Role of dorsal rhizotomy in spinal cord injury–induced spasticity

Report of 3 cases

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Selective dorsal rhizotomy (SDR) is a highly effective and well-established neurosurgical procedure in the treatment of cerebral palsy–associated spastic diplegia.2,11,14,15 It reduces spasticity by decreasing the excitatory input entering the spinal cord after the transection of selected sensory nerves of the cauda equina.19 Class I evidence supports the utility of SDR for the immediate and permanent reduction of spastic diplegia in patients with cerebral palsy, improving motor function and performance of ADL.14 Selective dorsal rhizotomy has primarily been studied in patients with cerebral palsy, and there have been rare reports of its use and outcomes in patients with multiple sclerosis or neurodegenerative disorders.6 To our knowledge, the use of SDR has not been reported in children with SCI. We present 3 cases in which pediatric patients with SCI-associated spasticity were treated with SDR at the University of Washington (2 patients) and University of Utah (1 patient).

Case Reports

Case 1

History and Examination. A 9-year-old girl sustained a gunshot wound that resulted in an American Spinal Injury Association (ASIA) Grade A SCI of T-2. After the in-
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jury, she suffered progressively worsening spasticity in her lower extremities. Despite multiple interventions including large doses of oral baclofen and repeated botulinum toxin injections to her hip flexors, knee extensors, and knee flexors as well as phenol blocks to her hip adductors, she continued to experience severe spasticity. She suffered severe pain, difficulty sitting in her wheelchair, and problems with transfers in and out of the wheelchair. At times, her spasms were so severe she would fall out of her wheelchair, one such occasion resulting in a tibial fracture.

**Operation.** Given the limited benefit received from her previous medical interventions, the implantation of an ITB pump was considered when the patient was 14 years old; however, the patient lived a significant distance from a center equipped to perform interrogations and refills, and therefore the family declined. As an alternative, SDR was considered, and, after counseling, the patient and her family agreed to proceed. The patient’s preoperative Ashworth scores were 3 in the bilateral hip adductors and her family agreed to proceed. The patient’s preoperative Ashworth scores were 3 in the bilateral hip adductors and quadriceps, 2 in the bilateral hamstrings, and 1 in the bilateral ankles.3

The patient underwent SDR with stimulation-mapped guidance and sectioning of 80% of the sensory roots from L-1 to S-1 via L1–2 laminectomies.

**Postoperative Course.** The patient did well, and at 9-month follow-up, she was found to have significantly reduced tone in her lower extremities, with Ashworth scores of 1 in each muscle group with the exception of a score of 2 in her right knee extension. At the last follow-up, she was completely weaned off her oral baclofen therapy and she had improved ADL performance and less difficulty with static sitting in her wheelchair, transfers to and from the wheelchair, and pain.

**Case 2**

**History and Examination.** An 11-year-old girl who had suffered a high lumbar SCI during infancy as a result of nonaccidental trauma presented with significant spasticity in all lower-extremity muscle groups with an Ashworth score of 3/5. This was negatively impacting her quality of life and function. Initially after the injury, she had significant lower-extremity weakness, with an Ashworth score of 1/5 in her bilateral hip abductors and hip extensors as well as a score of 0/5 in her bilateral dorsiflexors. She was ambulatory with excessive lateral trunk sway, hip circumduction, and excessive knee extension throughout the stance phase and used braces and crutches.

**Operation and Postoperative Course.** To treat the spasticity, which had developed over time, she underwent SDR through an osteoplastic laminotomy from L-2 to S-1. Sixty-four percent of the nerve roots were sectioned at these levels. Ten months after surgery, her Ashworth scores in all muscle groups were 1/5, and she had improved strength in her dorsiflexion to a score of 1/5. She was able to walk more easily with the assistance of her braces and crutches.

**Case 3**

**History and Examination.** A 13-year-old boy with C-2 quadriplegia experienced progressive lower-extremity spasticity, extensive knee flexion, and extension spasms. He had suffered a traumatic fall at 1 year of age that resulted in the quadriplegia. His Ashworth scores at presentation were 2/5 in his bilateral dorsiflexors and 1/5 in the remaining muscle groups, but he had sustained lower-extremity clonus with minimal manipulation. Oral baclofen provided marginal relief, and the patient’s caregivers were having significant difficulty with wheelchair mobility and personal hygiene care.

**Operation.** Surgical options including placement of a baclofen pump and dorsal rhizotomy were discussed. As the patient had a long history of methicillin-resistant *Staphylococcus aureus* colonization and infections, the decision was made to pursue SDR and avoid the complication of hardware infection with placement of a baclofen pump. The patient subsequently underwent sectioning of 50%–75% of the sensory nerve rootlets from L-2 to S-1 via a single-level L-2 laminectomy without complication.

