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Subarachnoid to subarachnoid shunt for correction of nonfunctioning baclofen pump in a severe case of chronic debilitating post-spinal cord injury spasticity

Adewale A. Bakare, BS,¹ Jonathan Weyhenmeyer, MD,¹ and Albert Lee, MD²

¹Department of Neurological Surgery, Indiana University School of Medicine, Indianapolis, IN;

²Goodman Campbell Brain and Spine and Department of Neurological Surgery, Indiana University School of Medicine, Indianapolis, IN

Author Emails:

bakaread@gmail.com

jweyhen2@gmail.com

alee@goodmancampbell.com

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Corresponding author:

Albert Lee, MD
IU/Goodman Campbell Brain and Spine
355 W. 16th Street
Suite 5100
Indianapolis, IN 46202
Tel: 317-396-1300
Email: alee@goodmancampbell.com

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Abstract

Background and Importance: Perhaps the most disabling condition seen in patients with spinal cord injury is spasticity. Spasticity is characterized as hyperreflexia and hypertonicity as a result of damage to the supraspinal tracts in the aftermath of SCI. Intrathecal baclofen (ITB) is the mainstay therapy for spasticity unresponsive to oral baclofen. One of the problems associated with post-SCI spasticity unresponsive to ITB is development of scar tissue that prevents the diffusion of baclofen in the desired spinal cord area. This case offers a unique strategy to deal with multilevel scar tissue.

Clinical Presentation: This is a 46-year-old paraplegic male with eighth thoracic SCI whose spasticity had been well managed with ITB therapy for many years but recently suffered intractable spasticity necessitating multiple re-operations for non-functioning ITB catheter secondary to extensive scar tissue and intrathecal adhesion. A subarachnoid to subarachnoid shunt was placed, that solved the problem of extensive scar tissue preventing adequate baclofen therapy.

Conclusion: After undergoing multilevel thoracic and lumbar laminectomies with subarachnoid-subarachnoid spinal shunt; the patient's spasticity was finally under control with adequate daily baclofen infusion. This case demonstrates a creative way to address failed ITB catheter before considering other measure like neuroablative procedures (i.e., rhizotomy or myelotomy). Given the case presented, we reinforce the recommendation that ablative procedures, which have far greater complications, should be reserved for patients that have failed medical or different non-ablative therapies.

Keywords: spinal cord injury; spasticity; intrathecal baclofen; multilevel scar tissue; subarachnoid-subarachnoid shunt; neuroablative

Background and Importance

In 2016, the National Spinal Cord Injury statistical center estimated a prevalence of 54 cases of spinal cord injury (SCI) per million persons in the United States.¹ Damage to the upper motor neurons in SCI results in poor regulation of the alpha motor neuron response to tonic muscle stretch reflexes that are constantly relayed to the spinal cord.^{2,3} Subsequently, the alpha motor neurons persistently fire action potentials to opposing muscle groups. As such, damage to the UMN tracts causes hyper-excitability states that manifest as spasticity, hypertonia, and hyperreflexia.^{2,3,4} Unfortunately, spasticity is an inevitable, chronic neurological sequela of SCI.^{3,5} SCI patients in chronic spastic states experience debilitating quality of life characterized by pressure ulcer, poor ambulation, wheelchair distress, and impaired activity of daily living.²

There are a number of medications that have been shown to improve spasticity caused by UMN lesions, e.g., baclofen, benzodiazepines, dantrolene.^{2,6} Oral baclofen has long been known to reduce spasticity and hypertonicity but the dosage required will often lead to intolerable adverse effects.^{2,7} As a result, intrathecal baclofen (ITB) has become a mainstay in the management of spasticity in the SCI and cerebral palsy populations.

In this case report we present a spastic paraplegic patient with extensive scar tissue. The patient's spasticity had been well managed with ITB therapy for many years but recently suffered severe unremitting truncal and lower extremity spasticity necessitating multiple re-operations for non-functioning ITB catheter secondary to extensive scar tissue and intrathecal adhesion. We present a novel unpublished manner in dealing with scar tissue to improve patient spasticity due to SCI.

