

Health Quality Ontario

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Ontario Health Technology Assessment Series

Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy: A Health Technology Assessment

KEY MESSAGES

What Is This HTA About?

Lumbosacral dorsal rhizotomy is a surgical procedure for children with spastic cerebral palsy. The procedure is intended to permanently decrease lower limb spasticity by cutting spinal nerves. The surgery is always followed by physical rehabilitation.

What Did the HTA Find?

For children whose lower limb spasticity significantly limits motor development, dorsal rhizotomy effectively reduces spasticity and (with physical therapy) increases motor function and functional independence. Motor gains are related to level of disability. Less disabled children with some mobility are more likely to achieve motor skills like running or jumping. More disabled children generally gain skills like crawling, sitting, or standing. Functional independence and caregiver burden also improve for many children after surgery.

Major surgical complications are infrequent. Families interviewed about treatment satisfaction after surgery felt that their children had improved and that they were satisfied with treatment. However, parents also said that surgery and post-operative rehabilitation are highly stressful, time-consuming, and interfere with care for their other children.

Families report a lack of medical information upon which to make an informed decision. They also face enormous financial burdens incurred both at the point of surgery and after surgery, as well as the lack of rehabilitation supports after surgery.

Funding dorsal rhizotomy for spasticity in children with cerebral palsy could cost the government \$1.3 million per year.

HEALTH TECHNOLOGY ASSESSMENT AT HEALTH QUALITY ONTARIO

This report was developed by a multi-disciplinary team from Health Quality Ontario. The clinical epidemiologist was Gaylene Pron, the health economists were Brian Chan, Hong Anh Tu, and Xuanqian Xie, the patient engagement specialists were Mark Weir and David Wells, and the medical librarian was Caroline Higgins.

The medical editor was Elizabeth Jean Betsch, and others involved in the development and production of this report were Merissa Mohamed, Kellee Kaulback, Ana Laing, Claude Soulodre, Vivian Ng, Andrée Mitchell, Nancy Sikich, and Irfan Dhalla.

We appreciate the reviews and consultations on the *Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy: a Health Technology Assessment Report* from the following:

For the clinical section: Dr Paul Cooper, Department of Neurology, London Health Sciences Centre, Ontario; Dr James Drake, Department of Neurosurgery, The Hospital for Sick Children, Toronto, Ontario; Dr Jean-Pierre Farmer, Division of Neurosurgery, The Montreal Children's Hospital, Montreal, Quebec; Dr Darcy Fehlings, Division of Developmental Pediatrics, Holland Bloorview Kids Rehabilitation Hospital, Toronto, Ontario; Dr Golda Milo-Manson, Division of Developmental Pediatrics, Holland Bloorview Kids Rehabilitation Hospital; Dr Unni Narayanan, Division of Orthopedic Surgery, The Hospital for Sick Children, Toronto, Ontario; and Dr Paul Steinbok, Division Pediatric Neurosurgery, British Columbia's Children's Hospital, Vancouver, British Columbia.

For the economic section: Julie Chiba Branson, Operations Manager Child Development Program, Holland Bloorview Kids Rehabilitation Hospital, Toronto, Ontario.

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ABSTRACT

Background

Cerebral palsy, a spectrum of neuromuscular conditions caused by abnormal brain development or early damage to the brain, is the most common cause of childhood physical disability. Lumbosacral dorsal rhizotomy is a neurosurgical procedure that permanently decreases spasticity and is always followed by physical therapy. The objectives of this health technology assessment were to evaluate the clinical effectiveness, safety, cost effectiveness, and family perspectives of dorsal rhizotomy.

Methods

We performed a systematic literature search until December 2015 with auto-alerts until December 2016. Search strategies were developed by medical librarians, and a single reviewer reviewed the abstracts. The health technology assessment included a clinical review based on functional outcomes, safety, and treatment satisfaction; an economic study reviewing cost-effective literature; a budget impact analysis; and interviews with families evaluating the intervention.

Results

Eighty-four studies (1 meta-analysis, 5 randomized controlled studies [RCTs], 75 observational pre-post studies, and 3 case reports) were reviewed. A meta-analysis of RCTs involving dorsal rhizotomy and physical therapy versus physical therapy confirmed reduced lower-limb spasticity and increased gross motor function (4.5%, $P = .002$). Observational studies reported statistically significant improvements in gross motor function over 2 years or less (12 studies, GRADE moderate) and over more than 2 years (10 studies, GRADE moderate) as well as improvements in functional independence in the short term (10 studies, GRADE moderate) and long term (4 studies, GRADE low). Major operative complications, were infrequently reported (4 studies). Bony abnormalities and instabilities monitored radiologically in the spine (15 studies) and hip (8 studies) involved minimal or clinically insignificant changes after surgery.

No studies evaluated the cost effectiveness of dorsal rhizotomy. The budget impact of funding dorsal rhizotomy for treatment of Ontario children with cerebral palsy was \$1.3 million per year.

Families reported perceived improvements in their children and expressed satisfaction with treatment. Ontario families reported inadequate medical information on benefits or risk to make an informed decision, enormous financial burdens, and lack rehabilitation support after surgery.

Conclusions

Lumbosacral dorsal rhizotomy and physical therapy effectively reduces lower-limb spasticity in children with spastic cerebral palsy and significantly improves their gross motor function and functional independence. Major peri-operative complications were infrequently reported. Families reported perceived improvements with dorsal rhizotomy, and surgery and post-operative rehabilitation were intensive and demanding.

