Brown-Sequard syndrome revealing intradural thoracic disc herniation

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Abstract Brown-Sequard syndrome (BSS) is a rare form of severe myelopathy characterised by a clinical picture reflecting hemisection of the spinal cord. This syndrome is mostly due to a penetrating injury to the spine but many other non-traumatic causes have been described. Intradural thoracic disc herniation (TDH) is one of the rare aetiologies of this syndrome. Despite progress in imaging techniques, diagnosis and treatment remain difficult. We retrospectively reviewed one of the largest reported series of six patients with BSS revealing intradural TDH between 2003 and 2007. There was a marked female predominance and the mean age was 44 years. Before surgery, half of the patients had a severe neurological deficit. The mean duration of symptoms until surgery was 8.5 months (range 0.5–24 months). Spine magnetic resonance imaging (MRI) or spine computer tomography scan showed calcified TDH between T5–T6 and T9–T10. The intradural location of the thoracic herniation was strongly suspected from the clinical data. All the patients underwent posterolateral transpedicular surgery with an operative microscope to open the dura mater. The intradural location of the herniation was overlooked in one case and the patient underwent a second procedure. The dura mater was carefully closed. Two patients' condition worsened immediately after the surgery before slowly improving. All the other patients improved their neurological status immediately after the surgery and

at 12 months follow-up. BSS with TDH on the spine MRI scan may be a warning symptom of the intradural location of the herniated disc. In such cases, spine surgeons are advised to use an operative magnification and to open the dura mater to avoid missing this potentially curable cause of severe myelopathy.

Keywords Brown-Sequard syndrome · Intradural disc herniation · Calcified thoracic disc herniation · MRI · Transpedicular approach · Spinal surgery

Introduction

Brown-Sequard syndrome (BSS) involves corticospinal tract compression resulting in ipsilateral loss of motor function, and interruption of the ascending fibres in the posterior white column resulting in ipsilateral loss of tactile discrimination, and of vibratory and position sensation. Additionally, BSS is characterised by spinothalamic tract dysfunction resulting in contralateral loss of pain and temperature sensation. The syndrome is most frequently observed in association with traumatic injuries to the spinal cord and extramedullary spinal cord tumours [19, 32]. Spine degenerative diseases [23, 28], such as thoracic disc herniation (TDH), are a very rare cause of severe myelopathy. Nevertheless, intradural TDH appears to be more closely linked to BSS than the more common extradural TDH [1].

To the best of our knowledge, our series of six patients is the largest series entirely focused on intradural disc herniation revealed by complete or incomplete BSS reported so far. The goal of this study was to analyse the clinical and physiological consequences of intradural thoracic herniation, and its surgical implications for the spine surgeon.

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Materials and methods

Between January 2003 and March 2007, thirty patients were operated for TDH in our neurosurgery department. Six of the patients (20%) had BSS revealing intradural TDH. These patients were the subjects of the study. We retrospectively reviewed gender, age, neurological signs (Levy score in Table 1), radiological data (CT or MRI), duration of symptoms before diagnosis, level of the herniation, partial or complete intradural location of the herniation during surgery, and immediate and late postoperative outcome. The size of each herniated disc was measured as a percentage of the area of the spinal canal on the axial MRI or CT scan. All the patients underwent postoperative follow-up at 12 months and long-term follow-up (12–62 months) after the operation, with a mean of 37 months.

Results

The patients' data are summarised in Table 2.

There were 4 women and 2 men with a mean age of 44 years (range 34–53 years). The mean duration of symptoms until surgery was 8.5 months (range 0.5–24 months).

