascular pathology. Ventral spinal SAH may be medically managed with close observation in the absence of vascular malformation if the patient is neurologically intact, whereas dorsal spinal SAH may require surgical exploration even with minor neurological impairment because of its relatively easier access.^{1,7,10,13,14}

ABBREVIATIONS AVM = arteriovenous malformation; CT = computed tomography; MRI = magnetic resonance imaging; SAH = subarachnoid hemorrhage.

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In some cases, spinal bleeding can extend into the cranial compartment, making cranial imaging necessary in patients who develop mental status changes.^{8,10,15} To the best of our knowledge, this is the first report in English of a traumatic subaxial cervical spinal SAH causing Brown-Séquard syndrome in the absence of cranial involvement that was treated with laminectomy.

Illustrative Case

History and Examination

An 83-year-old female patient presented to the emergency department with severe right-sided neck and shoulder pain after sustaining a ground-level fall without loss of consciousness. The patient's fall resulted in large bruising within the occiput (Fig. 1A). The patient's home medications included diltiazem and rivaroxaban for her diagnosis of atrial fibrillation. Eight hours after the patient's arrival, the patient developed remarkable right-sided weakness. Physical examination revealed a patient with Glasgow Coma Scale of 15. The patient presented with severe torticollis and left-sided loss of light touch sensation. Strength testing revealed 0 out of 5 right upper extremity motor strength and 1 out of 5 right lower extremity strength using the standard neurological muscle strength grading. Strength testing of the left upper and lower extremities was fully intact. The patient's presentation was clinically consistent with Brown-Séquard syndrome. The initial computed tomography (CT) scan without contrast of the cervical spine was negative for fractures as well as signs of a stroke. The scan was remarkable only for arthritic

changes of the cervical spine (Fig. 1B). Magnetic resonance imaging (MRI) of the cervical spine with and without contrast was then performed which revealed a subarachnoid hematoma compressing the spinal cord from the right dorsolateral position (Fig. 1C–D).

Surgical Technique

The patient was urgently transported into the operating room for surgical intervention. After general anesthesia was administered, the patient was positioned prone on a Skytron table (Skytron LLC). The patient's head was clamped with Sugita pins and then positioned in gentle neck flexion (Fig. 1A). The patient underwent a traditional C3–5 laminectomy using a posterior midline approach, followed by a midline durotomy because there were no pathologic findings in the epidural space. An organized hematoma was identified with, an intact arachnoid membrane (Fig. 2A). The arachnoid membrane was carefully dissected under a microscope after which three of the dentate ligaments were released on the right side to provide further exposure of the hematoma (Fig. 2B). The right C4 nerve root was found to be severely compressed (Fig. 2C). The hematoma was then carefully dissected and removed, unveiling a compressed spinal cord displaced toward the contralateral side. Signs of neural tissue affliction were noted such as hyperemia within the right posterior surface of the spinal cord and within the C4 nerve root (Fig. 2D). No active bleeding was found after a careful direct inspection. A dural suture was performed with running stitches, followed by spinal fusion via lateral mass screws and rod placement (Fig. 3).

Outcome

The immediate postoperative neurological examination was unchanged. Postoperative imaging showed satisfactory screw and rod placement after SAH evacuation (Fig. 4). The patient was instructed to discontinue her rivaroxaban for 2 weeks following surgery. Eleven days postoperatively, the patient was diagnosed with several clinical complications including acute cystitis, recurrent pulmonary coccidioidomycosis, metabolic encephalopathy, a gastrointestinal bleed, deep vein thrombosis of the right cephalic vein, pseudomembranous colitis, and neurogenic bladder. All of the patient's complications were managed appropriately and were unrelated to her cervical spine SAH, with the exception of neurogenic bladder. The patient was discharged to an inpatient rehabilitation facility for 3 weeks and remained without neurological improvement on physical examination. A long-term neurological assessment was not feasible because the patient moved out of state.

