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Case Report

Rehabilitation in a case with cauda equina syndrome after spinal manipulation: A long and difficult process

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ABSTRACT

In this case report, the rehabilitation process of a 38-year-old female patient who developed cauda equina syndrome due to spinal manipulation for the treatment of lumbar disc herniation was presented. The aim was to discuss the outcomes of rehabilitation after cauda equina syndrome, as well as the challenges associated with it. Despite the difficulties experienced during the rehabilitation process, a comprehensive multidisciplinary rehabilitation program is beneficial in terms of gaining independence and returning to daily activities, social life, and work for patients with cauda equina syndrome.

Keywords: Cauda equina syndrome, intervertebral disc herniation, rehabilitation, spinal manipulation.

Cauda equina syndrome is a clinical condition that occurs due to the compression of the cauda equina within the lumbosacral spinal canal. Sensorimotor losses at lower extremities, sphincters dysfunctions, decreased sexual functions, and saddle anesthesia are typical clinical signs of cauda equina syndrome.[1] The annual incidence of cauda equina syndrome is reported to be 5 to 10 per million.[2] Intervertebral disc herniation, often at the L4-5 and L5-S1 levels, is the most common cause of cauda equina syndrome.[3] Spinal stenosis, tumors, epidural hematoma and abscess, iatrogenic causes, and trauma are other causes.[1] Spinal manipulation has also been reported as a rare cause of cauda equina syndrome in patients with or without prior lumbar disc herniation.[4-6] Spinal manipulation should not be performed in the presence of unstable fractures or spondylolisthesis, local primary or metastatic bone tumors, spinal cord tumors, osteomyelitis, Paget's disease, severe osteoporosis, progressive or abrupt neurological deficits, inflammatory phases of rheumatological diseases, or uncontrolled bleeding disorders.[7]

Cauda equina syndrome is an emergency. Surgical decompression is the main part of early treatment. It has been shown that interventions in the first 48 h yield better results than the later ones.[2] A meta-analysis indicated that some patients with a long-term history of cauda equina syndrome have persistent bowel, bladder, and sexual dysfunctions, as well as sensory and motor losses.[8] After the surgical intervention, a multidisciplinary rehabilitation approach is key to restoring function. Progressive strength exercises, stretching exercises, electrotherapy, and gait training using robotic devices and a treadmill all play key roles in improving motor functioning.[9] Dietary and lifestyle change, voiding and defecation maneuvers, catheterization, timed voiding and defecation, pharmacological therapy, biofeedback, and surgical treatments are parts of bladder and bowel management.[10,11] Although the mechanism of action is not well understood, sacral nerve, pudendal nerve, and spinal cord stimulation may improve bladder and bowel function, sensorimotor deficits, and pain secondary to cauda equina syndrome.[12-14]

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This case was presented as a poster at the 8th Medical Rehabilitation Congress in Ankara, Türkiye, between November 9-13, 2022.

Short- and long-term outcomes of patients with cauda equina syndrome were previously reported in the literature.[4,8] However, studies exploring the rehabilitation process and outcomes are limited. In this case report, we aimed to discuss the outcomes of the rehabilitation period, as well as patient expectations and challenges associated with this period.

CASE REPORT

A 38-year-old female patient was admitted to the emergency department for low back pain, urinary retention, bowel incontinence, and muscle weakness in the lower extremities. The patient had a history of low back pain for 10 years and was previously diagnosed with lumbar disc herniation. Although surgery was recommended in the past, she preferred spinal manipulation and attended multiple sessions. The symptoms started after the last session, and magnetic resonance imaging of the lumbar spine in the emergency department revealed central disc extrusion at the L5-S1 level (Figure 1). The patients was diagnosed with cauda equina syndrome and had an urgent partial L5 laminectomy, a right S1 foraminotomy, and the removal of sequestered discs. Written informed consent was obtained from the patient.

Two months after the operation, the patient was referred to our physical medicine and rehabilitation clinic for rehabilitation. On physical examination, muscle strength was 3/5 in hip flexors, 4/5 in hip extensors, abductors, and adductors, 3/5 in knee flexors, 4/5 in knee extensors, 3/5 in ankle flexors and extensors, and 4/5 in big toe extensors bilaterally. The patient had hypoesthesia in the right S1 dermatome and anesthesia in the bilateral S3, S4, and S5 dermatomes. Patellar tendon reflexes were normal, but Achilles tendon reflexes were absent. An indwelling urinary catheter was placed due to impaired bladder sensation. The patient also complained of fecal incontinence, and her anal sphincter tone was decreased. The patient was able to walk without assistance but was unable to jog or run.

The following rehabilitation program was recommended for the patient: strengthening exercises, robot-assisted therapy, walking-balance training, neuromuscular electrical stimulation, and analgesic current for an hour, five days a week for 10 weeks.