Table 1 Levy score

Classification	Symptoms and signs
Grade 0	Normal
Grade I	Walking with assistance
Grade II	Strength greater than gravity
Grade III	Strength less than gravity
Grade IV	Paraplegic (no motor response)

Table 2 Data of patients harbouring intradural thoracic herniation

Case	Age (years)	Sex	Symptoms before surgery (months)	Levy score and preoperative clinical signs	Herniation location	Size of the herniation (% of the area of the spinal canal)	Surgery	Follow-up (months)	Immediate postoperative Levy score	Levy score at 12 months
1	53	M	0.5	Grade III	T9–T10 intradural/small epidural component	60	TPA + HL	62	Grade II	Grade I
2	39	F	1	Grade I Sexual discomfort	T6–T7 intradural/epidural	60	TPA	40	Total recovery	Total recovery
3	43	F	9	Grade III	T6–T7 intradural	50	TPA + HL	44	Grade IV	Grade II
4	50	M	12	Grade II	T8–T9 intradural	50	TPA	12	Grade I	Total recovery
5	45	F	6	Grade II Headache	T5–T6 intradural	60	TPA	26	Grade I	Grade I
6	34	F	24	Grade III Intercostal neuralgia	T6–T7 intradural/epidural	60	TPA	38	Grade IV	Total recovery

TPA transpedicular approach, HL hemi-laminectomy

Preoperative symptoms were severe BSS with grade III on the Levy score in 3 patients and moderate BSS in 3 patients, with grade II or I on the Levy score. Spine MRI showed TDH between T5–T6 and T9–T10. The herniated disc accounted for an average of 57% of the spinal canal space (range 50–60%). All the patients underwent spinal surgery in the prone position. A unilateral posterolateral approach with partial removal of the facet and pedicle of the vertebral body inferior to the herniation level was performed. In two patients, an additional hemi-laminectomy was performed. The dura mater was opened under operating microscope. The disc herniation was completely or almost completely intradural in four cases and both intradural and epidural in two cases. One patient (case 1, Table 2) underwent two procedures after the intradural location of the herniation was overlooked during the first procedure 2 weeks earlier. The dura mater was closed tightly whenever possible. If not, it was sealed with muscle and biological glue.

There was no operative mortality.

Two patients (cases 3 and 6) got worse immediately after the surgery (from grade III to grade IV) before slowly improving and eventually achieving a better Levy score at 12 months follow-up (one patient fully recovered and one patient was grade II). All the other patients improved their neurological status immediately after surgery and at 12 months follow-up.

Illustrative cases

Case 1

This 53-year-old male had a history of chronic thoracic back pain. He experienced sudden backache and right leg weakness while driving his car. On examination, he had

severe right BSS (grade III on the Levy score) with a sensitive left T10 level.

The spine MRI (Fig. 1) demonstrated a right bulging calcified disc herniation at the T9–T10 level with spinal cord compression. A right posterolateral transpedicular approach with laminectomy was performed. A relatively small calcified epidural herniation, attached to the dura mater, was removed under operating microscope. There was a spontaneous tear in the right lateral side of the dura mater at the location of the herniation. It was sealed with glue. The patient failed to improve postoperatively. A postoperative MRI and CT scan (Fig. 2) revealed the same bulging calcified disc at the same T9–T10 level. Intradural location of the thoracic herniation was then suspected. The patient underwent a second procedure via the same approach. The previous lateral tear of the dura mater was enlarged under operating microscope and a bulging calcified intradural herniation embedded in the right lateral part of the spinal cord was totally removed. The dura was stitched up under microscope and a muscle graft and glue were used. The patient slightly improved postoperatively

(grade II). Twelve months after, the patient improved to grade I. MRI revealed only a persistent T2-weighted hypersignal of the spinal cord at T9–T10, similar to the preoperative MRI.

Case 2

This 39-year-old active female had a long history of thoracic backache. Three years earlier, she had experienced sudden and transient left leg weakness after her fourth delivery. She was admitted for a 1-month history of right BSS with slight right leg weakness (grade I on the Levy score) and left side hypalgesia and numbness. She also complained of urinary and sexual discomfort.

Spine MRI and CT scan demonstrated a right bulging calcified disc herniation at the T6–T7 level with spinal cord compression (Fig. 3).

