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Selective dorsal rhizotomy for spastic diplegia secondary to stroke in an adult patient

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Abstract

Background:

Selective dorsal rhizotomy (SDR) is often recommended for children with spastic paraparesis and cerebral palsy. SDR reduces spasticity in the lower extremities for these children with spastic paraplegia. However, SDR is infrequently recommended for adults with spasticity. Spastic diplegia in adult patients can be due to stroke, brain or spinal cord injury from trauma, infection, toxic-metabolic disorders, and other causes. Although rarely considered, SDR is an option for adult patients with spastic diplegia as well.

Case Description:

The authors describe a patient who underwent a SDR with a successful postoperative outcome. This man suffered a hypertensive and hemorrhagic stroke secondary to intravenous drug abuse at age 46. A SDR was performed after two failed intrathecal baclofen pump placements due to recurrent infections, likely resulting from his immunocompromised status. The patient underwent lumbar laminectomies and dorsal rhizotomies at levels L1-S1 bilaterally. Postoperatively, the patient's spasticity was significantly reduced. His Ashworth spasticity score decreased from 4/5 to 1/5, and the reduction in tone has been durable over 3 years.

Conclusion:

SDR in older patients with spastic paraparesis may be considered as a treatment option.

Keywords: Intrathecal baclofen, selective dorsal rhizotomy, spastic diplegia, spastic paraparesis, spastic paraplegia, spasticity

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INTRODUCTION

Brain damage may impair upper motor neuron function, resulting in an absence of inhibitory influence on the lower alpha motor neurons.[10] This may cause an uninhibited reflex arc between the alpha motor neurons and the afferent nerve fibers from the muscles.[10] The loss of cortical inhibition allows for an exaggerated reflex response and uninhibited muscle contraction.[22] This resulting “muscle disorder” or hypertonicity is referred to as spasticity.

Two surgical treatments for spasticity include intrathecal or intraventricular baclofen therapy (intrathecal baclofen [ITB]) and selective dorsal rhizotomy (SDR). ITB therapy can be effective in both children and in adults.[1,2,3] Baclofen is a gamma-aminobutyric acid (GABA) agonist, and intrathecal delivery of the drug through an implanted pump and an intradural spinal catheter allows for the direct administration of baclofen into the cerebrospinal fluid (CSF).[21] Baclofen binds to GABA_B receptors inside the dorsal horn of the spinal cord. This inhibits muscle afferents, with a resulting presynaptic inhibition of alpha motor neurons.[21] This re-establishes the normal balance between excitatory and inhibitory input, decreasing muscle hyperactivity and hypertonicity.[3,15] Complications of the baclofen pump placement surgery include CSF leak, infection, as well as pump and catheter malfunctions.[10,22]

Selective dorsal rhizotomy is most commonly used to treat pediatric patients with spastic diplegia and cerebral palsy. It has also been used to treat spastic diplegia associated with other diagnoses, but it is rarely performed in adults.[4,12,13,22] In the case of adult-onset spasticity, there is often a lack of awareness of the efficacy of this procedure and so often SDR is not offered or considered as a treatment option.[11] In children with cerebral palsy, SDR is recommended earlier in an effort to preserve the child's ability to ambulate, ideally between the ages of two and five.[6,7,16,17] In SDR, the affected sensory nerve rootlets are identified visually and confirmed electrophysiologically by examining electromyographic (EMG) responses from muscles in the lower extremities.[6] In this way, it is determined which rootlets are contributing to spasticity. Once identified, the dorsal rootlets are sectioned (selectively cut). This minimizes the sensory signal originating in the muscle, restoring the balance between sensory input and motor output, and subsequently reducing or eliminating spasticity.[6]

In this article, a rare case is reported in which an adult patient, who suffered a stroke 7 years prior to presentation, underwent a SDR with long-lasting clinical success. Although this is considered very unusual, this patient's unique circumstances prompted the neurosurgical team to consider this “typically pediatric” procedure. This patient serves as an example of how the benefits of SDR can be utilized effectively in the adult population.

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CASE REPORT

A 46-year-old man, former professional athlete, presented with a hypertensive stroke secondary to use of intravenous (IV) drugs, including heroin and cocaine. While admitted to the hospital, it was discovered that he was human immunodeficiency virus (HIV) positive. The patient's stroke resulted in hypoxic-ischemic brain damage leading to severe spasticity. He developed 4/5 bilateral lower extremity spastic diplegia as per the Ashworth scale. Weakness of his right arm and face were additionally noted. The patient's speech was dysarthric but understandable.

For 7 years poststroke, the patient underwent physical therapy, botulinum toxin injections, and took oral baclofen for his increased tone without relief. The patient was wheelchair bound due to the increased tightness and loss of strength in both of his legs. He was unable to stand up or mobilize independently,

and needed assistance with daily activities such as bathing and dressing. The goals of his spasticity management included improvement in mobility and gait, as well as a better range of motion. Unable to achieve these goals with pharmacological treatment, his physiatrist administered an intrathecal test dose of baclofen through a lumbar puncture. A test dosage of 75 ug of ITB was administered. The patient responded positively and underwent surgical implantation of a 40 ml SynchroMed II pump with a T10 level catheter. Postoperatively, the patient did well and was transferred back to inpatient rehabilitation after 48 hours of flat bed rest and IV cefazolin.

