ISSN: 2577-8005

Case Report

Medical & Clinical Research

Spinal Cord Stimulation inpatient with Tethered Cord syndrome: A Case Report

Simonova Jana[1], Komanova Blanka[1], Pataky Frantisek[2], Hudak Vladimir[1], Simon Robert*[3] and Cuchrac Lukas[1]

[1]*1st Department of Anesthesiology and Intensive Medicine, Louis Pasteur University Hospital, Faculty of Medicine, Pavol Jozef Safarik University in Kosice, Slovak Republic, Rastislavova 43, 04001 Kosice*

[2]*Department of Neurosurgery, Louis Pasteur University Hospital, Faculty of Medicine, Pavol Jozef Safarik University in Kosice, Slovak Republic, Rastislavova 43, 04001 Kosice*

[3]*2nd Department of Surgery, Louis Pasteur University Hospital, Faculty of Medicine, Pavol Jozef Safarik University in Kosice, Slovak Republic, Rastislavova 43, 04001 Kosice*

***Corresponding author**

Simon Robert, 2nd Department of Surgery, Louis Pasteur University Hospital, Faculty of Medicine, Pavol Jozef Safarik University in Kosice, Slovak Republic, Rastislavova 43, 04001 Kosice

Submitted: 20 Aug 2020; **Accepted:** 25 Aug 2020; **Published:** 05 Sept 2020

Abstract

Background: *Neuromodulation techniques are an important part of the chronic refractory neuropathic pain treatment. Their effectiveness is insufficiently documented in patients with tethered cord syndrome.*

Case Description: *We present the case of a 32-year-old woman with a history of myelomeningocele repair, followed by a detethering surgery complicated with cerebral fluid leakage. Her intractable pain in her left leg and low back was successfully treated with spinal cord stimulation. Pain intensity decreased from 8/10 to 1-2/10 on her visual analogue scale without regular analgesic intake and her quality of life improved significantly.*

Conclusions: *A review of the literature documents only three case reports of similar efficacy of spinal cord stimulation in the treatment of pain in adult patients with tethered cord syndrome.*

Keywords: Lipomyelomeningocele, Spinal Cord Stimulation, Tethered Cord Syndrome, Chronic Pain

Abbreviations

TCS: Tethered Cord Syndrome;
VAS: Visual Analogue Scale;
MRI: Magnetic Resonance Imaging;
PW: Pulse Width;
CSF: Cerebrospinal Fluid;
LMM: Lipomeningomyelocele

Introduction

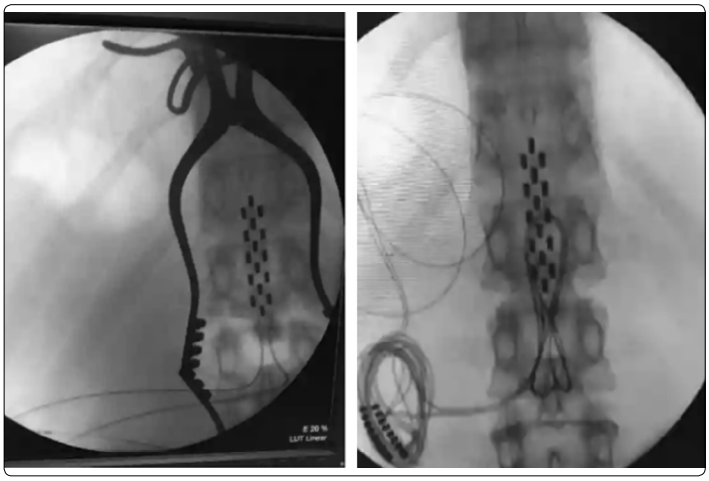
Tethered cord syndrome (TCS) is a neurologic disease caused by tissue attachments that limit the movement of the spinal cord within the spinal column. These attachments cause an abnormal stretching of the spinal cord. This functional disorder may be congenital or acquired. The lower tip of the spinal cord is normally located opposite the disc between the first and second lumbar vertebrae (in the upper part of the lower back). In patients with myelomeningocele, the spinal cord fails to separate from the skin of the back during development, preventing the spinal cord from ascending normally; therefore, the spinal cord is low-lying or tethered. In patients with lipomyelomeningocele, the spinal cord will have fat at the lower end, which may connect to the fat that overlies the thecal sac, which may also lead to tethering. Although the skin is separated and closed at birth, the spinal cord stays in the same location after closing. As the child continues to grow, the spinal cord may stretch with increased tension, causing damage and disrupt blood supply to the spinal cord [1]. Recent theories suggest that the major mechanism in these patients is impaired oxidative metabolism in the affected spinal cord due to extensive arachnoid and fibrous scars and tension [2]. Other causes of TCS are dermal sinus tract, diastematomyelia, lipoma, tumor, thickened/tight filum terminal and a history of spinal trauma or surgery [2-4].

