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Functional recovery in Brown-Séquard plus syndrome: A case report

Deja Rush, Karnesha Goins, Keli Doe, Damirez Fossett

ABSTRACT

Introduction: Brown-Séquard syndrome (BSS) is a rare neurological condition characterized by hemiplegia and hemianesthesia. It occurs as the sequelae to a hemi-transection of the spinal cord, and constitutes an incomplete spinal cord injury. We report a case of presumed BSS that resulted from a stab wound to the thoracic spine.

Case Report: A 64-year-old male presented to Howard University Hospital with complaints of back pain and loss of left lower extremity motor function. Neurological examination revealed weakness in the L2 through S1 nerve root distributions in the left lower extremity and hypoesthesia along the L2 through S1 dermatomal distributions in the right lower extremity. Imaging disclosed a foreign body extending from the muscle layer through the T6–T7 disc space. The patient was treated operatively with removal of a knife blade; the handle of which had been broken off at the surface of the skin. Post-operatively, prior to his discharge to an acute rehabilitation center, the patient's motor symptoms improved while his sensory symptoms worsened. He was lost to follow-up for approximately six months and returned with a debilitating spastic paraparesis.

Conclusion: The presenting symptoms of BSS are not always uniform, and thus may constitute a Brown-Séquard plus syndrome (BSPS). Surgical intervention is rare; however, it may occasionally be necessary in the setting of penetrating trauma. Long-term functional recovery for BSS as documented in the literature is variable. With aggressive physical therapy and rehabilitation, a good outcome is attainable. The functional outcome of BSPS, however, may not be as favorable.

Keywords: Brown-Séquard syndrome, Brown-Séquard plus syndrome, Functional recovery, Surgical management

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INTRODUCTION

Brown-Séquard syndrome (BSS) is defined as an incomplete spinal cord injury that results in the loss of ipsilateral motor function and the contralateral loss of pain and temperature sensation below the level of the lesion [1]. It is rare, accounting for 1–4% of spinal cord injuries, and occurs due to hemi-transection of the spinal cord from either a traumatic or non-traumatic event [1, 2]. Although a traumatic etiology is most common (i.e., stab wound, gunshot injury, and motor vehicle accident), non-traumatic causes such as spinal tumors, infection, ischemia, or disc herniation may also occur [1].

Given that injuries resulting in BSS can occur at any level along the spinal cord, patients may present with a myriad of neurologic deficits. In more severe cases,

where the cervical or thoracic spinal cord are involved, BSS can eventually progress to complete ipsilateral paralysis [1]. The diagnosis requires a thorough patient history, neurologic evaluation, and appropriate imaging demonstrating a unilateral spinal cord injury. In cases of BSS not caused by acute trauma, further work-up, such as serology, cultures for infectious etiologies, or gadolinium enhanced magnetic resonance imaging (MRI) for neoplastic causes, can be performed to determine the source of injury. The treatment of BSS is individualized, and in instances of trauma may require surgery. Prognosis varies depending on the extent of spinal cord injury; recovery normally takes weeks but may take up to two years [2]. Typically, however, the prognosis is excellent with 75–90% of patients returning to walking independently at the time of discharge from rehabilitation [1].

Our case describes a patient who underwent surgery to remove an embedded knife blade; an injury that resulted in a Brown-Séquard syndrome. In follow-up, however, the patient's condition significantly worsened.

CASE REPORT

A 64-year-old male presented to our trauma center with complaints of upper back pain and the inability to move his left leg after being stabbed during an altercation. The knife handle was found on the ground at the scene of the stabbing, but the knife blade itself was missing. On arrival to the hospital 35 minutes later, examination revealed a single 1 cm midline laceration at the T6 level with no external evidence of a retained foreign body. Neurologically, the patient had diminished motor strength in the left L2 through S1 nerve root distributions, but intact sensation to pain and light touch. Conversely, he had full motor strength throughout the contralateral L2 through S1 nerve root distributions, but diminished sensation to pain and light touch. The remainder of his exam was benign, with intact rectal tone and perianal sensation. A chest X-ray and computed tomography (CT) imaging of the thoracic spine revealed a large radiopaque foreign body, presumed to be the retained knife blade. It transversed the posterior spinal column, spinal cord, and the intervertebral disc between T6 and T7 (Figures 1 and 2).

