Spinal Cord (2007) 45, 744–748

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Case Report

Brown–Sequard syndrome produced by cervical disc herniation with complete neurologic recovery: report of three cases and review of the literature

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Study design: Case report.
Objective: To report three cases of Brown–Sequard syndrome (BSS) associated with cervical disc herniation.
Method: We describe clinical and radiographic review of three patients who presented with BSS caused by cervical disc herniation. Three patients presented with ipsilateral motor weakness and diminished sensation to pain and temperature on the contralateral side. Magnetic resonance images of the cervical spine in all cases, showed a large paramedian disc herniation at C5–C6, with ipsilateral severe spinal cord compression. Microsurgical removal of the herniated disc via anterior foraminotomy was performed and complete decompression of the spinal cord was achieved.
Results: Postoperatively, the neurological symptoms recovered rapidly with a complete remission of their symptoms.
Conclusion: Although BSS is rarely associated with degenerative cervical spine disease, cervical disc herniation should be kept in mind and prompt evaluation is indicated. Anterior foraminotomy suffices for spinal cord decompression with improvement of the neurological function.
Spinal Cord (2007) **45,** 744–748; doi:10.1038/sj.sc.3102025; published online 6 February 2007

Keywords: Brown–Sequard syndrome; cervical disc herniation; foraminotomy

Introduction

Brown–Sequard syndrome (BSS) involves corticospinal tract compression resulting in ipsilateral loss of motor function and spinothalamic tract dysfunction resulting in contralateral loss of pain and temperature sensation. BSS is observed most frequently in association with traumatic injuries to the spinal cord and extramedullary spinal cord tumors.[1–3] On the other hand, cervical disc herniation has rarely been considered to be a cause of BSS, and only 26 cases have been reported. We report three cases of disc herniation at the C5–C6 level with severe hemicompression of the spinal cord, resulting in BSS. To the best of our knowledge, these are the first reported cases of BSS caused by cervical disc herniation, which were successfully treated by anterior foraminotomy.

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Case report

Case 1
A 56-year-old man suffering from a sudden onset of right hemiparesis was brought to the emergency room. During the 4 days before admission, he experienced right shoulder pain, which was managed conservatively. He was admitted to the Department of Neurology and treated under the diagnosis of focal infarction, because the brain magnetic resonance images (MRI) showed the presence of a multifocal lacunar infarction in both basal ganglia, the centrum semiovale and the periventricular white matter. There was no history of trauma to the head or neck. On admission, motor examination revealed right hemiparesis (grade 4/5), with particular weakness of the intrinsic muscles of the right hand (grade 2/5). On the day following admission, his right hemiparesis worsened progressively with significant weakness developing in the right leg. Difficulty in urinating was then noted and catheter insertion became necessary. Finally, 4 days after admission, the patient

was referred to our department after the cervical MRI was checked. Neurologic examination revealed diminished sensation to pain and temperature on the left side below the T10 dermatome. Right-arm weakness had progressed to a grip strength of grade 1/5, and the patient was unable to ambulate because of significant weakness in the right leg, which was consistent with a diagnosis of BSS. The patellar tendon reflexes in the lower extremities were hyperactive bilaterally. Cervical spine X-rays showed diffuse spondylosis, which was most marked at C5–C6. MRI of the cervical spine showed a large disc herniation at the C5–C6 level causing focal right-sided cord compression (Figure 1a and b). Right anterior foraminotomy was performed to decompress the spinal cord. Using an operating microscope, a 5×8 mm hole was made medial to the uncal joint with a drill. There was a large tear in the right posterolateral annulus. A large amount of herniated disc material, which had herniated posterior to the annulus, was found to be compressing the right side of the cord. The herniated disc fragment was gently removed with a microforceps and complete decompression of the spinal cord was achieved. It was confirmed with a blunt nerve hook that no more disc material was remained, and the small hole was packed with thin gelatin sponge. Postoperatively, the patient improved rapidly and was discharged on the 7th postoperative day. One month later, the patient was able to walk without assistance and was found to have slowly regained strength in his right arm. The postoperative MRI, obtained 1 month after surgery, showed complete removal of the disc fragment and satisfactory decompression of the spinal cord (Figure 1c). Two months later, the numbness and weakness had disappeared and he continued to do well at 30 months of follow-up. Mild narrowing of the intervertebral space at C5–C6 was shown in the lateral view of cervical spine, obtained 2 years after surgery.