The bladder function was assessed by urodynamic studies, and it was consistent with a noncontractile detrusor and decreased bladder sensation. The patient was started on clean intermittent catheterization six times a day with fluid restriction. Before each catheterization, voiding was attempted with Valsalva and Crede maneuvers. Spontaneous urination started after four weeks. By monitoring the patient's spontaneous urination, the daily frequency of clean intermittent catheterization was gradually reduced and then discontinued. A timed voiding program was implemented to avoid complications from urinary retention. Postvoid

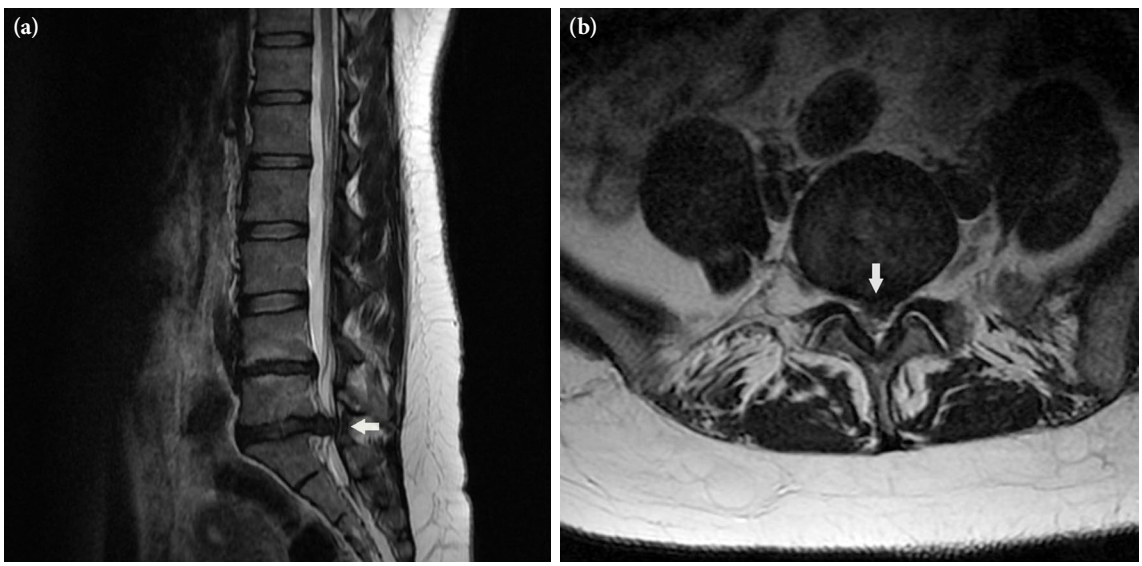

Figure 1. **(a)** Central disc extrusion (white arrows) at the L5-S1 level in the sagittal and **(b)** axial planes of magnetic resonance imaging of the lumbar spine.

residual volume was measured via a bladder scan and was planned to be less than 100 mL. Repeat urodynamic studies yielded similar results as the initial tests.

A bowel rehabilitation program for neurogenic bowel dysfunction was also started. The patients was advised to increase the consumption of fiber-rich nutrients. Timed toilet activity was planned at least once a day, half an hour after meals. As a result of our interventions, the frequency of bowel incontinence decreased to once weekly from at least once daily after six weeks.

The patient was single and expressed concern about how this condition might affect her sexual life after marriage. Possible rehabilitation approaches were also discussed with the rehabilitation team. Biofeedback-guided pelvic floor exercises were added to the rehabilitation program to strengthen the pelvic floor and increase sensory input. Since the patient stated that she started to feel a very slight sensation in that area, additional neuromuscular electrical stimulation was tried, but it was unsuccessful and was discontinued to avoid complications.

The patient was exhausted trying to cope with this situation, accept it, and manage her medical problems simultaneously. As an important component of comprehensive rehabilitation, the patient's psychological state was also evaluated. During the entire rehabilitation process, the patient's morale and mood were monitored by a psychiatrist and psychologist.

The patient was discharged after 10 weeks of rehabilitation. Upon discharge, the patient was able to successfully urinate with the Valsalva and Crede maneuvers. Fecal incontinence also improved. The motor dysfunction in the lower extremities of the patient recovered almost completely. The patient still had saddle anesthesia.

After discharge, the patient was followed up in our outpatient clinic at regular intervals. Additionally, she continued her rehabilitation program in different centers and with different approaches. During follow-up, the patient reported that magnetic field therapy had been applied in another center to improve bladder and bowel functions. However, during the second session of the therapy, the patient experienced a burn to the gluteal region and was operated on twice. The patient was followed up with permanent catheterization due to prone positioning in the peri- and postoperative period. The patient was readmitted to the rehabilitation program in our clinic to review the urinary status and wound care. Six months later, she was able to urinate spontaneously, control bowel continence, and walk comfortably. The patient returned to work as well.

