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CASE REPORT Against the odds: extraordinary recovery from complete cauda equina syndrome following L3 fracture. Time still matters

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INTRODUCTION: Cauda equina syndrome secondary to lumbar fracture is a relative rare event. Although it is usually considered as an emergency, there is still controversy in the literature regarding the optimal timeframe of surgical intervention in complete spinal cord and cauda equina injuries.

CASE PRESENTATION: We report a case of a 24-year-old victim of a road traffic accident admitted with an L3 fracture causing complete cauda equina syndrome, who underwent early surgery within 12 h and made an extraordinary recovery (from AIS A to E).

DISCUSSION: Although the timing of surgery in complete traumatic spinal cord injury and cauda equina syndrome remains controversial, this case highlights the importance of early surgical intervention even in complete injuries.

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INTRODUCTION

Cauda equina syndrome (CES) is a severe neurological disorder resulting from an injury to the cauda equina and causing polyradicular symptomatology, including lower limbs and sphincter deficits. CES not only affects the physical well-being of the individual, but can also have psychological consequences that may have long-lasting effect and lead to poor functional outcome. The etiology of the CES is varied and includes disc herniation,^{1,2} neoplastic lesions³ or trauma to the lumbar vertebrae.^{4,5} However, lower lumbar vertebral trauma as a direct cause of CES is not very common.⁵

There is still controversy in literature regarding the optimal timeframe of surgical intervention in CES. Most of the studies are retrospective with inconsistent follow-up to determine the statistical significance of surgical timing in the functional recovery. However, CES is generally recognized as a spinal surgical emergency, and the decision to decompress early can produce good functional recovery, helping the patient to return to a normal life after suffering a potentially catastrophic neurological insult.

We present a case of a patient who suffered CES caused by a traumatic L3 vertebral body fracture, following a road traffic collision. This case report describes a significant functional recovery with an exceptional improvement in the American Spinal Injuries Association (ASIA) Impairment Scale from A (complete injury—no sensory or motor function preserved in sacral segments S4–5) to E (normal sensations and motor functions in all segments).

MATERIALS AND METHODS

A 24-year-old patient was admitted to the Major Trauma Centre following a road traffic collision. Initial trauma screen demonstrated paraplegia with sensory level at L3, no voluntary anal contraction or sensation to deep anal pressure, as well as urinary retention. The International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI) were used to identify the ASIA Impairment Scale (AIS) and neurological level, and it was found to be L2 AIS A. Computer tomography (CT) of the spine showed a superior

endplate fracture of the third lumbar vertebra (AOspine type A3), with a retropulsed fragment lying within the spinal canal and causing >90% stenosis (Figure 1).

The patient developed some paraesthesia in both lower limbs ~ 8 h following injury, but with persistent numbness and no improvement in motor, bladder or bowel function. A decision was made for an early surgical intervention, with a hope to give the patient the best chances of neurological recovery. To avoid any further delays to treatments, a decision was made to proceed with surgery on the basis of the CT scan results, without performing further imaging (such as, MRI). Surgery was in fact carried out within 12 h from the original trauma. A posterior fusion and decompression from L3 to L5 was performed. Distraction and lordotising maneuvers (ligametotaxis) produced a good reduction of the retropulsed fragment, as confirmed on post-operative CT scan (Figure 2).

Following surgical treatment, the patient underwent specialist neurorehabilitation on the rapid access Acute Rehabilitation Ward and, 7 days post injury, distal power (ankle dorsiflexion) improved from 0 to 2 on the Medical Research Council (MRC) grade for motor function. He was then transferred to a specialist Spinal Cord Injury Rehabilitation Centre, where he continued his rehabilitation for another 2 months.

RESULTS

On discharge from the Spinal Cord Injury Centre, the patient was able to walk unaided with normal bladder, bowel and sexual functions. X-rays of the lumbosacral spine, performed 6 months postoperatively, showed good position of the pedicle screws and normal alignment of the lumbar spine (Figure 3). At his last follow-up in the Rehabilitation Outpatient Clinic, 2 years postoperatively, the patient was independent in all his activities of daily living, with normal bladder and bowel functions. There was no complaint of any back pain, sleep disturbances or symptoms of post-traumatic stress disorder. The ISNCSCI

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Figure 1. CT scan sagittal (a) and axial (b) showing L3 superior endplate fracture with retropulsed fragment causing severe canal stenosis.



Figure 2. Post-operative sagittal (a) and axial (b) CT scan showing satisfactory reduction of the fracture.

improved significantly, with final AIS graded as E (Figure 4). He described some occasional paraesthesia in a patchy area around the left shin, but without any objective residual neurological deficit (normal power, light touch and pinprick sensation in all areas). Six months post surgery, he was able to return to work and also play golf.

DISCUSSION

The cauda equina is a bundle of spinal nerves roots, consisting of the second through fifth lumbar nerve pairs, the first through fifth sacral nerve pairs and the coccygeal nerves, all of which arise from the lumbar enlargement and the conus medullaris of the spinal cord. The cauda equina provides sensory and motor innervation of most of the lower limbs, the pelvic floor and the sphincters. Therefore, in a CES, multiple signs of sensory, motor and sphincter dysfunction may appear, including saddle anesthesia, motor weakness of the lower extremities, bladder and bowel dysfunction.