Case 2

A 47-year-old man presented with a 2-week history of numbness in the left hand and left shoulder pain, which subsequently developed into left hemiparesis. He was then managed conservatively at a local hospital under the diagnosis of a cerebrovascular accident, until progressive left leg weakness and ataxia led him to be transferred to our hospital. There was no history of trauma to the head or neck. The motor examination revealed mild weakness of the left arm and a moderate weakness in the left leg with a spastic gait. The patient presented diminished sensation to pain and temperature in the right arm and leg, hypesthesia and hypalgesia in the left arm and hyperreflexia of the lower extremities, which were consistent with BBS combined with radicular symptoms. MRI of the cervical spine revealed a large left extradural paramedian disc herniation at the C5–C6 level, with ipsilateral severe spinal cord compression and high-signal intensity within the spinal cord adjacent to the herniated disc. (Figure 2a and b). Several disc fragments were removed by a left anterior foraminotomy, and complete decompression of the spinal cord was obtained. The postoperative course was uneventful, with gradual improvement of the patient's neurological function being observed. The patient regained normal motor strength and sensation at 2 months after surgery. Postoperative computed tomograms (CT) showed complete decompression of the spinal cord through the small tunnel (Figure 2c). At 18 months follow-up, good clinical outcome was noted, and flexion and extension dynamic roentgenograms demonstrates an acceptable stability, despite mild retrolisthesis during extension at C5–C6.

Case 3

A 45-year-old man presented with a 9-month history of right shoulder pain. During the last 2 months before admission, he developed right hemiparesis that progressively worsened. There was no history of trauma to the head or neck. Neurological examination revealed right hemiparesis (grade 4/5), with particular weakness in wrist extension (grade 3/5) and diminished sensation to pain and temperature on the left side below the T1 sensory dermatome. Cervical spine X-rays showed mild

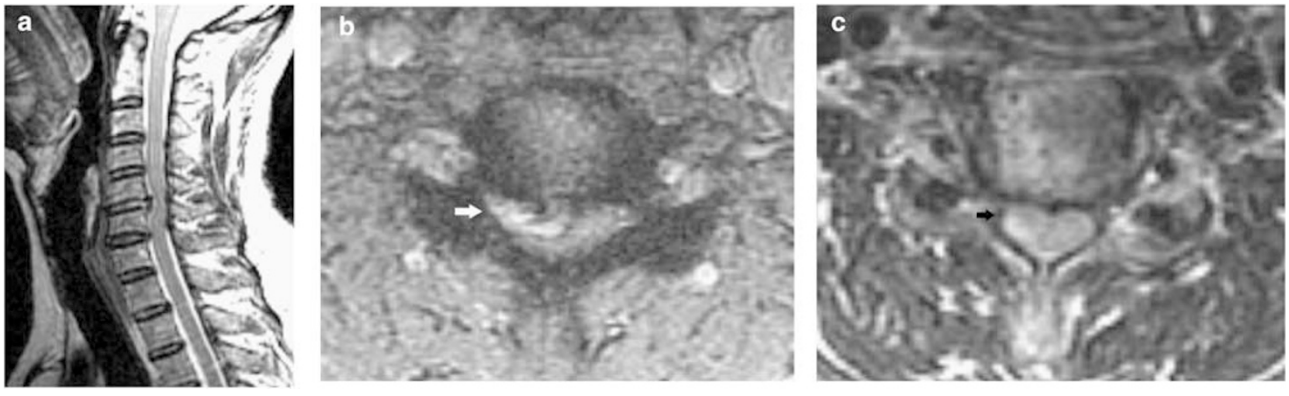

Figure 1 Preoperative T2-weighted sagittal (**a**) and axial (**b**) MRI in Case 1 revealing a large C5–C6 disc herniation, compressing the right side of the spinal cord (arrow). T2-weighted axial MRI (**c**), obtained 1 month after surgery, showing excellent decompression of the spinal cord (arrow)