TABLE OF CONTENTS

LIST OF FIGURES	7
BACKGROUND.....	8
Health Condition.....	8
Technology.....	11
Research Questions.....	12
CLINICAL EVIDENCE REVIEW	13
Objective	13
Methods.....	13
Results	15
Short-Term Effectiveness of Dorsal Rhizotomy	17
<i>Gross Motor Function.....</i>	<i>17</i>
<i>Short-Term Effectiveness for Functional Independence and Caregiver Assistance</i>	<i>32</i>
<i>Treatment Satisfaction with Dorsal Rhizotomy at Short Term</i>	<i>42</i>
Long-Term Effectiveness of Lumbrosacral Dorsal Rhizotomy.....	45
<i>Long-Term Effectiveness for Gross Motor Function</i>	<i>45</i>
<i>Long-Term Functional Independence and Caregiver Assistance.....</i>	<i>52</i>
<i>Treatment Satisfaction at Long Term.....</i>	<i>56</i>
Safety of Dorsal Rhizotomy for Spastic Cerebral Palsy.....	60
<i>Peri-operative Complications</i>	<i>60</i>
<i>Bladder Dysfunction</i>	<i>63</i>
<i>Sensory Abnormalities</i>	<i>67</i>
<i>Spinal Bony Abnormalities</i>	<i>69</i>
<i>Hip Instability.....</i>	<i>81</i>
Discussion	87
Conclusions.....	89
ECONOMIC EVIDENCE REVIEW	90
Objectives.....	90
Methods.....	90
<i>Sources</i>	<i>90</i>
<i>Literature Screening.....</i>	<i>90</i>
<i>Inclusion Criteria</i>	<i>90</i>
<i>Exclusion Criteria</i>	<i>90</i>
<i>Outcomes of Interest.....</i>	<i>90</i>
<i>Data Extraction.....</i>	<i>91</i>
<i>Study Applicability Appraisal.....</i>	<i>91</i>
Results	91
<i>Literature Search.....</i>	<i>91</i>
Discussion	92
Conclusion.....	93
BUDGET IMPACT ANALYSIS	94
Objectives.....	94
Methods.....	94

<i>Target Population</i>	94
<i>Resource and costs</i>	95
<i>Analysis</i>	97
<i>Sensitivity analysis</i>	97
<i>Expert Consultation</i>	97
Results	97
<i>Base Case</i>	97
<i>Sensitivity Analysis</i>	98
Discussion	98
Conclusions.....	100
PUBLIC AND PATIENT ENGAGEMENT	101
Background	101
Methods.....	101
<i>Engagement Plan</i>	101
<i>Recruitment of Participants</i>	102
<i>Interview Approach</i>	102
<i>Data Extraction and Analysis</i>	103
Results	103
<i>Physical and Emotional Experience of Living With Cerebral Palsy</i>	103
<i>Experience of Treatments for Cerebral Palsy</i>	104
<i>Exploring Dorsal Rhizotomy</i>	105
DISCUSSION.....	110
CONCLUSION.....	111
ABBREVIATIONS	112
GLOSSARY	113
APPENDICES.....	114
Appendix 1: Literature Search Strategies	114
<i>Clinical Evidence Search</i>	114
<i>Economic Evidence Search</i>	115
Appendix 2: Evidence Tables.....	117
Appendix 3: Evidence Quality Assessment	159
Appendix 4: Budget Impact Analysis Inputs.....	163
Appendix 5: Letter of Information and Consent Form	164
Appendix 6: Interview Guide	167
REFERENCES	168

LIST OF TABLES

Table 1. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Randomized Controlled Studies	20
Table 2. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Comparative Studies	24
Table 3. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Pre-Post Cohort Studies	28
Table 4. Short-Term Effectiveness of Dorsal Rhizotomy for Functional Independence and Caregiver Assistance Evaluated With PEDI.....	34
Table 5. Short-Term Effectiveness of Dorsal Rhizotomy for WeeFIM-Evaluated Functional Independence.....	39
Table 6. Long-Term Changes in Gross Motor Function After Dorsal Rhizotomy for Children With Spastic Diplegia.....	49
Table 7. Long-Term GMFM-66 Interval Changes by Baseline Motor Disability After Dorsal Rhizotomy	49
Table 8. Maximum GMFM-66 Score and Age at Attainment of Maximum Scores for Lund Dorsal Rhizotomy Cohort and Ontario Control Cerebral Palsy Population	51
Table 9. Long-Term PEDI Scores for Functional Independence in Children with Spastic Diplegia After Dorsal Rhizotomy.....	54
Table 10. Treatment Satisfaction At Long Term With Dorsal Rhizotomy	58
Table 11. Peri-Operative Complication Events.....	62
Table 12. Bladder Dysfunction Following Lumbosacral Dorsal Rhizotomy	66
Table 13. Clinical Studies Evaluating Spinal Bony Abnormalities After Dorsal Rhizotomy for Children with Spastic Cerebral Palsy.....	70
Table 14. Scoliosis in Children with Spastic Cerebral Palsy After Dorsal Rhizotomy.....	73
Table 15. Spinal Sagittal Deformities of Kyphosis or Lordosis After Dorsal Rhizotomy.....	76
Table 16. Spondylolisthesis and Spondylolysis After Dorsal Rhizotomy.....	79
Table 17. Radiographic Evaluation of Hip Instability After Dorsal Rhizotomy	84
Table 18: Number of Patients Receiving Dorsal Rhizotomy Between 1996 and 2001 in Ontario	94
Table 19: Dorsal Rhizotomy Eligibility Assessment Unit Costs.....	95
Table 20: Dorsal Rhizotomy Physician Unit Costs.....	96
Table 21: Dorsal Rhizotomy and Standard Care Total Costs per Patient	98
Table 22: Net Cost of Dorsal Rhizotomy per Year	98
Table A1: Evidence Base of Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy	117
Table A2. Short-Term Effectiveness of Lumbrosacral Dorsal Rhizotomy for Spastic Cerebral Palsy	126
Table A3. Clinical Studies Evaluating Long-Term Effectiveness of Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy.....	144
Table A4. Clinical Studies Evaluating Safety of Lumbrosacral Dorsal Rhizotomy for Spastic Cerebral Palsy.....	154
Table A5: GRADE Evidence Profile for Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy	160
Table A6: Risk of Bias in Observational Uncontrolled Pre-Post Intervention Studies.....	161
Table A7: McHarm Quality Assessment of Safety Based on Observational Studies.....	162
Table A8: Total Net Cost of Dorsal Rhizotomy per Year Sensitivity Analyses	163