A right posterolateral transpedicular approach was performed. Using the operating microscope, a large calcified intradural and extradural disc herniation embedded in the spinal cord was totally removed with lateral opening of the

Fig. 1 a, b Preoperative spinal T2-weighted MRI: bulging thoracic disc herniation at the T9–T10 level with severe compression of the spinal cord

Fig. 2 a Postoperative spinal CT scan showing a calcified thoracic disc herniation at the same T9–T10 operated level. **b** T2-weighted MRI showing this very large thoracic disc herniation at T9–T10 with severe deformation of the spinal cord

Fig. 3 **a, b** T2-weighted MRI revealing a midline to right-sided bulging thoracic disc herniation that compressed the ventral cord at the T6–T7 level. **b** Spinal CT scan showing the herniation is calcified

dura mater. The dura mater was sealed with a fat graft, and surgical and biological glue.

The patient fully recovered from her right leg paresis. Her bladder and sexual functions improved considerably.

Discussion

Brown-Sequard syndrome is an incomplete spinal cord lesion characterised by a clinical picture reflecting hemisection of the spinal cord. It was first described in the 1840s after Dr. Charles Edouard Brown-Sequard sectioned half of the spinal cord in animals [7–9].

Complete hemisection with classic clinical features of pure BSS is rare, whereas incomplete hemisection causing BSS plus other signs and symptoms is more common [26]. It is mostly caused by penetrating spine injuries [19, 32]. Non-penetrating aetiologies include blunt trauma [14], spine tumours or metastasis [3], epidural haematomas after anticoagulant therapy and thrombolytic therapy [6, 11]. Other causes include demyelinating diseases [5] or spinal cord herniation [10, 17]. Spine degenerative diseases, such as cervical disc herniation or TDH [23], are a rare cause of BSS [28].

Most TDH are asymptomatic and it has been estimated that 15–20% of the population have incidental TDH visible on MRI [4, 33, 34]. Symptomatic TDH are very rare and their surgical treatment amounts to <0.5% of all disc operations [1]. Most cases occurred between the third and the fifth decades of life with a marked female predominance, perhaps due to calcium–phosphorus metabolism status [20, 35]. Our series is consistent with the literature as regards gender predominance.

Clinical signs of symptomatic TDH vary considerably from back pain to profound myelopathy or BSS [1]. The incidence of BSS seems to be higher in patients with intradural TDH [1, 16]. This entity represents <7% of all

TDH cases in the large series reported in the literature [35]. In our series, 20% of the patients with operated TDH had an intradural TDH, which is far higher than the literature data. Intradural TDH may be overestimated due to the small number of our patients with TDH or due to the bias of our recruitment of severe myelopathy with TDH.

Most intradural disc herniations are found in the lumbar spinal canal. Only five percent are found in the thoracic canal. In our series of 30 patients with TDH, the six patients with intradural herniated disc had BSS, whereas BSS was not observed in the remaining 24 patients with extradural herniated discs. In most of the cases, the intradural location was highly suspected from the MRI finding, and confirmed during surgery due to a dural tear close to the disc herniation or a CSF leakage after removal of the extradural portion of the disc herniation. Case 1 is highly representative of the misdiagnosis of an intradural TDH. The patient had severe BSS with a very large calcified disc herniation. During the first procedure, we removed only the extradural component. The dura tear was small and hidden and the patient did not improve after surgery. The immediate postoperative MRI showed almost the same very large TDH. We checked that the surgery was performed at the correct level. The almost completely intradural location of the herniation was then highly suspected and confirmed during a second procedure with the opening of the dura mater.

Anatomically, intradural disc herniation corresponds to penetration of the disc material, which is often calcified, into the subarachnoid space through a tear in the posterior longitudinal ligament and dura mater [12, 36]. The pathogenesis of intradural TDH has not been fully elucidated. But it seems to be related to adherence between the ventral part of the thoracic dura mater and the posterior longitudinal ligament. For some authors [1, 15], a history of traumatic injury may cause chronic adhesion between the

ligaments and dura, resulting in rupture through the dura as a secondary event. Like other authors [2, 27], we find this aetiology debatable. The adhesion could be caused by osteophytes or disc protrusion which is often calcified [16, 20, 30, 35]. In our series, all the TDH were calcified and occupied more than 50% of the cross sectional area of the vertebral canal. Calcification is an important consideration since more than 60% of this hard herniation was found to be intradural or incorporated into the dura mater in a recent series [20]. Moreover, there is a strong correlation between the size of the herniated disc and whether or not it is symptomatic [4, 22].