Case Presentation

We present a 32-year-old woman who underwent operation due to myelomeningocele as a 5-day-old newborn. In November 2016 she underwent resection of an intradural lipoma (at the site of the previous surgery) and neurosurgical release of the terminal cone with a posterior approach. In June 2017, she underwent surgery due to formation of a cerebrospinal fluid (CSF) pseudocyst at the site of the previous surgery. Evolution has been marked by

improvements in sphincter disorders and motor functions in the sacral part of the lower limb, but low back pain and neuropathic left leg pain persist. Magnetic resonance imaging (MRI) documented the regression of the cerebrospinal fluid pseudocyst. The patient was followed and treated by a neurologist – in the evening she was given pregabalin, zolpidem and alprazolam. She does not tolerate non-steroidal anti-inflammatory drugs and tramadol. Due to the increasing intensity of pain – visual analogue scale (VAS) 8/10 – despite these conservative therapies, she was examined by a neurosurgeon and was sent for consultation to our workplace. Severe pain reported by the patient – sharp electrical appearance (VAS 8/10) in her left leg and in her left low back.

MRI showed a sufficient spinal canal diameter; therefore, we decided to implant a surgical lead 5-6-5 Sure Scan MRI (Medtronic, USA) under general anaesthesia. According to the patient's clinical examination, mainly the left S1 dermatome area was needed to be covered. In order of a correct electrode placement, we used the technique of intraoperative stimulation and muscle contractions in the related area with following settings: frequency 2Hz, current 4.8, pulse width 360 µs. We found an appropriate position of the lead tip at the Th12 level (Figures 1, 2).



Figures 1 & 2: Positions of the Lead

Following a successful trial period (we obtained coverage more than 80 % of the pain area with a significant decrease of pain intensity in her back and leg area) an Intellis pulse generator Medtronic, USA was implanted in May 2019. Initially, the following parameters were set:

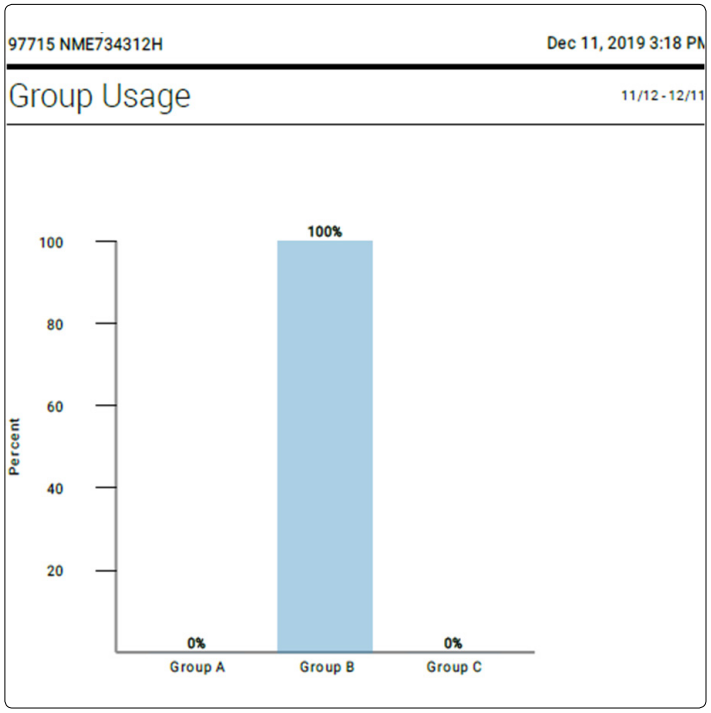
Program A:
2+, 3+, 4-, intensity: 3.6, pulse width: 300 µs, rate: 80 Hz (for her right leg/back)

Program B:
10+,14+, 15-, intensity: 4.1, pulse width: 300 µs, rate: 80 Hz (for her left leg/back)

Program C1:
2+, 3+, 4-, intensity: 4.4, pulse width: 300 µs, rate: 80 Hz

Program C2:
Connection of programs A and B.
Coverage of the painful area and pain relief were satisfactory, the patient was discharged from the hospital.