Clinical Presentation

The patient is a 46-year-old male with a past medical history significant for paraplegia, neurogenic bowel and bladder, and chronic spasticity secondary to severe traumatic brain injury (TBI) and eighth thoracic level (T8) SCI sustained in a motor vehicle accident (MVA). The patient's spasticity was previously well managed with ITB infusion for almost 20 years. His original intrathecal catheter entered the thecal sac at the second to third lumbar interspace (L2-3) and was threaded up to the tenth thoracic level (T10). Over the course of the last 3 years, the

patient presented to our neurosurgery clinic on several instances with intractable torso and lower extremity spasticity. The patient's persistent spasticity resulted in multiple operations for intrathecal catheter revision. Unfortunately, he continued to have residual and increasing spasticity that was not relieved by these interventions.

During an ITB catheter revision in early 2015, he was noted to have a significant amount of subarachnoid scar that had prevented the intrathecal catheter from threading to the desired spinal level. Post-operatively, he continued to complain of disabling upper trunk spasticity in the area extending from his xyphoid to his shoulder. Given his persistent symptoms, we elected to pursue an additional operation 3 months later to attempt to mitigate his increased spasticity. Prior to the operation there was concern that an intrathecal catheter extending from the lumbar spine would not be able to be threaded past the arachnoid adhesions and dural scar in the thoracic spine. With this in mind, we elected to perform bilateral laminectomies at T8 and T7, and a partial laminectomy at T6 to facilitate direct placement of an intrathecal catheter in the thoracic spine. The intraoperative microscope was utilized to improve visualization in order to ensure that the catheter was not infiltrating the spinal cord parenchyma. The intraoperative microscope allowed for direct visualization of the old intrathecal catheter and arachnoid scar that had extended from the T8 level to T9-10 levels. We initially attempted to enter the thecal sac at the T8 spinal level and thread the intrathecal catheter rostral to the T6 level. Due to the extensive scar, the intrathecal catheter could not be threaded higher than the T6 level. Instead, we elected to place the catheter at T6 using fluoroscopic x-ray guidance and then ligated the old, nonfunctional lumbar catheter through a separate incision.

While the patient had some relief of spasms in the upper trunk area postoperatively, the lower extremity spasticity and abdominal area spasticity worsened. The lower truncal spasticity was so bad that patient frequently complained of worsening diarrhea-type symptoms with incomplete evacuation despite adequate bowel regimen. The patient had been placed on bolus dosing of ITB, 40mg oral baclofen daily, and large, scheduled doses of oral valium. His oral valium needs were high enough that his family noticed significant cognitive decline. Unfortunately, his lower extremity symptoms persisted. His spasticity continued to be readily apparent on physical exam with lower extremity scissoring. A CT myelogram showed the patient had a myelographic block impeding cerebrospinal fluid flow (CSF) flow as illustrated in Figure 1. Subsequently, the

myelographic block had prevented the ITB from providing adequate coverage in the lower extremities.

Given the patient's persistent spasticity, two surgical options were discussed with him over several clinic visits. First, the patient could undergo placement of a second baclofen pump with catheterization of the caudal intrathecal space. Second, the patient could undergo a subarachnoid to subarachnoid shunting procedure whereby a subarachnoid-to-subarachnoid shunt would be placed between the thoracic and lumbar spinal levels. After extensive discussions, the patient elected for the subarachnoid to subarachnoid shunting procedure.

The patient was taken back to the OR in late 2016 for placement of the subarachnoid to subarachnoid shunt as described in Figure 1. The procedure began with a T4-5 laminectomy and an L2-3 laminectomy with intradural lysis of adhesions at both levels. Next, an intrathecal catheter was tunneled into the thecal sac at the L2-3 level. The baclofen catheter was then threaded rostrally from the lumbar laminectomy site up to the T11 vertebral level. Next, a standard bactiseal antibiotic catheter was placed intradurally at both the L2-3 and at the T4-5 laminectomy site, creating a subarachnoid-to-subarachnoid shunt. The shunt catheter was then split with scissors and an inline low-pressure standard shunt valve was connected between the two catheters. The shunt was arranged such that there was an inferior to superior direction of flow (e.g., lumbar to thoracic flow pattern). We elected to directly supply the lower thoracic spine with ITB and shunt to the upper thoracic spine because the patient's lower trunk and leg symptoms were more difficult to deal with than his upper trunk area. This allowed a baclofen gradient with a higher concentration of baclofen in the lower thoracic levels. If the patient's symptoms had been reversed e.g., worse spasticity in the upper extremities and thoracic regions, we would have placed the intrathecal catheter in the superior thoracic distribution and reversed the direction of CSF flow on the shunt valve from superior to inferior.