The clinical implications of intradural TDH may be severe due to the anatomic features of the thoracic spinal cord. Indeed, the spinal canal in the thoracic region is small and mostly occupied by the spinal cord with a minimal subarachnoid space [29]. Furthermore, the thoracic spinal cord has a weak blood supply often dependent on a single artery [13]. Intradural herniation may damage the spinal cord through a combination of mechanical injury and vascular insufficiency [18]. This could explain the higher incidence in the literature and in our series of severe neurological signs with complete or incomplete BSS or paraplegia in patients with intradural location. Arce et al. [1] reported that 70% of patients had different degrees of symptoms of spinal cord compression or BSS at diagnosis of intradural disc herniation. The lateral hard or calcified disc protrusion could act as a knife or a stab wound into the medulla and cause damage to the ascending sensory pathways and the descending pyramidal tract, subsequently leading to hemidysfunction.

The intradural location of TDH may be suspected on T2-weighted spine MR images. An obtuse angle with an enlarged subarachnoid space around the herniation may suggest its intradural location [21]. But no image is specific and the intradural location of the herniation can only be confirmed during the operation. It is the combination of a severe neurological deficit such as BSS and a bulging calcified TDH on imaging that may lead to suspicion of partial or total intradural location of the disc herniation.

The surgical approach for TDH has changed over the years and is now divided between transthoracic and posterolateral surgery, which mostly depend on the spine surgeon's policy. Because of its poor outcome, laminectomy alone has practically been abandoned [1]. Like other authors [24, 25, 31], we adopted a posterolateral transpedicular approach with partial removal of the facet and pedicle of the vertebral body inferior to TDH. To avoid mistaking the operative level, a skin tattoo of the spinous process of the vertebra inferior to the herniation level was done the day before the surgery. A fluoroscopic arm was used just before the skin incision to check the level of the

herniation again. Under operative microscope, the pedicle was drilled and the dura mater was opened. The dural wound was carefully closed with a fat or muscle graft and biological glue after removal of the intradural herniation, since dural tears lead to a high risk of cerebrospinal fluid fistula. Moreover, a dural tear may lead to spinal cord herniation [17], followed by secondary neurological deterioration. Except for one patient who underwent a second procedure 2 weeks after the first one, the intradural position of the disc herniation was found for all the patients during the surgery.

According to our experience, when there is a combination of BSS and a large radiological calcified herniation we recommend opening the dura mater, if the correct level is proved, when there is a discrepancy between the size of the resected herniation and its radiological size. Moreover CSF leakage or a dural tear close to the herniation are good indicators of an intra-dural part of the herniation.

We used the posterolateral transpedicular approach which was effective for all the patients in our series, even when the thoracic herniation was almost median. Two patients got worse immediately after surgery (from grade III to grade IV) before slowly improving and eventually achieving better neurological status at 12 months follow-up. In these two cases the initial neurological status was poor (grade III) and the calcification of the intradural herniation was very extensive. The neurological worsening is may be due to the surgical traumatism of the spinal cord even if the manoeuvres to separate the herniation from the spinal cord were soft and strictly centrifugal, under surgical microscope. A spinal cord oedema may have occurred following the surgery, resulting in temporary worsening in patient with a very fragile compressed spinal cord.

It is difficult to predict the surgical result even in patients with a poor clinical condition. One of the patients [case 6] has unpredictably fully recovered after 1 year. This patient was the youngest and the rehabilitation was intensive but not specific. Additionally he was managed post operatively by corticosteroid medication during 1 week although we cannot conclude on the role of this medication for the neurological improvement. This very interesting result confirms us firstly to propose a surgical procedure even if the initial neurological status is poor, and secondly it confirms us not to be pessimistic if the patient got worse post operatively.

All the other patients improved their neurological status immediately after surgery and at 12 months follow-up.

Conclusion

Spine surgeons examining patients with BSS related to a large calcified TDH on an MRI or CT scan should be aware

of the possible partial or complete intradural location of the herniated disc. We therefore recommend intradural exploration during surgery, which may be facilitated through the use of an operative magnification.

Conflict of interest statement None of the authors has any personal or institutional conflict of interest.

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