LIST OF FIGURES

Figure 1: PRISMA Flow Diagram — Clinical Evidence Review	17
Figure 2: PRISMA Flow Diagram — Economic Evidence Review	92

BACKGROUND

Health Condition

Cerebral palsy is the most common cause of childhood physical disability.^{1,2} It is a spectrum of neuromuscular conditions caused by abnormal brain development or damage to the brain and can occur in utero (from infection, vascular disruption, etc.) or can be acquired after birth.³ One formal definition describes cerebral palsy as:

[A] group of permanent disorders of the development of movement and posture causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour; by epilepsy; and by secondary musculoskeletal problems.⁴

The prevalence of cerebral palsy is approximately 2 per 1,000 live births.⁵ Low birth weight, premature birth, and multiple gestations are the strongest risk factors.⁶⁻⁸ Although cerebral palsy is usually caused by an interference in brain development in utero, approximately 10% of cases occur postnatally through cerebral infection, cerebral trauma, or cerebrovascular accidents.^{9,10} Mild cerebral palsy might also be detected only when children fail to reach developmental milestones, presenting as late as 5 years of age.¹¹ Population prevalence rates based on registries that collect and record case ascertainment at later ages, 2 and often 5 years of age, could more accurately reflect the burden of the condition.^{1,12} A cerebral palsy prevalence of 3.6 per 1,000 children aged 8 years was reported in a population-based disabilities surveillance program.¹³

Life expectancy of children with cerebral palsy depends on the severity of the disability and can be similar to that of the general population or be much lower.^{14,15} The severity of disability and presence of comorbidity are the most important predictors of long-term survival.¹⁶⁻¹⁸ Respiratory problems are the most common cause of death for all cerebral palsy variants.¹⁹ Improvements in treatment and care, particularly gastrostomy tube feeding, have increased the likelihood of survival.¹⁵

The defining characteristic of cerebral palsy is the dysregulation of muscle tone, which is needed to maintain normal posture and to facilitate movement.²⁰ Dysregulation results in spasticity, which has been defined as “disordered sensory motor control, resulting from an upper motor neuron lesion, presenting as intermittent or sustained involuntary activation of muscles resulting in hypertonia (increased muscle tone).”²¹ Spastic muscles can reduce a child’s mobility and ability to participate in play and other normal healthy activities. Spasticity can also impede normal musculoskeletal tissue growth, and multiple corrective orthopedic surgeries are frequently required.²² Other motor symptoms of muscle overactivity can also include flexor and extensor spasms, co-contractions, and dystonia (involuntary movements resulting in twisting, tremors, and abnormal postures).²³ These effects can result in weakness and lack of dexterity, which can also be functionally disabling and less amenable to treatment.

In cerebral palsy, other neurologic disabilities or comorbidities frequently coexist.^{24,25} A range of comorbidity has been reported for children with cerebral palsy, increasing with the level of motor disability and depending on the extent, magnitude, and location of congenital or acquired injury to the central nervous system.^{25,26} Comorbidities have involved all body systems: eating disorders (dysphagia, difficulty chewing, nasal regurgitation, choking on liquids); cognitive impairment; sensory impairments (visual or auditory); and communication difficulties

(dysphonia, laryngeal weakness, and dysarthria [difficulty speaking]).^{24,27} Seizures also occur in almost half of children with cerebral palsy; epilepsy occurs most often in children with little or no ability to walk.²⁷ Children with cerebral palsy are also more likely than their peers to have behavioural and emotional problems and frequently have chronic pain.²⁸⁻³⁰

Classification systems of cerebral palsy have been based on severity, anatomic distribution, and motor function. Spastic cerebral palsy is the most common form and represents 80% to 85% of cases.³¹ Major spastic anatomic categories include spastic hemiplegia (unilateral involvement of one or two limbs); spastic diplegic (bilateral involvement with more leg than arm involvement) or spastic quadriplegic (bilateral involvement with equal movement or more arm than leg involvement).³² Other dyskinetic or abnormal involuntary movements, can occur in isolation or accompany spasticity and are referred to as mixed spastic-dyskinetic cerebral palsy.^{32,33} Diagnosis of these conditions can be clinically challenging.^{34,35}

In addition to anatomic classification, one classification system is based on motor function, the Gross Motor Functional Classification System (GMFCS).³⁶ The GMFCS is a five-level classification of severity based on gross motor function and includes Level I (no functional impairment); Level II (can need assistive device); Level III (assistive device needed for ambulation); Level IV (limited self-mobility, wheelchair often required), and Level V (wheelchair bound, unable to sit independently). Although there is a range of physical abilities for children, even within these levels, children with spastic hemiplegia are generally at Levels I and II and those with spastic quadriplegia are at Levels IV and V. In general, children classified as Levels I to III are classified as ambulatory; Levels I and II are independent ambulatory, and Level III is dependent ambulatory. Children classified as Levels IV and V are nonambulatory.

Management of Cerebral Palsy

There is no cure for cerebral palsy. Given the complexity of the condition and its symptoms, an integrated multi-disciplinary approach at specialty treatment centres is recommended.³⁷⁻³⁹ Team members can include rehabilitative physical and occupational therapists, physiatrists, developmental pediatricians, neurologists, speech and language pathologists, psychologists, counsellors, education specialists, orthopedic surgeons, or neurosurgeons. Treatment approaches that focus on lower limb dysfunction are diverse and generally depend on the severity and distribution of spasticity.⁴⁰ First-line treatments for children with cerebral palsy often include physical rehabilitation approaches that are continually evolving and often directed at gait training, strength training, and fitness or exercise training programs.⁴¹ Various orthotic devices, such as splints, casts, braces, or molds, are also often employed during training.⁴²

A range of pharmacologic agents are used either locally or systemically for spastic overactive muscles.^{43,44} Local anesthetic agents, such as lidocaine and bupivacaine, are used to relax muscles by blocking nerve conduction. Chemical neurolysis agents, such as ethyl alcohol and phenol, impair nerve conduction by destroying portions of nerves. Botulinum toxins have also been used to selectively and reversibly reduce upper and lower-limb spasticity in order to decrease muscle tone.⁴⁵ Baclofen, a central nervous system depressant, has been administered orally or intrathecally to manage spasticity. However, baclofen administered intrathecally via a spinally implanted pump would not be considered a first-line treatment because of the invasiveness of the procedure. It would be more likely to be considered as a treatment for cases unsuccessfully treated with less invasive interventions and as an alternative to dorsal rhizotomy. Botulinum toxin or baclofen administered either orally or intrathecally can be adjunct therapy after surgery if spasticity remains troublesome.