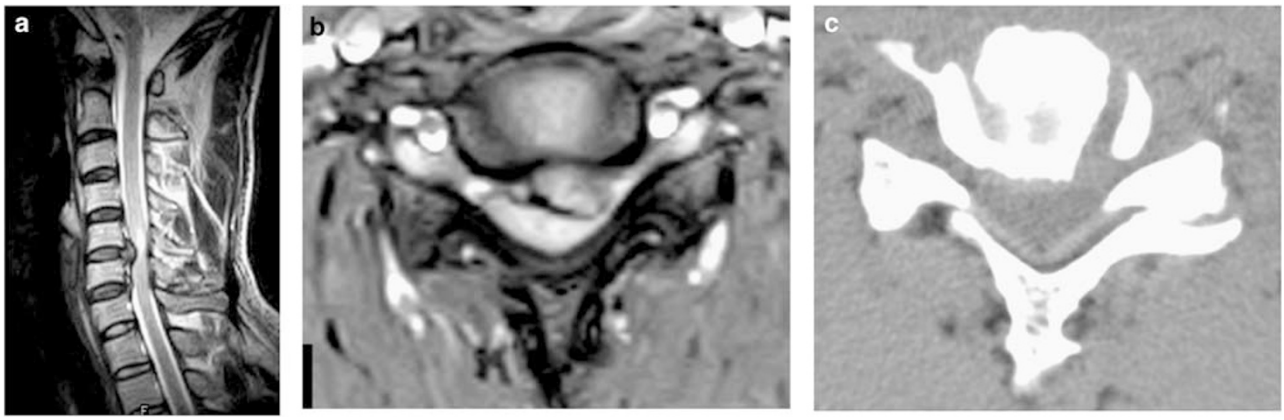

Figure 2 Preoperative T2-weighted sagittal (**a**) and axial (**b**) MR images in Case 2 demonstrating a large left paramedian disc herniation at the C5–C6 level, with ipsilateral severe spinal cord compression and high-signal intensity within the spinal cord adjacent to the herniated disc. Postoperative axial CT image (**c**) revealing complete decompression of the spinal cord through a small tunnel

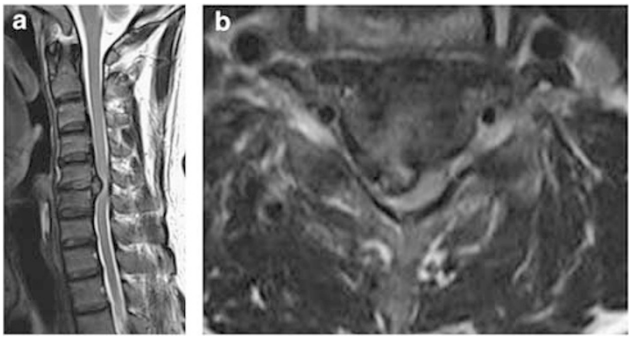

Figure 3 Sagittal T2- (**a**) and axial T1-weighted (**b**) MR images in Case 3 demonstrating a large herniated disc causing significant compression of the right side of the spinal cord at the C5–C6 level

narrowing of the disc space at C5–C6. MRI of the cervical spine showed C5–C6 large herniated disc causing a severe right hemicompression of the spinal cord (Figure 3a and b). Spinal cord decompression was achieved by removal of large disc material using right anterior foraminotomy. Motor weakness and sensation were improved completely at 1 month after surgery. Postoperative CT revealed sufficient decompression of the spinal cord. Cervical spine roentgenogram at 1-year follow-up shows the preserved stability, despite narrowing of the intervertebral space at C5–C6.

