

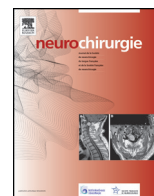


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Clinical case

## Partial cauda equina syndrome after an uneventful minimally invasive microdiscectomy in a patient with Crohn's disease



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### ABSTRACT

Cauda equina syndrome is a serious condition resulting from dysfunction of the lumbosacral nerve roots and characterized by impairment of bladder, bowel, sexual and lower limb functions. We report the case of a 48-year-old woman who had Crohn's disease for more than twenty years. The patient was undergoing immunotherapy with infliximab and developed a partial cauda equina syndrome after an uneventful minimally invasive microdiscectomy (L<sub>5</sub>–S<sub>1</sub>) that completely cured her sciatica. A postoperative magnetic resonance imaging examination showed root clumping but no compressive lesion. We discuss a possible relationship between the cauda equina syndrome and the patient's active Crohn's disease, treatment and surgery.

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### 1. Introduction

Cauda equina syndrome (CES) is a serious condition resulting from dysfunction of the lumbosacral nerve roots. When severe, it is characterized by impairment of bladder, bowel and sexual functions with sensory and motor impairment of the lower limbs. The potential causes are disc herniation, tumor, hematoma or trauma, possibly surgical.

Crohn's disease is an autoimmune condition characterized by chronic and progressive inflammation of the gastrointestinal mucosa.

We report a case of partial CES after an uneventful minimally invasive lumbar microdiscectomy (L<sub>5</sub>–S<sub>1</sub>) for sciatica in a patient with Crohn's disease for a period of approximately twenty years who had been treated with infliximab for the past 15 years.

### 2. Clinical case report

A 48-year-old woman (BMI = 24), diagnosed with Crohn's disease in 1994, presented to our department in March 2015 with left S1 lumbosciatica, which had started suddenly two months earlier.

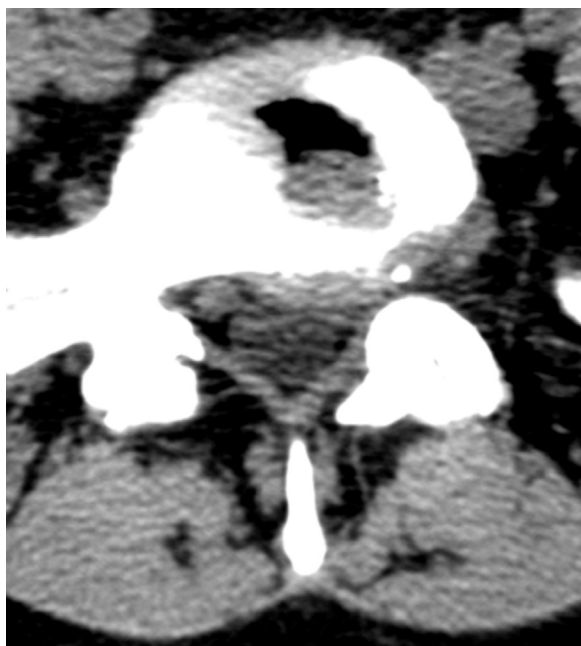
Ninety percent of the pain was in the leg and the degree of pain fluctuated between 8 and 10 on the Visual Analog Scale (VAS). When treated with analgesics, the pain decreased at rest to a VAS score of 4. The patient was able to walk, but with pain and only for short distances. She had attempted acupuncture treatment but with no effect. There was no neurological deficit and patellar and Achilles reflexes were symmetric. Lasègue's test showed increased pain on the left side at 45°. CT-scan showed left disc protrusion at L<sub>5</sub>–S<sub>1</sub> compressing S<sub>1</sub> (Fig. 1) and a grade 1 L<sub>4</sub> retrolisthesis on L<sub>5</sub>. Electromyography showed left L<sub>5</sub>–S<sub>1</sub> peripheral chronic neuropathic dysfunction. In 1994, the patient was diagnosed with Crohn's disease and began treatment with Deltacortril and Salazopyrin. In July 1997, she experienced colonic reactivation with superficial rectal ulceration and was again treated with corticosteroids. In 2000, she started treatment with infliximab (Remicade, Janssen Biotech Inc.), a monoclonal antibody against tumor necrosis factor- $\alpha$ , and was still taking this medication at the time of her consultation in our Department.