Surgical treatment for children with cerebral palsy can include both orthopedic and neurosurgical procedures. Treatment objectives for orthopedic surgery, however, are generally to correct musculoskeletal disorders that develop in the growing child as a consequence of untreated spasticity.²² Untreated spasticity can cause shortening of muscles, muscle contractures, relative immobilization, and impaired longitudinal muscle growth. Various bony and soft orthopedic procedures, including tendon lengthening, tendon transfers, rotational osteotomies, and joint stabilization procedures, address these musculoskeletal disorders.⁴⁶ Hip displacement and dislocation is common among children with cerebral palsy, particularly those with decreased levels of physical function, and hip surveillance until adulthood is recommended.⁴⁷

Although neurosurgery for cerebral palsy has included deep brain stimulation and lumbosacral dorsal rhizotomy, deep brain stimulation is more often used for rare forms of cerebral palsy involving dystonia.^{48,49} Lumbosacral dorsal rhizotomy is the main neurosurgical approach for spastic cerebral palsy, but it is not the first treatment option, and it is always performed in conjunction with extensive post-operative physical rehabilitation.

Clinical Need and Target Population

Given that cerebral palsy is a multifaceted disorder with various potential neurologic causes, motor impairment, and comorbidity, careful selection of patients for dorsal rhizotomy is essential for successful spasticity reduction and functional improvement. In general, treatment objectives are to maximize function, to ease care, and to prevent secondary orthopedic problems and pain. Determining patient eligibility for dorsal rhizotomy is complex, and many authors have stressed that selection should be based on a multi-modal assessment covering various aspects of patients and their families and should be performed by a multi-disciplinary team.³⁷⁻³⁹

A systematic review of the patient selection criteria in 52 studies evaluating lumbosacral dorsal rhizotomy for spastic cerebral palsy being offered at more than 25 centres worldwide reported varied selection criteria.³⁸ The most common criteria identified were lower-limb spasticity that interfered with mobility or caregiver support, adequate muscle strength, absence of other movement abnormalities, gross motor function level, clinical diagnosis, level of cooperation, and motivation of the child and parent or caregiver.

Treatment objectives for dorsal rhizotomy depend on the functional status of the child, and parents have expressed many concerns for children with various disability levels.^{38,50} In children with some degree of mobility or assisted mobility, the main treatment objective is to manage spasticity that is interfering with their mobility or physical function. Therefore relevant outcomes would include improved gait, such as walking faster, longer, more efficiently, or with a more normal appearance, or increasing play or activity participation.

Children with cerebral palsy frequently have long-term functional limitations and require long-term or lifelong assistance in their activities of daily living.⁵¹⁻⁵³ In children with limited or no mobility, dorsal rhizotomy can also be performed if spasticity affects their positioning, comfort (reducing pain or improving sleeping), or need for caregiver assistance. Ease of care can refer to tasks performed for patients by a caregiver or by patients themselves with their unaffected side and are referred to as passive care or self-care activities. Difficulties with these tasks arise from complications of spasticity and subsequent soft tissue changes leading to skin breakdown, stiff or abnormal limb positioning, and difficulty washing and dressing. All of these events can increase children's need for caregivers' assistance and affect their psychological and physical health.⁵⁴ Therefore, for this group of children, any improvements in sitting, positioning, bowel

and bladder care, or wheelchair mobility would also be appropriate treatment objectives. These outcomes are also objectives that parents have requested in their goal-directed or patient-centred consultations for their children with cerebral palsy.

Technology

Dorsal rhizotomy is a neurosurgical procedure in which sensory nerves running through the spine, which are evaluated as probably causing overactive spastic muscles, are severed. The surgery reduces spasticity by decreasing the sensory input by irreversibly severing selected spinal sensory nerve rootlets. The operative details of the surgery have been described in detail by several authors.⁵⁵⁻⁵⁷ The degree to which dorsal rhizotomy reduces spasticity and supports normal growth and development is thought to minimize the need for corrective orthopedic surgeries and other medical therapies (baclofen, botulinum toxin A, etc.).³⁷

The surgery always requires intensive inpatient and long-term outpatient physical rehabilitation. Protocols for rehabilitation have been shown to vary considerably by treating centre and over time.⁵⁷ Children undergoing the surgery need to have access to and be able to participate in these intensive ongoing specialized physical services after surgery to rehabilitate muscles and function and to recover from any adverse effects, usually loss of muscle strength.⁵⁸

The surgical procedure itself also varies by centre and has evolved over time.^{59,60} Dorsal rhizotomy is also often called selective dorsal rhizotomy because the procedure involves the selection and targeting of specific nerves. However, the route of surgical access and the method of selecting spinal nerves thought to be responsible for spasticity can vary according to method of evaluation and definition of abnormality.⁵⁷ The number of abnormal nerve rootlets sectioned in various spinal regions also varies by treating surgeon.

Rhizotomy is performed with the patient under general anesthesia and can involve extended inpatient stays for post-operative recovery and physical rehabilitation. Therefore, as with any major surgery, there are surgical risks of potential complications.. In addition severing spinal nerves can have unintended effects, such as bowel or bladder dysfunction, sensory abnormalities, back pain, and possible spinal deformity. Eliminating spasticity can also adversely affect physical function if the child is using increased muscle tone from spasticity to assist in mobility, standing, or transfers. The unwanted consequence of muscle weakness after this surgery is the reason for the eligibility requirement for adequate muscle strength..