On December 11, 2019 after a 7-month follow-up, the patient reported sufficient pain relief in her back area and left leg (pain intensity 2/10) but the pain still persists in the area of her left toes. As Graph 1 shows, she mainly used program B.



Graph 1: Percentage of Program Utilisation

As an additional feature, we set up AdaptiveStim® mode – a new technology that uses an accelerometer (a device able to detect a change in position), which automatically adapts to changes in pain intensity when the patient`s position changes. AdaptiveStim® remembers the stimulation level we selected in each position and automatically adjusts the setting the next time the patient moves to that position. We modified program B (pulse width 480 µs and frequency 80 Hz) and program C2 (pulse width 500 µs and frequency 80 Hz) with better pain relief in this area (Figure 3). After the adjusting these settings patient is free of any pain medication and her quality of life has significantly improved. She was able to return to work. 15 months after implantation is pain relief still sufficient. It is not necessary to adjust the stimulation parameters. Patient is still free of any pain medication.

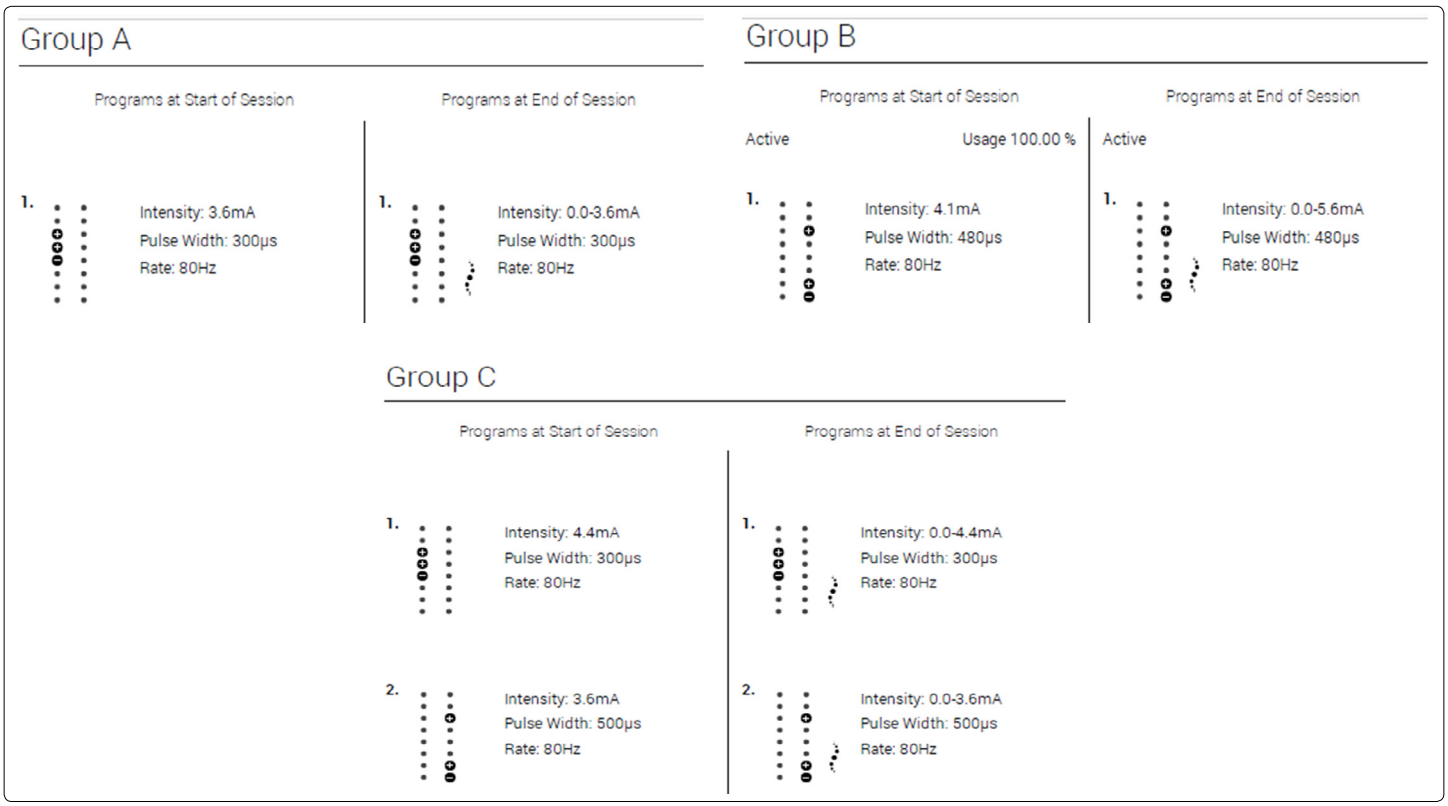


Figure 3: Adaptive Nerve Stimulation – Set Parameters

Discussion

We present a case of a young woman with chronic neuropathic lower back and leg pain after repeated detethering surgeries for TCS with a postoperative complication – cerebrospinal fluid leak and the formation of a pseudocyst at the site of previous surgeries. Following the failure of conservative treatment, we decided on neuromodulation treatment, because in many cases, SCS therapy in the patients with chronic neuropathic pain after spinal surgery have shown a significant improvement in patients' clinical condition and their subsequent return to active life [5].

Only three case reports on this topic were published – two of which were young adult patients – reported by Moens et al. in 2010 and Novik et al. in 2018 and one was a paediatric patient, reported by Tyagi et al. in 2016. Therefore, we took the liberty of supplementing the table created by Novik in his publication with the data of our patient from the presented case study (Table 1).