Review of the Literature

A systematic literature review was performed through querying PubMed. Search terms included the following phrases: "traumatic AND cervical AND spinal cord subarachnoid hemorrhage," "traumatic AND cervical AND spinal cord subarachnoid hematoma," "acute AND Brown-Séquard Syndrome AND hematoma," and "subarachnoid hemorrhage AND cervical spine." A search on Embase was performed using the key terms: "spinal cord subarachnoid hemorrhage and 'Brown Séquard syndrome'." Inclusion criteria included published articles reporting traumatic cervical subarachnoid hematomas. Exclusion criteria included penetrating injury of the spine, occurrence not within the cervical spine, hematoma within another meningeal compartment rather than subarachnoid, occurrence of other type of bleeding rather than organized hematoma, and other causes of nontraumatic bleeding.

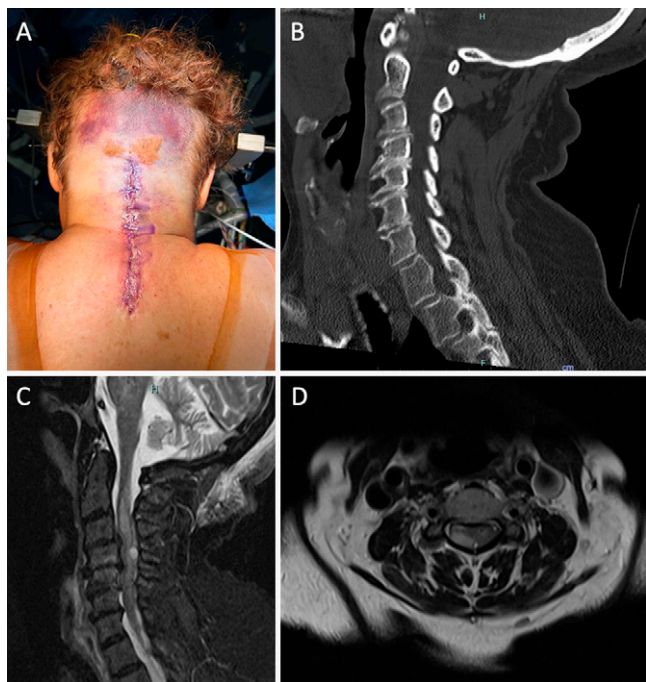


FIG. 1. A: Patient position on the surgical table with the head clamped on Sugita pins. Note the large bruise in the occipital region. **B:** Initial CT of the cervical spine demonstrating arthritic changes with osteophytes from C3 to C7 without fracture or malalignment. **C and D:** Pre-operative cervical MRI shows a hematoma located in the right dorsolateral aspect of the spinal cord and producing mass effect. The hematoma dimensions were 1.5 × 1.0 cm. Spondylolisthesis at the C3–4 segment is visible.

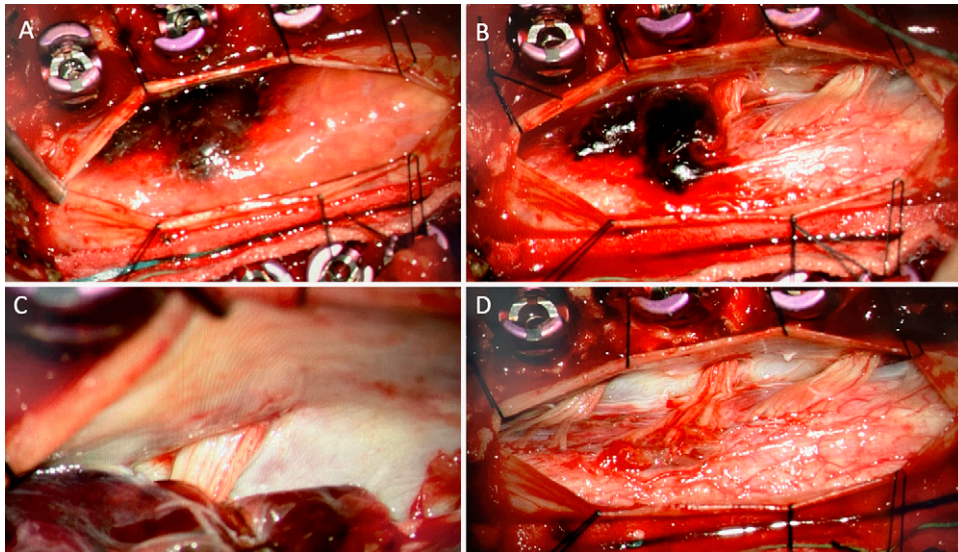


FIG. 2. A: Intraoperative aspect after dural opening. Arachnoid is preserved with the hematoma located in the right side displacing the cord laterally toward the left. **B:** After arachnoid dissection, the hematoma is better visualized. Dentate ligaments were released to provide further exposure. **C:** A hematoma is displacing the right-sided C4 nerve root. **D:** Image depicting a laterally displaced cord after removal of the hematoma with evident hyperemia.