Regulatory Information

The Ontario Physician Schedule of Benefits includes fee codes for surgeons conducting dorsal rhizotomy.

Context

At the time of this analysis, this surgery is not available in Ontario. It is, however, offered in: Vancouver, British Columbia, Calgary and Edmonton, Alberta, and Montreal, Quebec.

Two countries, Australia and England, have taken national approaches to introducing dorsal rhizotomy as a centralized specialized service. In 2006, the Australian Health Ministers Advisory Committee, based on the evidence reviewed by their Medical Services Advisory Committee, recommended that one Nationally Funded Centre with a multi-disciplinary team should be established for the Australian population of approximately 20 million through their Nationally Funded Centres Programs.⁶¹

In 2014 the National Health Service for England (which has a population of approximately 55 million) established centres for cerebral palsy through the Commissioning through Evaluation (CtE) program.⁶² They commissioned specialist dorsal rhizotomy procedures for more than 100 children with cerebral palsy at five designated hospitals across the country.

Research Questions

The research objectives of this health technology assessment were:

- to assess the short-term and long-term clinical effectiveness and safety, of and patient and family satisfaction for lumbosacral dorsal rhizotomy and physical therapy for children with spastic cerebral palsy.
- to review the published literature on the cost-effectiveness and cost-utility of lumbosacral dorsal rhizotomy in patients with spastic cerebral palsy and to analyze the budget impact of funding dorsal rhizotomy for children with spastic cerebral palsy in Ontario.
- to survey perspectives and values of patients with spastic cerebral palsy and their families through personal interviews about how cerebral palsy affects their quality of life and the value of treatments (including dorsal rhizotomy) to them.

CLINICAL EVIDENCE REVIEW

Objective

The objectives were to assess the short term and long term effectiveness, safety of and patient and family satisfaction for lumbrosacral dorsal rhizotomy with physical therapy for children with spastic cerebral palsy.

Methods

We performed a literature search on December 2, 2015 to retrieve studies published from inception to the search date. We used the Ovid interface to search the following databases: MEDLINE, Embase, Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, Health Technology Assessment, National Health Service Economic Evaluation Database (NHSEED), and Database of Abstracts of Reviews of Effects (DARE). See Appendix 1 for Literature Search Strategies, including all search terms.

Search strategies were developed by medical librarians using controlled vocabulary (e.g., Medical Subject Headings) and relevant keywords. The final search strategy was peer-reviewed using the PRESS Checklist.⁶³ Database auto-alerts were created in MEDLINE and Embase and monitored for the duration of the HTA review.

Literature Screening

A single reviewer reviewed the abstracts and, for those studies meeting the eligibility criteria, we obtained full-text articles. We also examined reference lists for any additional relevant studies not identified through the search.

Inclusion Criteria

- English-language full-text publications
- Studies published to December 2015 and in auto-alert updates until December 2016
- Systematic reviews, randomized controlled trials (RCTs), observational studies, case reports
- Studies with at least 1 month of follow-up and involving clinical outcome measures
- Studies involving the surgical intervention lumbrosacral dorsal rhizotomy for patients with spastic cerebral palsy

Exclusion Criteria

- Abstracts and conference proceedings
- Animal and in vitro studies
- Editorials, or commentaries
- Technical reports

Outcomes of Interest

- Gross motor function
- Functional independence
- Caregiver assistance
- Safety

- Patient or family satisfaction with treatment

Data Extraction

We extracted relevant data on study characteristics, risk of bias items, and PICOT (population, intervention, comparison, outcome, and time) using a standardized data form. The form collected information about the following:

- Source (i.e., citation information, contact details, study type)
- Methods (i.e., study design, study duration and years, reporting of missing data, reporting of outcomes, and whether or not the study compared two or more groups)
- Outcomes (i.e., outcomes measured, number of participants for each outcome, number of participants missing for each outcome, outcome definition and source of information, unit of measurement, upper and lower limits [for scales], and time points at which the outcome was assessed)

Analysis

Results of the studies were not pooled because of varying definitions, measurement, and follow-up periods of reported outcomes. Variability of the surgical approach and the clinical heterogeneity of the study populations also caused substantial inter-study and intra-study differences. Results were summarized in tables for the major outcome variables, stratified by levels of presenting motor disabilities where possible. A *P* value of .05 or less was considered statistically significant for effect estimates reported in clinical studies. Outcomes were also evaluated against minimal clinically important differences, where such values were known. Gross motor function measures, where possible, were evaluated in the context of expected growth and developmental patterns of children with cerebral palsy.

Quality of Evidence

The quality of the body of evidence for each effectiveness outcome was examined according to the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) Working Group criteria.⁶⁴ The overall quality was determined to be high, moderate, low, or very low using a step-wise, structural methodology. The risk of bias for non-randomized observational interventional studies with validated outcome measures was evaluated using a modified form of the ROBINS-1 measurement tool.⁶⁵ The quality of studies evaluating safety or complication reports was evaluated based on a modified form of the McMaster Quality Assessment Scale of Harms (McHarm)⁶⁶ tool for harms assessment and reporting. The study quality was judged to be high, moderate or low based on the key features considered in the scale: definitions, methods of collection, adequacy of reporting and sampling and follow-up. Systematic reviews were used as background information or sources of additional studies and were not evaluated for their quality.

Expert Consultation

In January 2016, experts on lumbrosacral dorsal rhizotomy for spastic cerebral palsy were first consulted. Consultants included physicians in the specialty areas of neurosurgery, orthopedics, neurodevelopmental medicine, and neurology. The role of expert advisors was to contextualize the evidence on lumbrosacral dorsal rhizotomy and provide advice on management practices of children with spastic cerebral palsy. The health technology assessment was performed independently of the consulted experts, and the statements, conclusions, and views expressed in this report do not necessarily represent the views of the experts.