The patient was taken to the operating theater within 16 hours of presentation. He was placed in the prone position and a standard posterior midline approach to the thoracic spine was performed. The knife blade was isolated in the paraspinal muscles, and its entry into the spinal canal was noted to be between the left T6 and T7 hemi-lamina. Cerebrospinal fluid emanated from the wound, indicating a dural laceration. Further dissection was performed to free the knife blade from the surrounding tissues (Figures 3 and 4). The spinous processes were removed and laminectomies at T6 and T7 were performed. The knife blade, which was seen

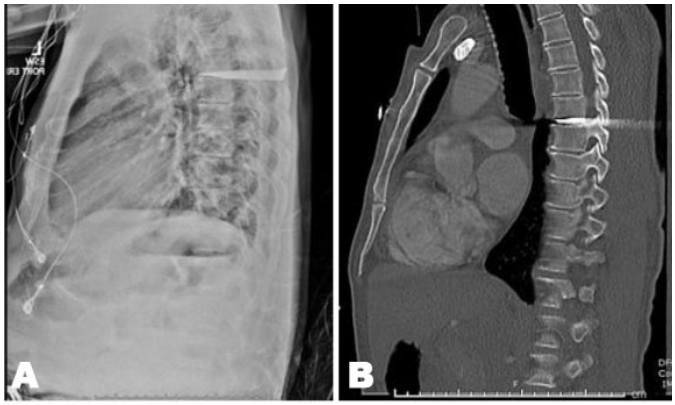

Figure 1: (A) Lateral chest X-ray showing a retained foreign body between T6 and T7, (B) Sagittal thoracic CT showing a foreign body traversing the spinal canal. Note that the foreign body comes to rest in the T6–T7 disc space, narrowly avoiding penetration into the abdominal aorta.

Figure 2: Axial CT showing the knife blade traversing the spinal cord.

Figure 3: Intra-operative photograph of the patient in the prone position, demonstrating a 1 cm stab wound to the mid-thoracic region. No evidence of a retained foreign body was grossly visible or palpable.

Figure 4: Intra-operative photograph demonstrating the retained knife blade lodged between the T6 and T7 hemi-lamina. For orientation purposes, the head is at the top of the photograph.

to be embedded within the left half of the spinal cord, was carefully removed and a primary dural repair was performed using 6.0 nurolon suture. Duragen and fibrin glue were placed in the epidural space to augment the dural closure (Tables 1 and 2).

Table 1: Manual muscle test grading upon presentation

	Left	Right
Hip flexion	0	5
Hip extension	0	5
Hip abduction	1	5
Hip adduction	1	5
Knee flexion	0	5
Knee extension	0	5
Plantar flexion	0	5
Dorsiflexion	1	5
Great toe flexion	5	5
Great toe extension	5	5

Grade 0 denotes no discernable muscle contraction. Grade 1 denotes a muscle contraction that is palpable but with no obvious motion. Grade 2 denotes a muscle contraction with motion that can only be appreciated with reduced gravity. Grade 3 denotes a muscle contraction against the force of gravity but no added pressure. Grade 4 denotes a muscle contraction against gravity with slight added pressure. Grade 5 denotes a muscle contraction against gravity and full added pressure [3].

Table 2: Light touch sensory test grading upon presentation utilizing the International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI)

	Left	Right
Sural nerve distribution	2	1
Saphenous nerve distribution	2	1
Deep peroneal nerve distribution	2	1
Superficial peroneal nerve distribution	2	1
Tibial nerve distribution	2	1

Grade 0 denotes absent sensation. Grade 1 denotes altered, including diminished or hypersensitive, sensation. Grade 2 denotes normal sensation [4].

On post-operative day 1, the patient's neurologic exam slightly worsened with the loss of sensation to pain and light touch extending further cephalad to the right hemithorax.

Pre-operatively, he only had the loss of pain and light touch below the level of the right hip. His exam was otherwise unchanged. Over the next 10 days the patient was treated with empiric antibiotics and he received daily physical and occupational therapy. Prior to discharge to an acute rehabilitation facility, his exam showed improvement in left ankle plantar and dorsiflexion.

However, paresthesias persisted in his right lower abdomen radiating to the right lower extremity.

DISCUSSION

In 1846 Charles-Édouard Brown-Séquard described a rare disorder that emerges secondary to hemisection of the spinal cord [1]. In this syndrome, which now bears his name, damage occurs to multiple spinal cord pathways including the corticospinal, medial lemniscus, and spinothalamic tracts. The meticulously precise cytoarchitectural design of the spinal cord dictates the profound neurologic sequelae of such an injury. Loss of innervation from upper motor neurons in the corticospinal tract leads to ipsilateral spastic paralysis below the lesion; the loss of innervation from lower motor neurons in the corticospinal tract leads to ipsilateral flaccid paralysis in the muscles supplied by that specific level. Disruption of the medial lemniscus results in ipsilateral loss of vibration, proprioception, and fine touch while impairment of the decussating spinothalamic tract results in the contralateral loss of pain and temperature sensation [1].