Two weeks postoperatively, the patient developed leakage of CSF from the lumbar incision and was subsequently transferred back to the neurosurgical service. The lumbar incision was oversewn and a lumbar puncture below the level of the catheter was done. A pressure of 24 cm H₂O was documented. A computed tomography scan of the head showed mild to moderate ventricular enlargement. The next day, a right frontal Codman ventriculo-pleural shunt was placed. The patient's ventricular CSF did not initially show any bacteria. However, the following day the lumbar puncture CSF grew *Staphylococcus epidermidis*, sensitive to oxacillin. The patient was placed on IV antibiotics to prevent infection of the newly placed pump.

After finishing 8 weeks of IV antibiotics, another area of the patient's incision site opened. Subsequently, the patient received a peripherally inserted central catheter line for another round of IV antibiotics. Due to these recurrent infections, it was decided to remove the patient's right-sided pump, and replace it with a new pump and spinal catheter on the left-side of his body. Postoperatively, the patient developed an infection at the abdominal incision site of the second pump, and the device was removed. At this time, the patient developed deep vein thrombosis and sepsis, and received 45 days of IV antibiotics through long-term IV access. Although the HIV was under control and the viral load was undetectable, it was believed that the patient's recurrent infections were due to his immunocompromised status. In addition, his urinary and fecal incontinence may have contributed to recurrent lower abdominal infections, secondary to contamination from his diaper.

At this time, botulinum toxin injections were re-instituted as the primary treatment for the patient's spasticity: 600 units of botulinum toxin were administered in the adductors, plantar flexors, and hamstrings every 3 months. He reported improvement of his gait 2 weeks after each injection, and experienced maximum effect of the treatment 4 weeks postinjection. The patient was able to walk with a rolling walker; however, he noted difficulty with balance while standing. The effect of the botulinum toxin lasted approximately 2.5 months before wearing off. The patient became septic with a *Staphylococcus* infection secondary to multiple, recurrent intramuscular botulinum toxin injections.

At this point, almost all options were exhausted, and the neurosurgeon proposed a SDR. The patient was explained the risks and unknown but potential benefits of SDR. The surgery would reduce the patient's tone, but could make it difficult for him to walk postprocedure if he was currently using his tone to ambulate. The risk of numbness in lower extremities, neurogenic bladder, and CSF leak were also discussed. The patient agreed to the procedure in an attempt to eliminate or decrease the frequency of the botulinum toxin injections.

The patient underwent selective dorsal rhizotomies at levels L2-S1 bilaterally. Osteoplastic laminectomies were performed from L1 to L5. The dura was retracted laterally. The L2 nerve root on the left was dissected out and confirmed electrically. The nerve root was separated out into the ventral motor and the sensory dorsal portions. Each dorsal sensory nerve was isolated using intraoperative microscopic dissection, and the rootlets were separated into thirds or quarters. Each motor nerve was stimulated, and then each sensory rootlet was stimulated. Rootlets were sectioned if stimulation of the rootlet was associated with clonus and prolonged contraction of the involved muscle groups. This was repeated for L2 through S1 bilaterally based on electrophysiological stimulation and the observed and recorded results. Approximately 50–75% of each dorsal sensory nerve on each side was sectioned.

Spinal nerve root function was tested in this procedure by recording spontaneous and electrically-triggered EMG activity from iliopsoas, adductor, quadriceps, tibialis anterior, gastrocnemius, biceps

femoris, and external anal sphincter muscles innervated by these roots. Portions of these roots were selectively sectioned based on triggered EMG discharge patterns.

At completion of the dorsal rhizotomy, the dura was sewn closed with a 6-0 Nurolon. The lamina of L1 through L5 was put back into position and laminar fusion was done using the Synthes laminar plating system. An epidural spinal catheter was placed for pain management control. There were no intraoperative complications.

Postoperatively, the patient did well and was transferred back to inpatient rehabilitation. His spasticity was significantly reduced according to the Ashworth scale. Prior to his SDR, his lower extremity spasticity was 4/5. Postoperatively after SDR, this patient's Ashworth scale is consistently 1/5 bilaterally. The patient experienced no numbness or change in bowel and bladder control postsurgically.

At the present time, 3 years postsurgery, the patient attends physical therapy twice a week. The patient has no spasticity. Although he still requires assistance to ambulate, he experiences increased comfort due to the reduction of his tone and associated pain. The patient continues to present with lower extremity weakness and cannot stand without support. His muscular strength is about 3/5 in both legs proximally and 2/5 distally. This weakness resulting from the stroke is permanent and cannot be improved via a dorsal rhizotomy, as most poststroke paresis is irreversible after 6 months. His hydrocephalus is well-controlled with his shunt set at 180 mm H₂O. His HIV viral load is undetectable. The patient is extremely satisfied with the outcome of his dorsal rhizotomy.