Table 1: Comparison Between Four Reported Cases of SCS in Patients with TCS

	Moens et al.	Tyagi et al.	Novik et al.	Šimonová et al. Current Study
Sex	Female	Female	Female	Female
Age of initial symptom presentation – diagnosis	23 years	12 years	31 years	28 years
Age of SCS implantation	37 years	19 years	55 years	32 years
Form of spinal dysraphism	LMM	LMM	LMM	Meningocele
Predominant symptoms	Severe chronic pain in back with heavy burning sensation in both legs Dysesthesia Hyperalgesia at her buttocks and her right posterior thigh	Sever back pain Left foot deformity (recurrent) Right foot numbness and weakness Allodynia Hyperpathia Urinary incontinence	Persistent low back, legs pain Right lower extremity paresthesia Almost complete anaesthesia of the groin area Hypoesthesia circumferentially in the right lower extremity Urinary retention	Low back pain (left side) and left leg pain
Number of previous interventions	2	2	4	3
Postoperative complications	N/A	CSF leak, neurologic deterioration	CSF leak	CSF leak – pseudocyst

VAS preoperative	9/10	8-9/10	9/10	8/10
VAS postoperative	2/10	1-2/10	0-2/10	1-2/10
Time of follow-up	N/A	10 months	24 months	15 months
Type of anaesthesia	Epidural	Epidural	General	General
Type of electrode	Specify 565 electrode (Medtronic Inc., Minneapolis, Minnesota, USA)	Specify 565 electrode (Medtronic Inc.)	Penta 5-column paddle lead SCS (St. Jude Medical)	565 Sure Scan MRI (Medtronic Inc.)
Level of lead placement and stimulation	Th11-12	Th8	L1	Th12
Mapping for optimal placement of the lead	Intraoperative stimulation	Intraoperative stimulation	Neurophysiological somatosensory-evoked potential	Intraoperative stimulation
Programming settings	Frequency 60 Hz PW 240 µs Current N/A Voltage 0.2	N/A	Frequency 40Hz PW 400 µs Current 1.7 Voltage N/A	AdaptiveStim® Frequency 80Hz PW 480 µs Current 4.1
Postoperative opioids intake	Cessation	N/A	Reduction >50%	No

As in our patient, in all three published cases, the cause of TCS was (lipo) myelomeningocele; the patients underwent repeated surgical interventions. In both published adult cases, the most annoying symptom was severe chronic neuropathic pain (VAS 9/10) [2, 4]. Our patient perceived pain in her left lower back and left leg, with an intensity of 8/10 on her VAS.