Given the multifaceted nature of the disease's pathophysiology, the clinical presentation of pure BSS is extremely rare; instead most reports describe patients who are suffering from what is known as Brown-Séquard plus syndrome (BSPS). The BSPS diagnosis broadens the defining characteristics of a BSS to encompass elements not seen in the classic syndrome. Nevertheless, BSS and BSPS type presentations still make up only between 2%

and 4% of all traumatic spinal cord injuries [1]. Based on our patient's initial neurological exam, his diagnosis did not fit the definition of a "pure" BSS. He had ipsilateral loss of motor function but not at every level below the lesion as evidenced by his ability to perform great toe extension, as well as hip abduction and hip adduction. He also did not show any signs of ipsilateral loss of vibration, proprioception, or fine touch. Contralaterally, our patient lost pain and non-discriminative touch but did not report the loss of temperature sensation. His presentation is therefore more consistent with BSPS [5].

As previously stated, BSS/BSPS can be caused by a litany of precipitating factors. However, the most common cause can be attributed to trauma; particularly penetrating trauma [1]. Koehler et al. reviewed over 600 published cases of BSS/BSPS and of the 81 reports with sufficient available data for their study, the largest percentage of origin was traumatic in nature (44.44%). Moreover, the authors stated that the typical inciting event was a stab wound [5]. This pattern of injury, though tragic, has been documented repeatedly. Peacock et al. reported 450 cases of traumatic spinal cord injury (SCI) caused by stabbings. Of those, 357 (79%) were characterized as incomplete lesions and 248 (55%) resulted in BSS [6]. Similarly, Lipschitz et al. reported 130 cases of traumatic SCI caused by stabbings. Of those, 91 (70%) were characterized as incomplete lesions with 16 (12%) and 18 (14%) cases of BSS and BSPS respectively [7]. Waters et al. reported 32 cases of traumatic SCI caused by stabbings. Of those, 20 (63%) were characterized as incomplete lesions and 10 (31%) resulted in BSS [8]. Similar to many of the above accounts, the origin of our patient's BSPS was trauma; a stab wound to the posterior thoracic spine.

The source of the injury in BSS/BSPS plays an important role in dictating the site of the injury. McKinley et al. reported 30 cases of incomplete SCI that resulted in BSS and Taylor et al. reported another series of 27 cases. Both noted the most common site of injury to be the cervical spine; however, only a small fraction of their cases involved stab wounds [9, 10]. In the Peacock study, however, the primary focus was penetrating trauma. 218 (61%) of the incomplete lesions occurred in the thoracic spine while 112 (32%) occurred in the cervical spine. Only 27 (7%) occurred in the lumbar spine [6]. In the setting of BSS/BSPS, stab wounds are the leading cause of penetrating trauma. Therefore, it is not surprising that the thoracic spine is the most common site of injury. Other segments of the spine may more commonly be the site of origin for non-traumatic BSS.

Treatment for BSS varies greatly based on the initial presentation. However, most agree that surgery in traumatic BSS/BSPS secondary to penetrating injuries is only indicated when there is a retained foreign body, evidence of a cerebrospinal fluid leak, or infection/sepsis [6, 7]. The literature suggests that patients with retained foreign bodies who undergo early surgical intervention have less complications and lower risk of infections. Prophylactic broad-spectrum antibiotics for 48 hours is seemingly sufficient in most cases. Though not explicitly addressed in the literature, steroids are generally avoided in setting of penetrating spine trauma especially when there is a concern for infection. In our case, reports of the altercation identified a knife handle with a missing blade at the scene. Our imaging studies showed a foreign body traversing the patient's spinal canal which was presumed to be the missing knife blade. As such, our team decided the appropriate treatment protocol to follow was urgent surgery. The goals of the operation were to remove the foreign object and repair the dura. The joint space was not violated, therefore a T6 and T7 decompressive laminectomy without instrumented fusion, and a dural repair were all that was deemed necessary. Our patient was maintained on antibiotic therapy until discharge to the rehabilitation facility because he was actively draining cerebrospinal fluid (CSF) at the time of surgery, and the stab wound itself was considered a contaminated wound.

Review of the literature indicates that in general BSS patients have a good neurologic prognosis. Because the injury to the spinal cord is incomplete, potential exists for substantial recovery.

McKinley et al. found that in a cohort of SCI patients, those with BSS achieved the best outcomes in terms of functional improvement upon discharge [9]. The average length of stay (LOS) in the acute hospital setting and acute rehabilitation were 10.2 and 27.2 days, respectively. Utilizing the modified Barthel Index (MBI), Roth et al. demonstrated statistically significant differences between scores at admission and upon discharge from the hospital in patients' abilities to perform the designated self-care and mobility tasks independently. Moreover, statistically significant changes in MBI scores were also shown during rehabilitation [11].