A minimally invasive disc herniation resection was performed under microscope via an 18 mm diameter METRx tube (Medtronic Sofamor Danek Inc., Memphis TN). Only the left compressed S1 root was visualized during the surgery. The surgery was uneventful. When the patient awoke, she reported that her sciatica had completely disappeared.

However, nine hours later, she complained of difficulty in urinating and also of perineal hypoesthesia. An urgent magnetic

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**Fig. 1.** Preoperative CT. Axial image demonstrating compression of the left S1 nerve root at the L<sub>5</sub>–S<sub>1</sub> level.

resonance imaging (MRI) was performed, which showed no compressive lesion but revealed clumping of the lumbosacral roots resulting in a “pseudo-cord” appearance (Fig. 2).

On the first postoperative day, the drain was removed and fraxiparine was started. Lower limb motricity was normal and there was a marked improvement in perineal sensitivity.

On the second postoperative day, the patient started to walk and reported no lumbosciatica. The patellar and Achilles' reflexes were present and symmetric. A gradual resolution of the sensory deficit in the perineum was again observed.

Three months postoperatively, the patient still complained of urinary retention with stress urinary incontinence improved by physiotherapy. She also reported difficulty distinguishing the passage of gas and feces.

Four months postoperatively, the patient still had urinary retention. MRI showed normal divergence of the lumbosacral roots but still partial clumping at the L5 level with contrast enhancement of a few roots (Fig. 3).

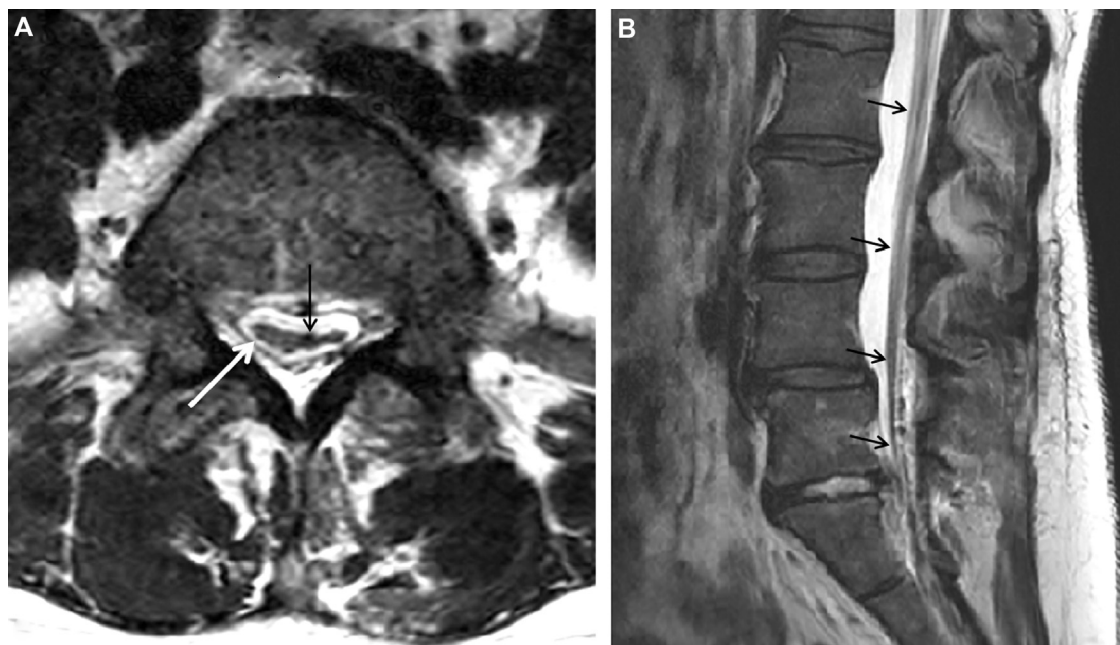
Urological examination, performed 6 months postoperatively, revealed a reduced micturition sensation with hypocontractility of the detrusor muscle, compatible with lower motor neuron dysfunction.