After the operation, the pump flow setting was decreased from 1700 mcg to 700 mcg per day. Postoperatively, the patient's spasticity was significantly improved the day after surgery and continued to improve during his hospitalization. He was monitored in the intensive care unit (ICU) for two days and his requirements for oral baclofen decreased substantially while his vital signs normalized. At his first post-operative clinic visit, his pump was further turned down to 496.6 mcg with continued control of his lower extremity and truncal symptoms. He was rapidly

weaned off his oral valium and now takes minimum oral baclofen as needed for spasticity. The patient is quite satisfied with his post-operative results.

Discussion

Post-SCI spasticity is usually unresponsive to oral baclofen owing in part to an impermeable blood-brain barrier resulting in low tissue concentration of drug in the spinal cord.^{2,7} Oral baclofen is not well tolerated at high doses due to intolerable adverse effects (e.g., respiratory depression) depressed mental status, that limit the ability to provide the dose necessary to achieve maximal therapeutic efficacy.^{2,7} Due to low therapeutic index of oral anti-spasticity medications, surgery tends to be the more effective treatment modality. The surgical options described in the literature can be grouped into either neuroablative or non-neuroablative surgery.⁵

ITB infusion is a well-established non-neuroablative surgical treatment for spasticity.^{2,5} The procedure involves intrathecal catheterization and programmable pump implantation with a refillable reservoir containing baclofen.⁶ The pump allows for regulated and targeted delivery of baclofen from the reservoir to the desired spinal cord area through the intrathecal catheter.⁶ While maximizing its effectiveness, ITB requires lower dosing and prevents systemic distribution thus minimizing side effects often seen in oral baclofen.

In clinical practice, it is not uncommon to have intradural adhesions and scar formation. These pathologies make catheter placement difficult and alter CSF flow dynamics especially after a long-term and recurrent intrathecal catheterization. Several case reports in the medical literature have reported granulomatous scar formation at the catheter tip preventing effective intrathecal delivery of drug.⁸⁻¹¹ This case is unique in the sense that the patient sustained a single-level SCI, yet developed scar tissue and intradural adhesion that extended through multiple vertebral levels presumably creating an anatomical myelographic block as evidenced by the CT myelogram. While myelographic block has been associated with several spinal pathologies including arachnoiditis, neoplasm, or degenerative diseases, its occurrence after SCI is not well documented in the literature.^{12,13} To our knowledge, this is the first report to hint at the occurrence of myelographic block post-SCI. It is possible that the scar tissue and intradural adhesions are the result of post-SCI secondary injury, recurrent spinal manipulation or intrathecal catheterization from prior spinal surgeries. Regardless of the source of the scar tissue,

the fact remains that patient's increased spasticity over the last three years coincided with the growth of extensive scar tissue and intradural adhesions. The myelographic block anatomically impedes the flow of baclofen due to poor CSF flow dynamics, thus; greatly increasing the dosage required to achieve local therapeutic effect.

Conclusion

Post-operatively, the patient was far more satisfied with the outcome of the surgery involving multilevel thoracic and lumbar laminectomies with the placement of a subarachnoid-subarachnoid shunt as a conduit for rostral flow of baclofen. Catheter placement for maximum efficacy is not well studied and this case exemplifies catheter insertion failure in a patient with multilevel scar tissue where threading of catheter tip is impeded. Placement of two catheters, one rostral and the other caudal to the scar location, has not been well documented in the medical literature. This patient's truncal and lower-extremity spasticity posed tremendous challenge because he needed rostral and caudal spinal cord coverage to ameliorate his symptoms. Unfortunately, the Ascenda catheter is not manufactured with a "Y" or "T" connector, which would enable multiple catheters with a single pump source.

Another option we considered was using two baclofen pumps, but this is not a feasible option currently in modern-day medical care. In theory, two baclofen pumps anatomically may provide some benefits, but will likely increase susceptibility to baclofen toxicity without necessarily achieving adequate dosing for spastic and pain control. This was the reason for leaving the ascenda catheters at T11 and placing the shunt catheter up to T4-T5 while inserting an inline low-pressure standard shunt valve with the direction of flow from lumbar to thoracic levels.