A total of 687 articles were found in our search. After a careful review of all articles, only seven articles met inclusion criteria which reported traumatic subarachnoid hemorrhage within the cervical spine. One article was excluded because it was written in Japanese. Only six cases reported in English language and were included in this review: Kim et al.,⁸ Wolfe et al.,¹² Rascón-Ramírez et al.,¹⁰ Domenicucci et al.,⁷ Russell et al.,¹¹ and Chang et al.⁶ (Table 1).

Discussion

Observations

Blunt nonpenetrating traumatic hematomas of the spine are a traditionally rare entity. Of all spinal hematomas, epidural is the

most common followed by subarachnoid and subdural hematomas.² Spinal hematomas are frequently idiopathic, consequent of vascular malformation or tumor with trauma being cited as an etiology in only 2.5% of all cervical hemorrhages.^{2,4} Hematoma formation in the subarachnoid compartment of the spine is rare and difficult to diagnose typically due to low suspicion when there is a lack of vertebral fracture or displacement on initial imaging. High suspicion is warranted avoid delay in treatment. Severe neurological deficit and disability is usually present in the absence of proper and timely treatment, often leading to death or permanent neurological deficits.

The pathophysiology of traumatic SAH within the cervical spine is not properly established, however, subarachnoid spinal hemorrhage is thought to originate by rupture of radicular blood vessels, arteries, or veins, close to where the root emerges from the spinal cord, within the subarachnoid space. Blood collection with subsequent clot formation under the arachnoid layer compresses both the spinal cord and nerve roots causing neuronal damage and neurological deficit. Cranial subarachnoid hemorrhages result from blood spreading through the cisterns and subarachnoid space. This is in contrast to spinal subarachnoid hemorrhages, which are organized clots with a preserved arachnoid layer. In some cases, blood can spread and reach the cranial cisterns causing mental status changes and coma.^{8,11} In this literature review, the only two cases with cranial involvement included young patients, at ages 17 and 19.^{8,11} This can validate the theory that the presence of spinal arthritis with stenosis of the spinal canal may favor the formation of organized blood clot. The presence of osteophytes and narrowing of the spinal canal would in theory prevent blood from spreading throughout the cerebrospinal fluid and consequently reach the cranial compartment.² The other reported cases of cervical SAH without cranial involvement included patients over the age of 65 who all demonstrated presence of remarkable arthritic changes of the cervical spine.

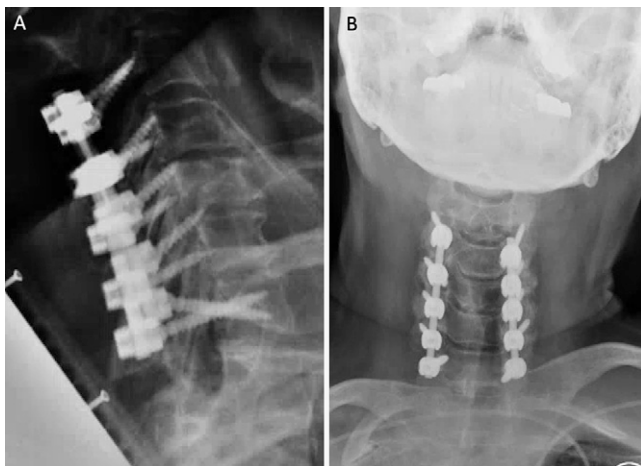


FIG. 3. Postoperative imaging (**A** and **B**) shows C3–7 laminectomy with proper screw and rod placement.