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DISCUSSION

Hypoxic-ischemic brain injury often results in the development of weakness and spasticity. Spasticity, in these cases, is the result of upper motor neuron dysfunction. Severe spasticity, unresponsive to oral medications and physical therapy is often controlled with intramuscular botulinum toxin or alcohol injections into the affected nerves. However, if the spasticity or dystonia is not focal in nature, then the number of injections required for each muscle group may be excessive. In addition, botulinum toxin and alcohol blocks are only temporary and the neural damage causing the spasticity is permanent, therefore a more durable treatment option should be considered. Medically intractable spasticity and dystonia require surgical intervention, either ITB, SDR, or in certain cases deep brain stimulation. While SDR is not the first choice of treatment in patients with spasticity, for this patient, other options such as physical therapy, oral medications, alcohol blocks, and botulinum toxin injections failed to control his spasticity. In addition, ITB therapy was attempted twice, but because of the patient's immunocompromised status and debilitated condition the incisions failed to heal well. The patient developed superficial and deep wound infections, prompting pump explantation.

Selective dorsal rhizotomy has the potential to reduce patients' spasticity and muscle tone, possibly resulting in the improvement of motor skills. This could provide increased ease of daily life activities, such as dressing and self-hygiene.[7,9,11,16] However, clear postoperative expectations as well as patients' preoperative functional conditions must first be evaluated. We have found that the postoperative functional improvements after SDR are comparable among pediatric and adult patients.[11] The benefits of SDR include improvement in ambulation and balance with or without a walking device following rigorous physical therapy.[14,16,20] For patients that are able to walk with assistance before surgery, postoperative goals include walking independently.[7] Risks of the procedure may include deformation of the spine.[23] In a study of 43 patients with severe spasticity due to cerebral palsy, 19 patients developed spinal distortions including scoliosis, lumbar hyperlordosis, thoracic hyperkyphosis, and L4–L5 spondylolisthesis postsurgically.[23] The presence of a former spinal condition, prior neurological damage, as well as older age place an individual at an increased risk for these outcomes.[23] In adult patients with a mature spine, spinal deformity may be less of a risk.

A spastic diplegic patient is considered to be a good candidate for SDR if conservative treatments such as botulinum toxin injection therapy and orthopedic treatments are producing intolerable or undesirable responses.[7,11] In addition, the individual's goals for treatment should be discussed with the patient and their family or caregivers.[16] For patients with mixed motor disorders such as dystonia and spasticity or rigidity and spasticity, ITB therapy may be more beneficial.[5] SDR, unlike placement of an ITB pump, is a permanent procedure. Finally, it is important that patients understand that physical therapy is an important component of their postoperative care plan.[16]

Current literature supporting the effectiveness of SDR in patients who have developed spasticity for reasons other than cerebral palsy is scarce.[11] There are few reports of dorsal rhizotomy in patients with other conditions.[11] In a study of 30 pediatric patients with spasticity secondary to anoxic or ischemic brain injury, neurodegenerative disease, hydrocephalus, spinal cord disease, or cerebral palsy, all demonstrated successful reduction in tone after SDR.[19] Two out of the eight patients in the study that were able to walk demonstrated an improved gait. Four subjects who were unable to walk showed improved sitting and standing. Two patients who were nonambulatory prior to the SDR were able to ambulate with support within 2 years postoperatively.[19] In a second study of 154 patients with various causes of spasticity, 100% of the subjects showed improvement of lower limb spasticity at early and late follow-up.[18] Other outcomes of SDR include improvements in gait, range of motion in affected joints, bladder function, upper limb spasticity, and speech and cognition at both early and late follow-up. These outcomes did not diminish over time. 20% of patients who were nonambulatory preoperatively were able to walk following surgery.[18]

The effectiveness of SDR specifically in poststroke spasticity is documented in a report of 2 patients who developed severe unilateral lower extremity spasticity following ischemic stroke.[8] These patients did not respond well to oral muscle relaxants. One patient was 68 years old at the time of his stroke, and SDR restored his ability to walk and significantly improved his ability to partake in daily activities. The second patient was 89 years old at the time of infarct, and his SDR eliminated some of the tone in his legs while eliminating all pain associated with the spasticity.[8] Our case report reinforces the clinical results previously reported. While these case reports are few, all seem to demonstrate that SDR is a viable and efficacious option in adults with spastic diplegia.

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CONCLUSION

The effectiveness and durability of SDR in adults is confirmed by the patient in this case report. The patient's spasticity remains resolved 3 years following his SDR. He is able to mobilize with the assistance of a walking device without experiencing the pain and discomfort that had been associated with his increased tone. The patient is satisfied with the results of the SDR and he reports that his quality of life has been significantly improved. SDR should be considered as a treatment option for adult patients with spastic paraparesis.

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Footnotes

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