Koehler et al. found that 80% of BSS patients and 85% of BSPS patients whose injuries were of traumatic origin fully or almost fully recovered [5]. Peacock et al. reported a good recovery (defined as walking with minor or no support) in 65.6% of patients and partial recovery in 17.1% of patients whereby major support was necessary. The majority of patients (71.6%) had a total LOS in the hospital of less than three months [6]. Lipschitz et al. reported that patients with incomplete injuries were more likely to be deemed fit to return to work. At the time of the study, the overwhelming majority of patients (96.7%) were at least "up and about" with some already having fully recovered [7]. In spite of the wide spectrum of patient presentations, the literature demonstrates consistency in the favorable prognosis of BSS and BSPS.

Most studies base prognosis of BSS and BSPS on recovery of motor function, while few address recovery of sensory modalities and/or bowel and bladder function. In a review of 27 cases, Taylor et al. assessed the extent and rate of recovery of Brown-Séquard spinal cord injuries secondary to trauma. They noted motor damage was the greatest at onset, contralateral weakness recovered quickly, and ipsilateral paresis began recovering after nine days, on average. The greatest degree of ipsilateral

motor recovery occurred within the first 18 months after injury, and rapid return of motor strength was deemed indicative of a good long-term prognosis. Spasticity occurred bilaterally on average 31 days post-injury, and increased muscle tone was noted most obviously in the extensor muscles of the lower extremities [10].

In this study, there was no definite correlation between the onset of motor and sensory recovery. At the onset, 1 (4%) patient demonstrated no sensory loss and 5 (24%) had a dissociated sensory loss that ultimately completely recovered. 21 (78%) patients had a loss of all sensory modalities but began to recover at 25 hours post-injury, on average. On the contralateral side, most of these 21 patients had little further recovery, however, 9 (43%) experienced a drop in the level of their sensory loss. Ultimately, complete dissociated sensory loss remained in 3 of these 21 patients (14%), while only partial dissociated sensory loss remained in 12 (57%) patients. On the ipsilateral side, 20 (95%) patients demonstrated loss of posterior spinal column function. This loss ultimately remained complete in 3 (15%) patients, was incomplete in 10 (50%), and disappeared in the remaining 7 (35%) patients. Most patients who displayed sensory dysfunction on the ipsilateral side, showed the full extent of their sensory recovery within 1 week. In regard to bladder function, 6 of the 27 patients (22%) retained complete function at the onset. Of the 21 (78%) patients who developed bladder issues, 17 (81%) had immediate retention that required treatment. Ultimately, 13 of these 17 (76%) patients had complete return of normal bladder control within the first six weeks. In regard to bowel function, 8 of the 27 (30%) patients retained complete function at the onset. Of the 19 (70%) patients who had bowel issues, 17 (89%) had fecal retention and 2 (11%) had fecal incontinence. Ultimately, 7 (37%) patients made complete recovery by six months, but 11 (58%) patients still had issues with constipation. Important to note, the authors stated that in general, bladder, bowel, and sexual function demonstrated a linked recovery pattern [10].

Unfortunately, our patient's recovery has not followed the general pattern that is described in the literature. His condition remained relatively unchanged while in the acute care setting; he was transferred to an acute rehabilitation hospital and ultimately lost to follow-up. Upon resurfacing several months later, the patient gave an interval history of having spent a very short period of time in the acute rehabilitation facility prior to transfer to subacute rehabilitation. It was while in subacute rehabilitation that he returned to us for follow-up. To our chagrin, his neurologic picture was dismal. Our patient had diminished sensation to pain and light touch extending from his right hemithorax to his right lower extremity. He continued to have urinary incontinence and was unable to feel the urge to void. Profound constipation remained an issue. Though there was slight improvement in dorsiflexion and plantar flexion of the left big toe, the left leg was significantly cooler to touch than the right leg below the knee. Most disconcerting was the fact that he was profoundly spastic in both lower extremities. His legs were flexed at the knees and could not be passively or actively extended and any slight stimulus to the lower extremities caused both legs to spontaneously withdraw. Rigidity in the right leg was worse than the left and in the recumbent position, the patient continually had spontaneous, non-painful bilateral lower extremity spasms. Unfortunately, at nine months post-injury, none of these findings have improved. Bilateral involuntary spasms and profound spasticity continue to be the most predominant issues for our patient despite the use of high-dose oral baclafen therapy.

CONCLUSION

The presenting symptoms of Brown-Séquard syndrome are not always uniform, and thus may constitute a Brown-Séquard plus syndrome. Surgical intervention is rare; however, it may occasionally be necessary in the setting of penetrating trauma. Long-term functional recovery for Brown-Séquard syndrome as documented in the literature is variable, but attainable, if the patient undergoes aggressive physical therapy and rehabilitation. The functional outcome of Brown-Séquard plus syndrome may not be so favorable.

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Author Contributions

Deja Rush – Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Karnesha Goins – Acquisition of data, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Keli Doe – Acquisition of data, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Damirez Fossett – Conception of the work, Design of the work, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.