Despite some improvement in the patient's functional status as a result of the rehabilitation interventions, the patient continued to seek additional treatment modalities to fully recover. The patient wished to return to her former healthy state in terms of sensorimotor, bowel, and bladder functions by applying to different centers. However, complications that occurred during the treatments in these centers hindered her hope and effort for a complete recovery. This difficult process helped the patient to better understand the course of her illness.

DISCUSSION

Although intervertebral disc herniation is the most common cause of cauda equina syndrome, there are multiple causes.[3] Spinal manipulation is one of the rare causes of cauda equina syndrome.[1,4] There are few studies in the literature that explore the relationship between spinal manipulation and cauda equina syndrome. Haldeman and Rubinstein[5] presented three patients that suggested a temporal relationship between cauda equina syndrome and spinal manipulation. Tamburelli et al.[4] presented a case with a history of cauda equina syndrome that started just a few hours after spinal manipulation. Similar to our case, their patient also had a history of intervertebral disc herniation. Yang et al.[6] reported a case where cauda equina syndrome symptoms started immediately after vigorous back massage and spinal manipulation. These case reports note that symptoms can appear acutely after manipulation sessions. In our case, cauda equina syndrome had a slower progression. It is the first reported case of the gradual onset of cauda equina syndrome after spinal manipulation. This delayed presentation of cauda equina syndrome in some cases may result in a failure to identify spinal manipulation as a cause of cauda equina syndrome.

Some mild and temporary side effects of spinal manipulation include headaches, fatigue, nausea, dizziness, and local or radiating pain. Severe side effects, such as cauda equina syndrome, stroke, and death, are rarely reported as a result of spinal manipulation.[6] To avoid severe adverse events, spinal manipulation should not be performed in the presence of the already mentioned contraindications. It is not completely contraindicated in individuals

with lumbar disc herniation; however, some individuals experience worsening symptoms following manipulation. In a case series of patients with exacerbation of symptoms of lumbar disc herniation or cauda equina syndrome after spinal manipulation, the majority of patients were over 50 years of age, had a history of recurrent lower back pain with bilateral radicular pain for more than five years, and had imaging studies demonstrating severe disc herniation occupying more than one-third of the spinal canal.[15] Our case also has similar characteristics, except for the patient's age. Spinal manipulation should be carefully planned for patients with these characteristics.

Cauda equina syndrome is associated with multiple long-term complications. In a meta-analysis, 852 patients with cauda equina syndrome who were followed for an mean of 39.2 months were analyzed; 43.3% of the patients had bladder dysfunction, 31.1% had bowel dysfunction, 53.3% had sensory deficits, 38.4% had motor weakness, and 40.1% had sexual dysfunction.[8] In our case, the follow-up period was approximately 31 months. Bowel and motor functions completely recovered, but bladder dysfunction and saddle anesthesia failed to fully resolve. Since there is a taboo about sexuality in the Turkish culture, sufficient information could not be obtained about the progress of the patient's sexual status.

Urinary incontinence significantly interferes with activities of daily living. Although urinary function did not completely improve in our case, urinary continence was achieved with the Crede and Valsalva maneuvers, a timed voiding program, and fluid restriction. At the last follow-up, our patient was independent in the activities of daily living and had returned to work.

Saddle anesthesia is another challenge in the long-term management of cauda equina syndrome. It also leads to sexual dysfunction and is important in determining the choice of treatment modality. Neuromodulation techniques, electrotherapy, and thermotherapy should be applied carefully to the anesthetic area. It may result in burns, as in our cases, and may cause functional regression, as seen in urinary management. Patients should be warned about the complications of these therapies.

In conclusion, patients with cauda equina syndrome experience multiple functional impairments. Cauda equina syndrome causes dependence on activities of daily living and difficulty returning to work. Our case also showed that the rehabilitation of cauda equina syndrome is difficult and requires a long period of rehabilitation. This case also reveals that the loss of functions in patients can be permanent. Rehabilitation programs are useful in increasing independence and restoring functions. After the surgical period, patients should be referred to rehabilitation services. Most of the studies in the literature investigate the short- and long-term results of cauda equina syndrome; however, there is not enough data on the rehabilitation course of these patients. Long-term studies are needed to better understand the rehabilitation course of cauda equina syndrome.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Concept, design, supervision: B.S.T.; Resources, data collection and/or processing, analysis and/or interpretation, writing manuscript, critical review: B.S.T., B.K.T.; Materials, literature search: B.K.T.

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