Ten months postoperatively, the patient described significant improvement in urinary and fecal symptoms. Stress urinary incontinence occurred only with sudden effort and fecal incontinence was only present with very loose stools.

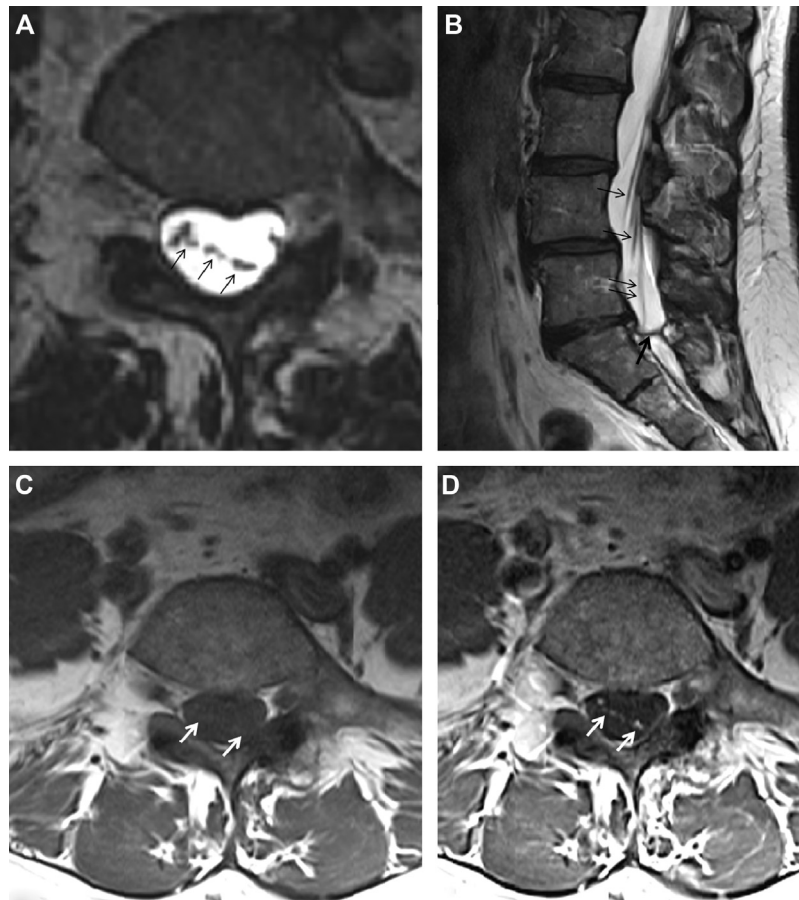
### 3. Discussion

To try to understand the appearance of a partial CES after an uneventful minimally invasive microdiscectomy, during which we visualized only the left S1 root and subsequently followed by resolution of the sciatica, we searched the literature: even if CES is a well-documented complication in patients undergoing lumbar discectomy, occurring with an incidence of 0.08 to 1.2%, in most cases, the precise origin of the complication remains unknown (Table 1) [1–3]. Moreover, we could not find any relevant information reported in the literature regarding a patient with Crohn's disease.

Although Crohn's disease is an autoimmune condition that can affect the entire gastrointestinal mucosa, it is considered a systemic disease because it often involves extraintestinal organs. Among the extraintestinal complications, neurologic manifestations have been reported: peripheral neuropathy, cerebrovascular diseases, demyelinating diseases, multiple sclerosis, and epilepsy. Peripheral neuropathy is the most common neurologic complication [4]. Several cases of chronic inflammatory demyelinating polyneuropathy have been reported [5,6].



**Fig. 2.** Early (9 hours) postoperative magnetic resonance images. A. Axial transverse T2-weighted view at L5 level showing clumped roots (white arrow). Very thin hypointense foci with blood micro-deposition were seen (black arrow). B. Mid-sagittal T2-weighted view demonstrating absence of compressive lesion but showing clumping of the cauda equina nerve roots resulting in a ‘pseudo-cord’ appearance (black arrows).