Discussion

BSS is characterized by ipsilateral loss of motor function, proprioception and vibratory sense, combined with contralateral loss of pain and temperature sensation. Complete hemisection with classic clinical features of pure BSS, is rare and incomplete hemisection causing BSS plus other signs and symptoms is more common.[4] Spasticity and hyperactive reflexes may not be present in the case of an acute lesion. It occurs most often after traumatic injury to the spinal cord.[1] Cervical disc herniation has rarely been reported as a cause of BSS. As the first three cases of BSS caused by cervical disc herniation reported by Stookey[12] in 1928, we could find only 26 cases of BSS caused by cervical disc herniation in the literature.[1–3,5–16] (Table 1). Among these 29 cases including ours, there were 20 male subjects and nine female subjects, whose ages ranged from 25–73 years (mean 45.6 years). The disc herniation involved one interspace in 27 cases and two contiguous interspaces in two cases. The disc herniation was at C2–C3 in two cases, at C3–C4 in three cases, at C4–C5 in five cases, at C5–C6 in 16 cases and at C6–C7 in five cases. There were 10 cases of intradural herniation and 19 cases of extradural herniation. Intradural disc herniation is very rare and accounts for less than 0.3% of all disc herniations and only 3% occur in the cervical region.[6] BSS was observed in 10 of these 17 cases of cervical intradural disc herniation.[11] With respect to the treatment, all patients underwent surgery after diagnosis. Six patients were treated by laminectomy or hemilaminectomy, three patients by anterior discectomy without interbody fusion, 14 patients by anterior discectomy with interbody fusion, two patients by anterior corpectomy and interbody fusion, one patient by anterior discectomy with interbody fusion and laminectomy and our own three patients by anterior foraminotomy. The prognosis of functional motor recovery were favorable in most cases, although minor residual deficits sometimes were remained. The outcomes of the cases of extradural disc herniation were better than those of the cases of intradural disc herniation. Complete recovery occurred in 10 of the 19 extradural cases and three of the 10 intradural cases. Interestingly, we observed the decreased sensation of pain and temperature at the dermatome below the level of compression in case 1. This discordance of sensory level with the injury level on radiological study has been reported in BSS by cervical disc herniation.[1] This can be explained by the fact that the spinothalamic tract crosses the midline of the spinal

Table 1 Summary of cases of Brown–Sequard syndrome caused by cervical disc herniation reported in the literature

Authors and Year	*Age (years), Sex*	*Level*	*Initial Symptoms*	*Surgery*	*Intradural or extradural*	*Outcome*
Stookey (1928)	44, M	C3–C4	Left leg weakness, neck pain	Lam	Extradural	?
	52, M	C5–C6	Neck pain	Lam	Extradural	?
	68, M	C6–C7	Neck pain	Lam	Extradural	?
Dürig *et al* (1977)	52, F	C5–C6	Thoracic pain	Lam	Intradural	Mi, Si
Roda *et al* (1982)	43, M	C6–C7	Thoracic pain	Lam	Intradural	Mi, Sc
Eisenberg *et al* (1986)	25, M	C5–C6	Left arm and neck pain	Lam	Intradural	Mi, Si
Schneider *et al* (1988)	50, F	C5–C6	Left leg numbness, neck pain	AD	Intradural	Mi, Si
Sprick *et al* (1991)	49, F	C6–C7	Right. arm and thoracic pain	ADF	Intradural	Mi, Si
Finelli *et al* (1992)	28, F	C5–C6	Bilateral hand and rt leg numbness	ADF	Extradural	No change
	61, M	C6–C7	Left hand numbness	AD	Extradural	CR
	46, F	C4–C5, C5–C6	Bilateral hand numbness	AD	Extradural	CR
Rumana *et al* (1996)	56, F	C4–C5	Left leg numbness	ADF	Extradural	CR
Antich *et al* (1999)	73, F	C2–C3	Neck pain	ADF	Extradural	CR
Kohno *et al* (2000)	33, M	C4–C5	?	ADF	Extradural	CR
	31, M	C5–C6	?	ADF	Extradural	Mi, Si
	38, M	C5–C6	?	ADF	Extradural	Mi, Si
	45, F	C4–C5, C5–C6	?	ADF	Extradural	Mc, Si
	34, M	C3–C4	?	ADF	Extradural	Mi, Si
Börm *et al* (2000)	40, M	C5–C6	Left arm pain, bilateral leg sensory disturbance	ADF	Intradural	CR
Clatterbuck *et al* (2000)	40, M	C4–C5	Neck pain and right arm pain	ADF + Lam	Intradural	Mi, Si
	52, F	C3–C4	Right leg weakness	ADF	Intradural	CR
	32, M	C5–C6	Right arm and leg weakness	ACF	Intradural	CR
Iwamura *et al* (2001)	45, M	C6–C7	Neck pain and stiffness	ACF	Intradural	Mc, Si
Kobayashi *et al* (2002)	64, M	C5–C6	Left leg paresthesia	ADF	Extradural	CR
	39, M	C2–C3	Neck and right shoulder pain	ADF	Extradural	CR
Mastronardi *et al* (2004)	36, M	C5–C6	Neck and left arm pain	ADF	Extradural	Mc, Si
Present case	56, M	C5–C6	Right shoulder pain	AF	Extradural	CR
	47, M	C5–C6	Left shoulder pain, left hand numbness	AF	Extradural	CR
	45, M	C5–C6	Right shoulder pain	AF	Extradural	CR