Results

Literature Search

The database search yielded 1,319 citations published by December 2015. After removing duplicates, we reviewed titles and abstracts of 738 citations to identify potentially relevant articles. Full texts of relevant articles were obtained for further assessment. We hand-searched the reference lists of the included studies, along with health technology assessment websites and other sources, to identify additional relevant studies. Database auto-alerts created in Ovid and CINAHL were monitored up to December 2016, and one additional relevant study⁶⁷ was identified. In total, eighty-four studies (1 meta-analysis, 5 RCTs, 75 observational pre-post interventional studies, and 3 case reports) met the inclusion criteria of reporting on one or more clinical outcomes after dorsal rhizotomy for spastic cerebral palsy.

Figure 1 presents the adapted flow diagram for the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA).⁶⁸

Systematic Reviews

Five systematic reviews were identified and excluded from this clinical review because they either did not address the research objectives of this review or did not have current literature search strategies.⁶⁹⁻⁷¹ Objectives for the systematic reviews were as follows: short-term outcomes after dorsal rhizotomy up to 2001⁷¹; long-term outcomes after dorsal rhizotomy up to 2010⁷²; all interventional strategies for ambulatory cerebral palsy⁶⁹; a review of systematic reviews for any interventions in cerebral palsy⁷⁰; and a review of selection criteria employed in studies of dorsal rhizotomy.³⁸ These reviews were used for background information and to identify any additional studies not found in this evidence search.

Evidence Review

The main reasons for undergoing dorsal rhizotomy are to decrease lower-limb spasticity and improve range of motion in order to improve function, to increase comfort or functional independence, and to decrease need for caregivers' assistance. Many clinical studies reported mainly on how surgery affects spasticity or range of motion. The quantitative measures reported for spasticity included myometry, dynamometry, and some version of the Ashworth Scale, an ordinal rating scale based on clinical examination or subjective evaluations. Clinical observations of these outcomes confirmed reduced spasticity and increased range of motion. The reduced lower-limb spasticity and increased range of motion in the short term were also shown in three RCTs⁷³⁻⁷⁵ comparing early surgical protocols and physical therapy with physical therapy alone.

Reduced lower-limb spasticity is now generally recognized as an accepted outcome of dorsal rhizotomy. How reduced lower-limb spasticity affects function among children with spastic cerebral palsy treated with dorsal rhizotomy as well as treatment satisfaction and safety of the surgery were the key outcomes evaluated in this review.

The primary studies including one or more of the clinical outcomes after dorsal rhizotomy are outlined alphabetically in Appendix 2, Table A1. The clinical studies were further classified according to whether the study objectives involved short-term outcomes (2 years or less) in Table A2 or long-term outcomes (more than 2 years) in Table A3. Studies evaluating safety and complication events are listed in Table A4. In these summary tables, studies were grouped by the country of origin and listed within country by region of the institution where services were

provided and by the date of publication. The study characteristics, patient characteristics, and types of outcomes being evaluated are also listed.

Studies evaluating treatment effectiveness with validated outcome measures on motor function or functional independence (as opposed to clinical observation only), in either the short term or the long term, were discussed and evaluated fully in the evidence review. Treatment satisfaction reported in any study was also evaluated in the review. All studies involving safety or harms assessments were also evaluated in the review.