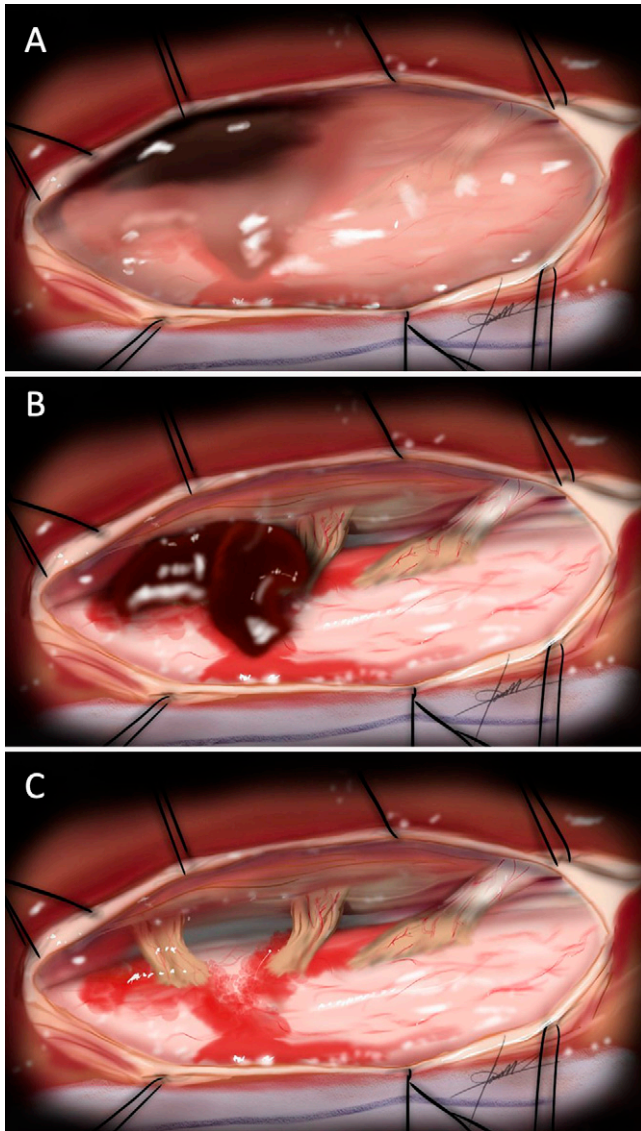


FIG. 4. Illustrations representing the intradural phase of the surgery. **A:** After dural opening, the is visualized under an arachnoid layer. **B:** After arachnoid dissection, the hematoma organized is visualized. **C:** After hematoma removal, the spinal cord is hyperemic.

Most cases of spinal hematomas are characterized by pain and have an acute course resulting in severe paralysis with or without bladder/intestinal dysfunction.² The clinical manifestations of spinal SAH are usually severe, commonly presenting with remarkable neurological deficits. Only one of the five patients included in this review presented with only neck pain.⁷ One patient presented with quadriplegia,⁶ one presented with quadriplegia,¹¹ and three patients, including the present report, presented with the clinical manifestations of Brown-Séquard syndrome.^{10,12} Brown-Séquard syndrome was first described by Mauritian physiologist and neurologist Charles-Édouard Brown-Séquard in 1850. This syndrome results from corticospinal interruption which produces ipsilateral paralysis below the level of the lesion, fasciculus gracilis and cuneatus (dorsal column) interruption resulting in ipsilateral loss of vibration, sensation, and proprioception,

and spinothalamic tract interruption leading to contralateral loss of touch, pain, and temperature sensation.¹⁶ Mori et al.⁹ also reported a case of a 43-year-old male with traumatic hyperextension cervical spinal subarachnoid hematoma who presented with Brown-Séquard syndrome managed with a laminectomy; however, this report was excluded from our literature review because it was written in Japanese.