**Fig. 3.** Late (4 months) postoperative magnetic resonance images. A. Reformatted axial transverse T2-weighted view from volumetric acquisition at L5 level, at the same level as in Fig. 2, showing incomplete resolution of inter-radicular adhesences (arrows). B. Mid-sagittal T2-weighted view in similar slice location as in Fig. 2 showing a declumping of the roots (thin arrows) with, at the level of L5, partial persistent root clumping (pairs thin arrows). Fibrotic bridging is observed at the L5–S1 level (thick arrow). C and D. T1 axial transverse views without and with contrast, in a similar slice location as in A, showing contrast enhancement within clumped roots and revealing persistent blood-root-barrier disruption (white arrows).

**Table 1**  
Cauda equina syndrome developing after uneventful lumbar discectomy, with normal postoperative radiological control.

Authors	Sex	Age (years)	Preexisting narrowing of spinal canal	Level	Visual aide	ioC	CT	MRI	Laminectomy	Complete recovery	Time for complete recovery (month)	Possible explanation
Henriques et al., 2001 [3]	F	31	+	L4–L5	LH	–	–	N	+	–	–	Postoperative swelling causing venous congestion
	M	49	+	L3–L4	LH	–	–	N	+	–	–	
	F	42	+	L4–L5	LH	–	–	N	+	–	–	
	M	54	+	L3–L4	LH	–	–	–	+	+	3	
	F	43	+	L5–L6	LH	–	–	–	+	+	3	
Jensen, 2004 [1]	F	67	–	L4–S1	?	Dural tear repaired with a fat graft	–	N	–	–	–	Secondary ischemia
Dimopoulos et al., 2005 [2]	F	37	–	L5–S1	?	–	–	N	+	+	FD	Venous stasis-induced ischemia of the conus medullaris
	M	44	–	L2–L3	?	–	–	N	–	+	12	
St-Luc, 2016	M	47	–	L3–L4	?	–	–	N	–	+	3	Crohn's disease, infliximab, surgical stress
	F	48	–	L5–S1	M <sup>a</sup>	–	N	Agglutination with tiny hemorrhagic deposits on the roots	UP	+ <sup>b</sup>	10	

FD: within a few days; ioC: intraoperative complication; LH: loupe and headlight; N: normal; UP: unilateral and partial.

<sup>a</sup> Microscope and 18-mm-diameter tubular retractor from the METRx microdiscectomy system.

<sup>b</sup> Continued mild bladder leakage 10-months postoperatively.

In our case, the patient developed a partial CES after an uneventful minimally invasive microdiscectomy that completely resolved her sciatica. During surgery, only the compressed S1 radicular could be observed through the 18 mm metal tube. No compression was observed on the postoperative MRI examination.

The only hypothesis to explain this partial CES was the presence of increased radicular sensitivity in relation to the Crohn's disease or/and the chronic treatment with infliximab. The patient had been receiving infliximab for at least 15 years, and this therapy has been reported to be associated with adverse neurological events (0.2% to 13.4%), including demyelinating conditions, chronic inflammatory demyelinating polyneuropathy, multifocal motor neuropathy and Guillain-Barré syndrome [5,7,8]. Therefore, infliximab could have placed our patient at an increased risk of developing acute peripheral sensory polyneuropathy.

Compared to the general population, patients with Crohn's disease have an increased level of general anxiety and depression, leading to a lower quality of life. Dysfunction of the autonomic nervous system is also frequently described [9]. These mood disorders and autonomic dysfunction influence visceral sensitivity [10], suggesting that our patient may have had a higher level of sensitivity even to a minimally invasive curative surgical procedure.

#### 4. Conclusion

The partial CES in our patient probably resulted from the combination of an autoimmune disease, immunotherapy and surgical stress, even though this was minimal. This phenomenon has been reported in patients with multiple sclerosis undergoing surgical procedures. A meta-analysis of 14 studies reported a significant association between stressful life events, such as a surgical procedure, and a multiple sclerosis relapse [11].

In reporting this case, we want to highlight the possible occurrence of an unexpected event, such as a partial CES, when

performing a minimally invasive surgical procedure on a patient with Crohn's disease treated with infliximab.

#### Disclosure of interest

The authors declare that they have no competing interest.

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