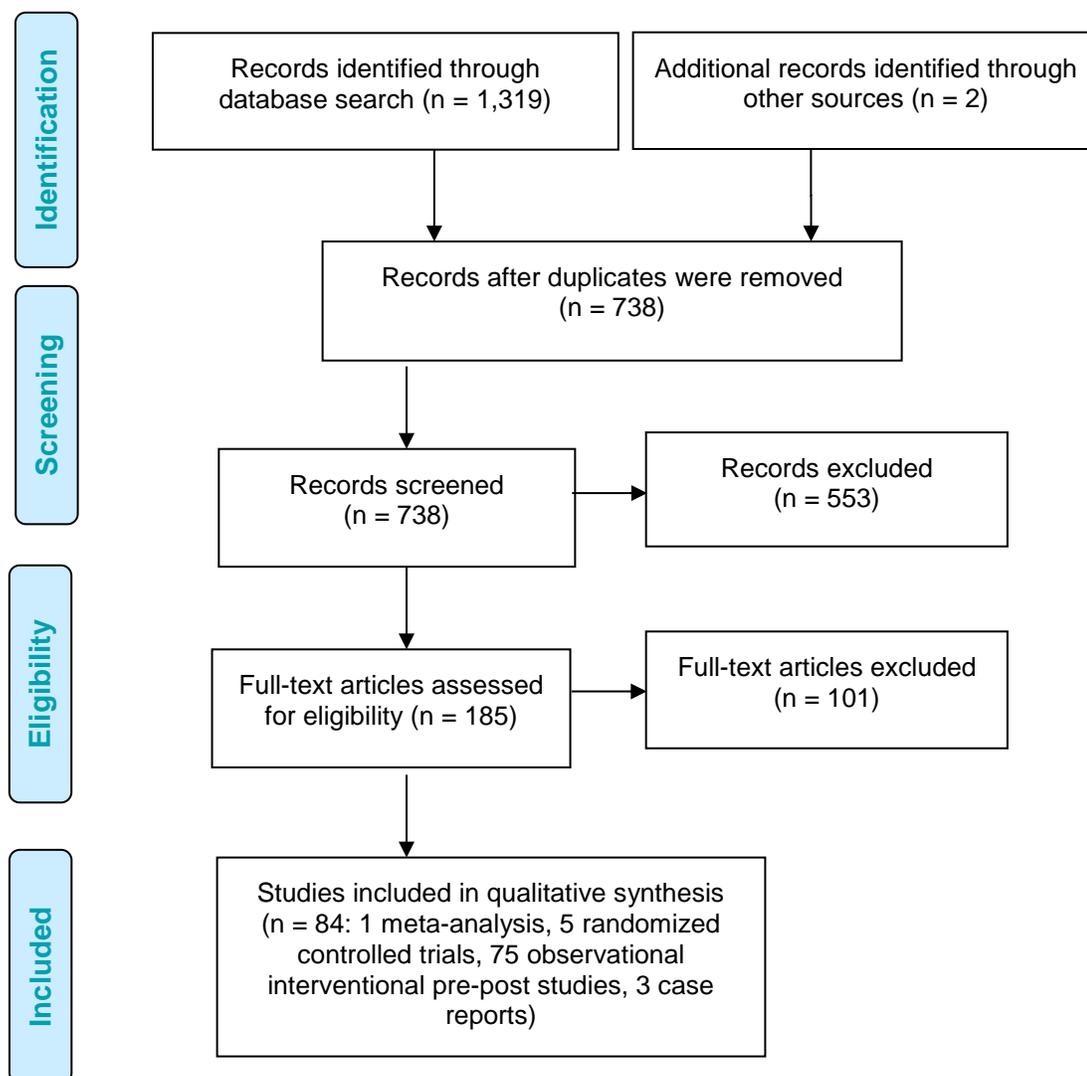


Figure 1: PRISMA Flow Diagram — Clinical Evidence Review

Source: Adapted from Moher et al, 2009.⁶⁸

Short-Term Effectiveness of Dorsal Rhizotomy

Those studies reporting short-term outcomes using validated outcome measures for gross motor function are detailed in Tables 1 and 2 and discussed in *Gross Motor Function*, below. Studies using validated outcome measures for functional independence and caregiver assistance are detailed in Tables 3 and 4 and discussed in under short-term effectiveness of dorsal rhizotomy. Treatment satisfaction with dorsal rhizotomy in the short term is detailed under treatment satisfaction for dorsal rhizotomy.

Gross Motor Function

Eighteen studies evaluated gross motor function in short-term follow-up (2 years or less) with standardized assessment tools involving gross motor function measure (GMFM). One study also reported on the gross motor performance measure (GMPM), which is derived from the GMFM.^{76,77} The GMFM is a reliable, valid, and responsive instrument to measure change in motor dysfunction in children with cerebral palsy.⁷⁸⁻⁸⁰ The GMFM scores are reported as total

scores and as total scores for any of the five subdomains: Item A: lying/rolling lie/crawling; Item B: sitting; Item C: crawling/kneeling, sitting; Item D: standing; Item E: walking/running/jumping. A change in GMFM score of 6% or greater has been estimated to represent a clinically important change.⁸¹

Goal scores calculated for the GMFM-88 are restricted to specific intended treatment objectives for a particular child (e.g., rolling would not be scored if the child is walking). The GMFM-66,⁸² based on the GMFM-88, has 66 items over the subdomains. The GMFM-66 is an interval-based scaled instrument to calculate total scores that requires a computer program (GMAE). The GMFM-66 instrument has been shown to be reliable and responsive to motor changes in children with cerebral palsy.⁸³⁻⁸⁶

Construction of developmental curves representing the average pattern of development for children with cerebral palsy allows prognosis of gross motor function for each motor classification level.⁸⁷ Children classified as GMFCS Level I will on average achieve 90% of their gross motor potential by 4.8 years, and children classified as GMFCS Level V (the greatest disability) achieve 90% by 2.7 years.

In this review, studies evaluating gross motor function in children after undergoing dorsal rhizotomy involved five RCTs, one meta-analysis, three pre-post cohorts with comparative groups, and nine pre-post longitudinal cohort studies. The results of the RCTs are listed in Table 1, the comparative pre-post cohort studies in Table 2, and the uncontrolled pre-post cohort studies in Table 3.

Randomized Controlled Trials

Of the five RCTs, three⁷³⁻⁷⁵ compared children with spastic cerebral palsy who received dorsal rhizotomy followed by physical therapy with those who received physical therapy only (Table 1). Two trials were in Canada: one in Vancouver (Steinbok et al⁷⁴) and one in Toronto (Wright et al⁷⁵); the third trial was in Seattle (WA) (McLaughlin et al⁷³). All trials involved rhizotomy surgeries performed more than 20 years ago with varied techniques of nerve assessment and nerve root dissection. None formally classified the study population by the Gross Motor Functional Classification System (GMFCS), a standard classification for the degree of motor disability among children with cerebral palsy. Comparator treatment arms in the trials all involved physical therapy but employed various protocols of duration, intensity, frequency, and rehabilitative goals.

All trials reported significant reduction in lower-limb spasticity (evaluated with the Ashworth Scale) and greater range of motion. The primary outcome measure in all trials, however, was how dorsal rhizotomy affected gross motor function (evaluated as the GMFM) at follow-up points of 9 months, 1 year, and 2 years. In the two Canadian trials,^{74,75} improvements in gross motor function in the dorsal rhizotomy and physical therapy arms were clinically relevant (6% change) and were statistically greater than in the comparator arm treated only with physical therapy. In both trials improvements in Item E (walking/running/jumping) were greater than those in Item D (standing) for children treated with dorsal rhizotomy and physical therapy than for the comparison group.

The third trial performed in Seattle by McLaughlin et al⁷³ differed from the other two in that gross motor function evaluated as the GMFM-88 total score or GMFM composite scores (Items C, D, and E) was not significantly different between the study arms at the 1-year or 2-year follow-up. This trial differed from the others in other ways. The average rate of dorsal root resection was significantly lower (25%) in this trial than in the others (45% in Vancouver and 41% in Toronto). In addition the age range (3.3 to 8.0 years old) for the target population in this trial was much

broader than the Vancouver (3.0 to 6.5 years old) or the Toronto (3.5 to 7.6 years old) trials. The physical disability of patients in this trial, measured as a baseline GMFM-88 score of 71%, was much higher (less disabled) in Seattle than in Vancouver (56%) or Toronto (53%).

A meta-analysis of the mean motor GMFM values from the RCTs was not done in this review because gross motor outcome measurement, follow-up, patient characteristics, and surgical protocols varied across the centres. However, patient-level data from all trial investigators were obtained by McLaughlin et al,⁷³ who performed a primary meta-analysis.⁸⁸ The combined mean change scores both for GMFM-88 (4.53; $P = .002$) and GMFM-66 (2.66; $P = .002$) were statistically significantly improved in the rhizotomy-and-physical-therapy arm compared with the physical therapy-only arm. Regression analyses also confirmed a statistically significant direct relationship between the amount of dorsal tissue sectioned (cut) and the mean change in gross motor scores both for the GMFM-88 ($P < .001$) and the GMFM-66 ($P = .02$)—the more tissue sectioned the greater the score change.