Russell et al.¹¹ reported a case of traumatic cervical SAH that occurred in association with a brachial plexus avulsion. That can corroborate the theory that harsh stretching of the nerve roots can cause shearing forces with consequent blood vessel rupture. Sequela of this complication is only aggravated by the use of anticoagulants. The aging population is increasing making the use of anticoagulants more prevalent in the management of conditions such as atrial fibrillation, cerebrovascular diseases, and coronary artery disease. A report by Domenicucci et al.⁷ identified that 40.5% of patients with spinal hematomas had some type of coagulopathy. In this review, Domenicucci et al.⁷ also perceived that 44.9% of all spinal hematomas were iatrogenic, mostly related to lumbar punctures, in which 33% of these patients also had coagulation problems. Domenicucci et al.⁷ also ascertained a better outcome in patients who were treated with surgery. Interestingly, Chang et al.⁶ reported a case of traumatic cervical subarachnoid hematoma at C1 after a head trauma.

Lessons

In the present case, urgent surgery was indicated for decompression of both the spinal cord and nerve roots. Use of a microscope was imperative for allowing a safe arachnoid dissection. Surprisingly, the hematoma was very well organized and rigid. Dissection from the spinal cord was performed very carefully because of tight adhesions present throughout the spinal cord and cervical nerve roots. A clear layer of arachnoid was identified as demonstrated in Fig. 2A with the hematoma undoubtedly laying within the arachnoid web layer. Release of the dentate ligaments was essential in allowing gentle mobilization of the spinal cord to facilitate the dissection. Because of the rigid aspect and tight adhesions present, this clot was likely more than 24 hours old, despite a more immediate time of the onset of symptoms (Fig. 4). After removal of the hematoma, a remarkable area of hyperemia was identified, suggesting significant neural tissue damage that corresponded with the patient's clinical presentation. The main differential diagnosis includes an arteriovenous malformation, which was not found after careful inspection under the microscope. The presence of arthritis and spondylolisthesis at C3–4 is believed to have facilitated the hematoma formation, in conjunction with the use of anticoagulants. From our literature review, five of the six patients were managed operatively, reinforcing the idea that these types of lesions require aggressive treatment.

This study is not without limitations. First, it describes traumatic cervical SAH from a small series of cases reviewed from the available literature in addition to our presenting case. Second, the nomenclature describing spinal subarachnoid and spinal subdural hematomas may be confusing because it has not been standardized in the literature.

Traumatic blunt nonpenetrating cervical spine subarachnoid hematomas are a rare entity that can occur in patients with arthritic changes and stenosis of the spinal canal. These hematomas may be aggravated or incited by the use of anticoagulants. Rapid neurological

TABLE 1. Literature review

Authors & Year	Age (yrs)	Gender	Presentation	Anticoagulant?	Treatment	Brain Involvement	Remarkable Arthritis?
Kim et al., 2015 ⁸	17	M	Hemiparesis	No	Laminectomy	Yes	No
Wolfe et al., 2017 ¹²	67	M	Brown-Séquard	Yes	Nonoperative	No	Unknown
Rascón-Ramirez et al., 2018 ¹⁰	83	F	Brown-Séquard	No	Hemilaminectomy	No	Yes
Domenicucci et al., 2005 ⁷	70	F	Neck pain	Yes	Nonoperative	No	Yes
Russell and Mangan, 1980 ¹¹	19	M	Quadriplegia	No	Laminectomy	Yes	No
Chang et al., 2012 ⁶	63	F	Quadriparesis	No	Laminectomy	No	No
Present case	83	F	Brown-Séquard	Yes	Laminectomy	No	Yes

deterioration and severe disability warrant early aggressive surgical treatment. Raised awareness is necessary in our aging population as the use of anticoagulants becomes more prevalent.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: de Andrada Pereira, Meyer, Alvarez Reyes, Hurlbert. Acquisition of data: de Andrada Pereira, Alvarez Reyes, Brasiliense, Hurlbert. Analysis and interpretation of data: de Andrada Pereira, Meyer, Hurlbert. Drafting the article: de Andrada Pereira, Meyer, Alvarez Reyes, Hurlbert. Critically revising the article: de Andrada Pereira, Meyer, Alvarez Reyes, Orenday-Barraza, Hurlbert. Reviewed submitted version of manuscript: de Andrada Pereira, Meyer, Alvarez Reyes, Orenday-Barraza. Approved the final version of the manuscript on behalf of all authors: de Andrada Pereira. Administrative/technical/material support: de Andrada Pereira, Meyer. Study supervision: de Andrada Pereira, Hurlbert. Illustration rendering: Orenday-Barraza.

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