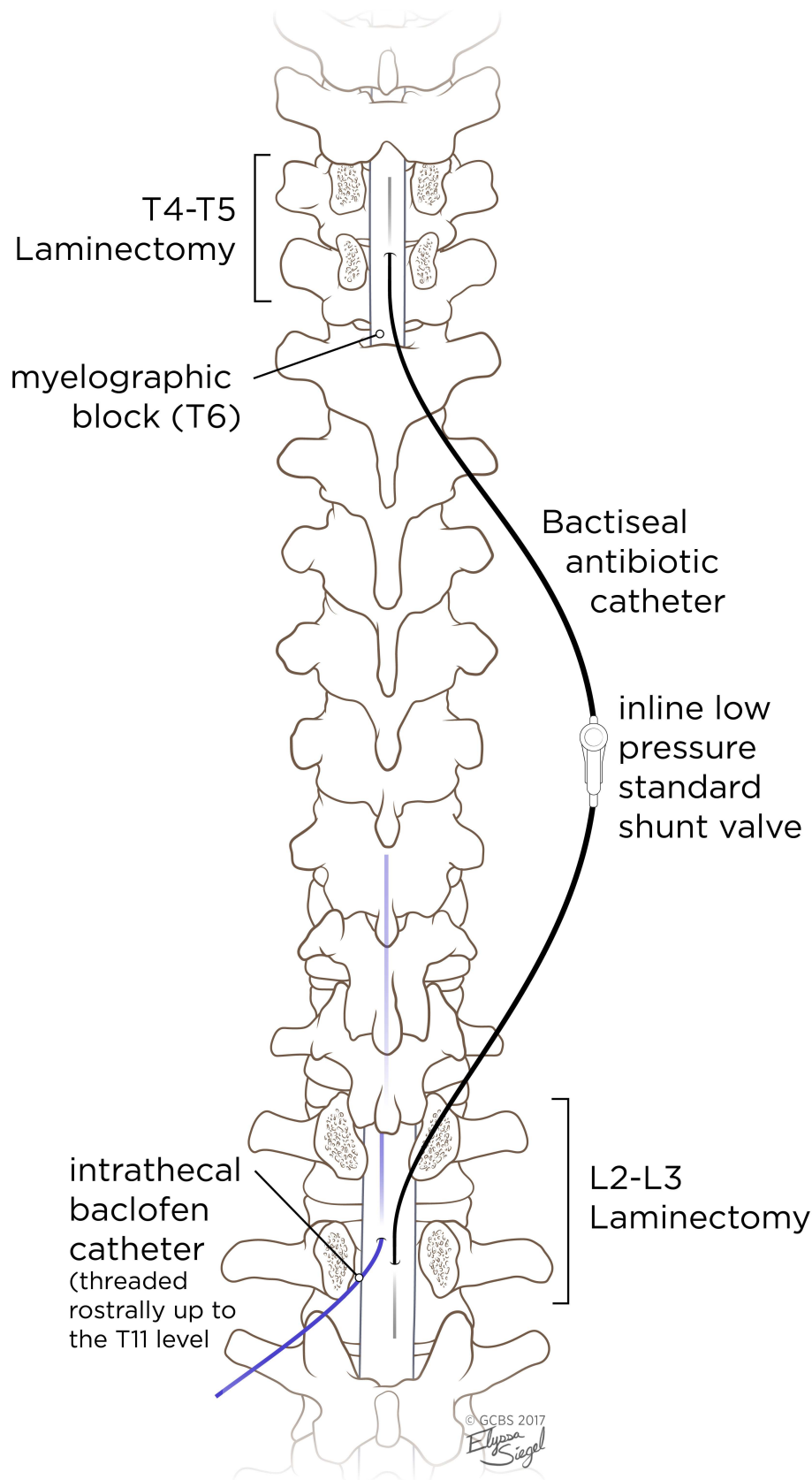
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Legends

Figure 1: Schematic illustration of subarachnoid-subarachnoid shunt placement showing the myelographic block at T6, laminectomies at T4-5 and L2-3, ITB catheter, and superior and inferior bacteseal antibiotic catheters connected to an inline low-pressure standard shunt valve.



HIGHLIGHTS

- Spinal cord injury affects about 54 of every million persons in the U.S.
- The most disabling condition seen in patients with spinal cord injury is spasticity.
- Intrathecal baclofen is standard for spasticity unresolved with oral baclofen.
- Scar tissue often develops preventing diffusion of baclofen in desired spinal area.
- We offer a novel strategy to deal with multilevel scar tissue.

ITB = Intrathecal baclofen

SCI= spinal cord injury

CSF= cerebrospinal fluid flow

ACCEPTED MANUSCRIPT