**Postoperative Course.** On initial postoperative follow-up, the patient had significant improvement, with a substantial reduction in his spasticity and near elimination of his clonus. At 8-week follow-up, his Ashworth scores were 0/5 in his bilateral extremities, except for a score of 1/5 in left dorsiflexion; however, at 6 months, he had complete return of his debilitating lower-extremity clonus with minimal manipulation of his upper or lower extremities. The patient ultimately had a baclofen pump implanted approximately 6 months after undergoing SDR, with an excellent, dose-dependent response.

**Discussion**

The National Institute on Disability and Rehabilitation Research estimates that the annual incidence of SCIs, excluding individuals who die at the scene of the injury, is near 12,000 cases a year. In 2012, approximately 270,000 people in the United States were living with SCI. Patients who have sustained an SCI can face significant physical limitations from loss of function and associated spasticity, as well as personal losses since the secondary effects of their injury affect their relationships with family and friends and their ability to work and care for themselves. Additionally, the financial burden of SCI is significant, with the average 1st-year treatment expense depending on the level of injury estimated to be between $334,000 and $1,000,000 and the additional yearly costs averaging $40,000–$177,000 (National Institute on Disability and Rehabilitation Research).

Despite decades of research, no surgical or medical treatment has been effective at restoring lost function after SCI. Long-term goals in the management of SCI include minimizing the consequences of the trauma, optimizing the conditions of long-term survival, and improving the patient’s quality of life.20 Patients with SCI routinely score significantly lower than their normative counterparts on quality of life measures aimed at assessing physical, emotional, and social function as well as mental health, bodily pain, and overall medical health.20 Spasticity-related issues are consistently rated among
SCI patients as a significant contributor to a lower quality of life. Spasticity after SCI occurs in 65%–78% of patients and contributes to joint contractures, pain, pressure sores, limited rehabilitation potential, and impaired mobility affecting personal hygiene and ADL. Particular to pediatric patients, spasticity in the growing patient can be especially harmful because of the bone and muscle development occurring during this time. Treatments to target SCI-related spasticity are limited and of varying effectiveness, although they may substantially improve a patient’s function and quality of life.

**Pathophysiology**

The most well-accepted definition describes spasticity as “a motor disorder characterized by a velocity-dependent increase in the tonic stretch reflexes with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex, as one component of the upper motor neuron syndrome.” The muscle stretch reflex originates in muscle spindles, sensory receptors within the muscle, when they perceive the mechanical deformation associated with muscle stretch. This initiates excitatory impulses down large-diameter sensory fibers, called Type Ia afferents, which are coiled around the muscle spindle. These Ia sensory fibers project centrally to form monosynaptic excitatory connections with the alpha motor neurons in the ventral spinal cord innervating the same muscle group, resulting in reflex contraction. Inhibitory connections to antagonistic muscles are also made by connections with local circuit neurons.

Given that spasticity is thought to arise in response to increased tonic stretch reflexes, much research has been centered on investigating the possible alterations in spinal reflex transmission that could be responsible. This simplistic model points to two areas toward which treatment should be focused: reducing the excitatory afferent input and increasing the reduced inhibition.

**Conventional Treatments**

Current conventional treatments for SCI-induced spasticity include physical and occupational therapy, oral and injectable antispasmodic agents, and ITB administration; most of these treatments aim at increasing the reduced inhibition, and each has varying degrees of efficacy.

Physical and occupational therapies are often first-line treatments focusing on muscle stretching and range of motion exercises to maintain or improve mobility and prevent the formation of contractures. Orthotics can also be used to keep a spastic limb in a neutral position, or progressive casting can help gradually stretch severely spastic limbs. However, as spasticity becomes more severe, the effects of these interventions become limited and can often be painful for the patient.

The most commonly used and efficacious oral medications in the treatment of SCI-induced spasticity include baclofen and tizanidine. Both drugs act in a systemic fashion and, although no single study has compared their effectiveness, in both cases it is usually mild and wanes over years of use.

Baclofen, a GABA<sub>4</sub> agonist, is the current pharmacological agent of choice. Baclofen binds to receptors in the lamella of the spinal cord where primary sensory fibers terminate. Because GABA is inhibitory in nature, baclofen ultimately decreases neurotransmitter release, inhibiting and limiting signal propagation. Despite its potential efficacy, it has a short half-life, requiring frequent dosing, and administration is often limited by medication side effects including sedation and ataxia.

Baclofen can also be continuously administered directly into the cerebrospinal fluid by long-term intrathecal drug delivery systems. Because baclofen crosses the blood-brain barrier poorly, this route optimizes the concentration of baclofen in the spinal cord while minimizing the systemic side effects? However, intrathecal infusion has been linked to tolerance, requires surgical intervention for initial placement followed by pump replacements when the generator lifespan has been exceeded, involves routine pump refills, and has the possibility of infection with each intervention.