The basis of the success of neuromodulation treatment is the correct placement of the leads – targeting of specific nerve structures and modulation their neuronal activity. Despite of the fact that in all three published cases paddle leads were used, we found significant differences in the lead placement [2, 4, 6]. In general, we can summarize that in all previously published case reports, the electrode was placed lower than in common daily practice, due to the elongation of the tethered or de-tethered spinal cord.

In case of patients with a TCS mapping process for optimal lead placement can be problematic – especially if the procedure is performed under general anaesthesia. Moens et al. and Tyagi et al. used epidural anaesthesia and intraoperative stimulation for exact lead placement, while Novik et al. performed this procedure under general anaesthesia and used neurophysiological somatosensory-evoked potentials for accurate lead placement [2, 4, 6]. In our case, we performed the procedure under general anaesthesia (without muscle relaxants) with intraoperative motor stimulation for optimal placement of the lead.

Success of SCS implantation is depended primarily on good patient selection and a good quality device with various programming options [5]. Our case is unique because we used AdaptiveStim® to better relieve pain in different body positions, because when the position changes, the spinal cord moves closer to or away from the electrodes that emit mild electrical pulses. The level of stimulation, which, for example, blocks pain when standing, may feel uncomfortable in lying position.

We also noticed differences in the set voltage. Moens et al. used the lowest voltage in their case report – only 0.2 what he explained by the anatomy of the spinal cord in TCS and its closer contact with the dura mater [4]. It is proven that the voltage needed for the recruitment of nerve fibers is related to the distance between the electrode and the spinal cord [7]. In our case, we used the highest current (4.1) and PW (480 µs). With these settings, we achieved significant pain relief in our patient without regular analgesic intake.

Conclusions

Based on our experience in accordance with the results of previously published case reports, despite the anatomical abnormality of the spinal cord in TCS, we can confirm the effectiveness of SCS in a patient with TCS. We emphasize the importance of the correct placement of the leads. The procedure under general anesthesia using technique with muscle contractions in the painful area is one of the suitable options.

Acknowledgements

We would like to thank the patient VP for permission to use the clinical findings to support this presentation.

Special thanks to Professor Jozef Firment – Head of 1st Department of Anaesthesiology and Intensive Medicine and Professor Miroslav Gajdoš – Head of Department of Neurosurgery for their general support.

References

1. American Association of Neurological Surgeons. Tethered Spinal Cord Syndrome. [cit. 2020-08-02]. URL<https://www.aans.org/en/Patients/Neurosurgical-Conditions-and-Treatments/Tethered-SpinalCord-Syndrome#:~:text=Tethered%20spinal%20cord%20syndrome%20is,closely%20associated%20with%20spina%20bifida>
2. Novik Y, Vassiliev D, Tomycz ND (2019) Spinal Cord Stimulation in Adult Tethered Cord Syndrome: Case Report and Review of the Literature. World Neurosurg 122: 278-281.
3. Jackson C, Yang BW, Bi WL, Chiocca EA, Groff MW (2018) Adult Tethered Cord Syndrome Following Chiari Decompression. World Neurosurg 112: 205-208.

4. Moens M, De Smedt A, D'Haese J, Droogmans S, Chaskis C (2010) Spinal cord stimulation as a treatment for refractory neuropathic pain in tethered cord syndrome: a case report. Journal of Medical Case Reports 4: 74.

5. Griger M, Rapčan R, Burianek M, Kočan L, Rapčanová S, et al. (2020) Wireless spinal cord stimulation - proposed algorithm before implantation. Medical Science 24: 2452-2459.

6. Tyagi R, Kloepping C, Shah S (2016) Spinal cord stimulation for recurrent tethered cord syndrome in a pediatric patient: case report. J Neurosurg Pediatr 18: 105-110.

7. Alo KM, Holsheimer J (2002) New trends in neuromodulation for the management of neuropathic pain. Neurosurgery 50: 690-703.

Citation: *Simonova Jana, Komanova Blanka, Pataky Frantisek, Hudak Vladimir, Simon Robert and Cuchrac Lukas (2020) Spinal Cord Stimulation inpatient with Tethered Cord syndrome: A Case Report. Journal of Medical & Clinical Research 5(8):184-188.*