The most common cause of CES reported in the literature is the central disc prolapse⁶ and can occur in 2–6% of all the disc herniations.^{7,8} However, it may also be caused by space-occupying lesions (for example, hematomas, abscesses, malignancies) and



Figure 3. Six months post-operative standing X-rays, showing good alignment and metalwork positioning.

trauma (blunt or direct injury). The CES may produce a variety of signs and symptoms depending on the involved nerve roots. The standard method of assessment of neurological status of an individual with suspected spinal injury is the American Spinal Injury Association (ASIA) Neurological Classification. The five categories of the AIS are A, B, C, D and E. AIS A, also known as complete injury, is characterized by no voluntary anal contraction, no sensation to deep anal pressure and no sharp/dull discrimination to light touch within the S4–5 dermatomes. In effect all neurological transmissions have ceased to exist below the level of injury. On the other hand, AIS E indicates normal sensory, motor and sphincter functions.

Although axonal regeneration and growth is inhibited in the CNS, peripheral nerves still have potential for regeneration and therefore for functional recovery. According to the Seddon system, peripheral nerve injury can be classified in neuropraxia, axonotmesis and neurotmesis. The neuropraxia, usually caused by compression or ischemia, is characterized by intact nerve structure and impaired axonal transport. Axonotmesis occurs when the axons and myelin sheets are completely interrupted, but the supporting structures (including the endoneurium) are intact. Neurotmesis occurs when the nerve is completely transected with loss of continuity. There is therefore a substantial difference in CES with incomplete injury of the nervous fibers, as usually in disc prolapses and post-traumatic subtotal spinal stenosis, and complete neural injury, as in penetrating injuries or in fractures with bony fragments causing direct nerve transection. In neuropraxia and axonotmesis caused by subtotal spinal stenosis, a functional recovery is usually possible if the anatomy of the spinal canal is restored, and the cauda equina is surgically decompressed. However, the optimal timeframe for surgical intervention in CES has been well debated in the past and is recognized as the most critical issue regarding CES management. It also has medico-legal implications. Qureshi and Sell⁹ in 2007 published a prospective longitudinal inspection cohort study of 33 patients to look at the outcomes at 3 months and 1 year with regard to the timings of the surgical decompression. The patients were divided into three groups based on the timing from symptom onset to surgery (< 24, 24–48 and > 48 h). They found no statistically significant difference in outcome between these three groups. However, patients who were continent at presentation had better outcome compared with the patients who were incontinent. Although the findings of this study seem to be very convincing, the results should be interpreted with caution. The drawback of this study is the small sample size, which could easily skew the results. Other studies have also failed to show any correlation between the timing of the surgical intervention and the outcomes in CES.^{10,11}

From the other hand, arguments for early decompression have been supported by the theory by Lee and Wolfe¹² that nerve compression will lead to disruption of the nerve-blood barrier. They theorized that proteins acting as antigens produced an autoimmune reaction, leading to a cycle of nerve Wallerian degeneration. Other authors, such as Sekiguchi et al.¹³ and Delamarter *et al.*¹⁴ supported the theory by Lee and Wolfe.¹² They also identified that demyelination and degeneration is related to the degree of compression,^{8,13} and there is a risk of complete nerve root atrophy, at the level of constriction, in severe and prolonged compression.¹⁴ These findings have greater implication on the timing of the surgical decompression in CES. A recent review of the CES suggested that the time course of the development of CES helps to determine the prognosis with regard to functional outcome, with the time since symptom onset inversely correlating with the likely return of function.¹

A retrospective study by Shapiro¹⁶ confirmed a better functional outcome in the group of patients who received early surgical intervention (< 24 h).

The most convincing evidence of functional recovery with early surgical intervention was shown by Ahn *et al.*¹⁷ in a meta-analysis of surgical outcomes for CES secondary to lumbar disc herniation. It included 42 studies and a total of 322 patients analyzed after the surgery. They found a significant difference in outcomes of the patients who were treated within 48 h as compared with that in the patients who were treated >48 h after the onset of the symptoms of CES, with greater improvements in those who were treated within 48 h. In 2001, Busse *et al.*¹⁸ published a series of 14 patients who underwent surgical decompression for CES. There was increased incidence of pain and impaired social physical functions in patients who underwent delayed decompression.

However, most of the above-mentioned studies have assessed surgical outcomes in CES caused by lumbar disc prolapses. Thongtrangan *et al.*⁵ assessed surgical outcomes in a series of 17 consecutive patients with CES following lower lumbar fractures. They concluded that surgical decompression and stabilization within 48 h offers a good chance of neurological recovery. However, in their study patients who presented with complete CES failed to show any neurological improvement despite early decompression and stabilization of the fracture.

To our knowledge our case is unique; in fact a recovery from a complete injury (AIS A) to full normal function (AIS E) is exceptional. On this basis, we believe that early surgery in complete injuries, which remains still controversial, can sometimes produce exceptionally good outcomes and therefore should be still supported.

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Figure 4. (a) Pre- and post-operative (b) ASIA scoring charts, showing improvements from AIS A to AIS E.

CONCLUSIONS

Complete CES following lumbar spine fractures is not very common. We have reported an exceptional recovery (AIS A to E) from a complete post-traumatic CES operated within 12 h. Although the severity of the initial injury is a contributing factor for neurological recovery, our case supports the argument for early surgery, even in complete injuries.

COMPETING INTERESTS

The authors declare no conflict of interest.

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