Botulinum neurotoxin is an injectable pharmaceutical agent that causes chemical denervation by inhibiting the release of acetylcholine at the neuromuscular junction, leading to a reduction in muscle contraction. Clinical effects at the injection site can last for approximately 4 months until recovery occurs through axonal sprouting and subsequent muscle reinnervation. Although botulinum neurotoxin can be clinically efficacious, it is limited by its focal effects and the need for repeated administration.

These traditional, and a multitude of experimental, therapies all have potential beneficial effects on SCI-induced spasticity; however, these beneficial effects come in varying degrees, without permanent effects and with several associated shortcomings. Despite the variety of agents tested, there have been few advances in the management of spasticity related to SCI, which is unfortunate because the growing consensus among patients living with SCI is that the alleviation of autonomic dysfunction or spasticity is a higher priority than walking when considering quality of life issues. It is imperative that we as clinicians and researchers continue to pursue interventions to target SCI-induced spasticity and related primary outcome measures.

**Selective Dorsal Rhizotomy**

Although dorsal rhizotomy was developed in the early 1900s, it did not gain popular acceptance until the late 1980s. Since its inception, variations of this procedure have been developed, but all involve the selective disruption of dorsal sensory roots to reduce the excitatory sensory input to the spinal interneurons and ultimately decrease the excitatory output of the alpha motor neurons. The procedure is performed via laminectomies at either the level of the conus medullaris or the level of the exit foramina of the lumbosacral roots. Despite skepticism regarding the validity of intraoperative electrophysiological stimulation, it remains a standard practice in SDR. Sensory rootlets, whose response to repetitive 50-Hz stimulation is sustained with synchronous muscle activation, are thought to be abnormal. Up to 80% of the...
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abnormally stimulated dorsal rootlets are then sectioned from L2–S1.

Selective dorsal rhizotomy combined with postoperative physical and occupational therapy has been demonstrated to effectively reduce spasticity, improve range of motion, and improve function in children with spastic cerebral palsy.\(^4\) Three randomized controlled trials and other nonrandomized prospective studies have provided strong evidence that SDR decreases lower-extremity spasticity in children with spastic diplegia.\(^4\) Additional studies have demonstrated maintenance of the postoperative benefits in the long term.\(^17\) When compared with age- and preoperative Gross Motor Function Classification Score–matched controls undergoing ITB therapy in this patient population, Kan et al.\(^7\) demonstrated that SDR provided a significantly larger magnitude of improvement in tone, passive range of motion, and gross motor function, with few patients requiring subsequent orthopedic interventions.

Clearly, SDR has an important role in the management of patients with spastic diplegia due to cerebral palsy. The cases presented in this article suggest that SDR may also have a role in the management of medically refractory spasticity secondary to other conditions, specifically SCI. Depending on the degree of the SCI, the goals of SDR may be focused on improvement in personal hygiene care, pain, and so on, and not on improvements in function, although the possibility of improving spasticity-limited function in patients with incomplete SCI can be considered. Two of the 3 patients with SCI treated with SDR had a significant postoperative improvement in their preoperative spasticity without complications at 9 and 10 months after intervention. These patients were able to obtain such significant improvement without the continuing long-term financial commitments and maintenance that corresponds with daily medication, intermittent injections, or pump refills. Although long-term follow-up was limited in this case series, one can extrapolate the positive role of SDR in patients with SCI-induced spasticity could have on both quality of life and the cost of care.

In 1 of the 3 patients treated, initial improvement was followed by complete return of his preoperative excess tone shortly after SDR. The reason for this response is unclear, but since this individual was the only patient with cervical pathology in this series, one possibility is that the pathophysiology of spasticity secondary to cervical SCI is different from that caused by thoracic and lumbar SCI. Selective dorsal rhizotomy has traditionally been used in the management of patients with spastic diplegia, so the outcomes in quadriplegic patients are not well known. Cervical pathology could be a contraindication to a successful SDR; however, important to this case is the fact that previous SDR is not a contraindication to placement of a baclofen pump. The quadriplegic patient presented here recently underwent baclofen pump placement and had excellent postoperative tone control.

**Conclusions**

Spinal cord injury–induced spasticity can be associated with several medical complications and significantly reduced quality of life. Conventional treatment methods are not curative, generally require the repeated administration of medication or repeated procedures, are limited in their effectiveness, and are coupled with side effects and tolerance. Selective dorsal rhizotomy, which reduces the excitatory afferent input, has had tremendous success in the management of spastic diplegia secondary to cerebral palsy and may have a role in the management of SCI-induced spasticity. Additional efforts should focus on patient selection and outcomes for SDR in this unique patient population.

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**References**

11. Pellizzari R, Rossetto O, Schiavo G, Montecucco C: Tetanus...

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