ACF, anterior corpectomy and fusion; AD, anterior discectomy; ADF, anterior discectomy and fusion; AF, anterior foraminotomy; c, complete resolution; CR, complete recovery; i, improved; Lam, laminectomy; M, motor function; S, sensory function; ?, details not reported

cord one to two segments rostral of entry level. Thus, the sensory deficit of pain and temperature on the contralateral side is likely to be demonstrable beginning at a dermatome a few levels below the cord injury. Additionally, the degree and level of sensory deficit may be related to the severity of the cord compression and the individual differences of distribution of anterior spinal artery.[1]

In our cases, we performed anterior foraminotomy and obtained good decompression of the spinal cord. Anterior discectomy followed by bone fusion, with or without a cervical plate, is widely used in the surgical treatment of cervical disc herniation. The main drawback of this procedure is that the functioning motion segment is lost, followed by an acceleration of degenerative changes at adjacent segments. The rate of symptomatic adjacent-segment disease following anterior cervical fusion ranges from 8 to 25%.[17,18] Cervical canal stenosis is the most common finding in the cervical spine X-rays of patients with BSS produced by cervical disc herniation.[1] In patients with cervical spondylotic radiculopathy or myelopathy, anterior foraminotomy leads to an excellent outcome without bone fusion and eliminates the risk of consequent adjacent-segment disease. Anterior foraminotomy provides direct elimination of the compressive pathological lesion, whereas preserving the remaining disc as much as possible. Additionally, anterior foraminotomy eliminates the bone fusion or postoperative immobilization.[19] Although the symptoms of BSS were quite severe in our patients, a 5×8 mm hole was sufficient for adequate decompression of the spinal cord and root, resulting in a favorable outcome. Regarding to the spinal instability, mild narrowing of the intervertebral space usually occur long time after foraminotomy. However, this procedure do not severely interfere the solid stability and the spinal motion, especially in foraminotomy of one level.

As a result the ready availability of MRI, the number of contemporary reports of BSS has been increased. As MRI can accurately indicate the presence or absence of a disc herniation, MRI is able to show the hemicord changes, it is spensable for the diagnosis of all patients with BSS. Although BSS is rarely associated with degenerative cervical spine disease, cervical disc herniation should be kept in mind and early evaluation is mandatory, even in the absence of pain or significant spine radiographic abnormalities. Sometimes BSS can be misdiagnosed as a cerebrovascular accident and diagnostic workup can be delayed, because of the similar motor weakness, as shown in our two cases. As serious symptoms can progress rapidly, early accurate diagnosis and immediate surgical treatment should be recommended to prevent serious morbidity.

Based on our cases, we emphasize the need for early diagnosis and prompt surgical intervention for the best chance of functional neurological recovery in cases of BSS associated with cervical disc herniation. Cervical disc herniation should be considered in the differential diagnosis of BSS, even in the absence of the typical symptoms.

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