The other two RCTs,^{89,90} both involving spastic diplegia, were performed at the Vancouver site and involved variations of dorsal rhizotomy (Table 1). In one trial,⁹⁰ patients receiving rhizotomy a year earlier were randomized to receive a year of therapeutic electrical stimulation (TES) for the abdominal and most proximal lower-limb muscles demonstrating weakness or to not receive TES. Parallel physical therapy continued in both groups. The mean change in GMFM-88 scores between the first and the second year was 5.5% in the TES-treated arm and 1.9% in the untreated arm. There were no significant differences between the trial arms in the 22 other secondary measurements involving spasticity, muscle strength, range of motion, seated postural control, or physiologic cost index.

The other RCT⁸⁹ compared children in a year-long intensified physical therapy program before dorsal rhizotomy with children not in an intensified physical therapy program before surgery. Both groups underwent standard physical therapy for 9 months after surgery. The mean change in GMFM scores at 2-year follow-up was the same, 10.4% and 10%, in the two arms. There were also no differences between the study arms in mean change scores for spasticity, range of motion, or muscle strength.

Table 1. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Randomized Controlled Studies

Study Groups Patient, n (M, F) Age, Mean ± SD (Range) ^a	GMFM Variant	Mean GMFM ± SD (Range)						
		Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change	12-month Post-op	Pre-op to 12-Month Post-op Mean Change	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change
RCT on DR and PT vs. PT alone								
Canada: Toronto, Wright et al,⁷⁵ 1998; 12-month follow-up—diplegia (n = 24)								
DR + PT 12 (7 M, 5 F) 57.8 months	GMFM-88 Total	51.9 ± 13.4	58.7 ± 13.5	6.8 ± 5.5	64.0 ± 13.2	12.0 ± 5.4	--	--
					Group difference <i>P</i> < .05			
PT 12 (7 M, 5 F) 58.3 months	GMFM-88 Total	56.5 ± 12.2	58.5 ± 10.7	2.0 ± 4.7	60.9 ± 12.5	4.4 ± 5.1	--	--
DR + PT	GMFM Item D	21.8 ± 15.9	30.1 ± 23.4	8.3 ± 8.2	33.1 ± 23.5	11.3 ± 8.2	--	--
					Group difference NS			
PT	GMFM Item D	19.6 ± 17.2	23.7 ± 12.1	4.1 ± 6.1	27.1 ± 19.6	7.5 ± 8.9	--	--
DR + PT	GMFM Item E	10.6 ± 8.2	14.8 ± 7.8	4.2 ± 3.3	23.4 ± 19.5	12.8 ± 6.1	--	--
					Group difference <i>P</i> < .05			
PT	GMFM Item E	13.2 ± 14.2	14.5 ± 15.4	1.3 ± 6.1	15.7 ± 17.1	2.5 ± 6.4	--	--
RCT on DR and PT vs. PT alone								
United States: Washington, McLaughlin et al,⁷³ 1998; 24-month follow-up—diplegia (n = 38)								
DR + PT 21 (11 M, 10 F) 6.4 ± 3.0 years (2.9–14.3 years)	GMFM-88 Total	70.3 ± 13.2	Graphic display	--	Graphic display	4.9 ± 7.6 0.8 (–3.5 to 5.0) NS	Graphic display	
PT 17 (12 M, 5 F) 7.2 ± 4.5 years (3.2–18.1 years)	GMFM-88 Total	71.3 ± 16.8	Graphic display	--	Graphic display	4.2 ± 5.3	Graphic display 7.2 ± 8.3	
DR + PT	GMFM-88 composite (Items C, D, E)	51.5 ± 20.2	Graphic display	--	Graphic display	7.0 ± 8.1 0.9 (–1.8 to 6.6) NS	Graphic display 9.6, 10.1	
PT	GMFM-88 composite (Items C, D, E)	53.0 ± 24.9	Graphic display	--	Graphic display	6.2 ± 9.0	Graphic display 9.8, 11.6 –0.2 (–7.4 to 7.1) NS	
RCT on DR and PT vs. PT alone								

Study Groups Patient, n (M, F) Age, Mean ± SD (Range) ^a	GMFM Variant	Mean GMFM ± SD (Range)						
		Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change	12-month Post-op	Pre-op to 12-Month Post-op Mean Change	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change
Canada: Vancouver, Steinbok et al,⁷⁴ 1997; 9-month follow-up—diplegia (N = 29)								
DR + PT 14 (NR) 50 months (35–75 months)	GMFM-88 Total	60.7 95% CI (51.4–70.0)	(at 9 months) 72.0	11.3 95% CI (7.4–15.2) Group difference P = .007	--	--	--	--
PT 15 (NR) 47 months (35–77 months)	GMFM-88 Total	62.7 95% CI (54.4–71.0)	(at 9 months) 67.9	5.1 95% CI (3.1–7.2)	--	--	--	--
DR + PT	GMFM-88 Item D	35.5	47.6	12.1	--	--	--	--
PT	GMFM-88 Item D	35.9	45.7	9.8	--	--	--	--
DR + PT	GMFM-88 Item E	20.7	31.1	10.4	--	--	--	--
PT	GMFM-88 Item E	18.6	23.1	4.4	--	--	--	--
RCT on DR and PT with TES vs. DR and PT only								
Canada: Vancouver, Steinbok et al,⁹⁰ 1997; 12-month follow-up—diplegia (N = 42)								
DR + TES 21 (NR) 7.2 years (4.3–10 years)	GMFM-88 Total	67	--	--	--	--	5.5 12–24 months P = .001	--
DR 21 (NR) 7.2 years (5.1–10.3 years)	GMFM-88 Total	69	--	--	--	--	1.9 12–24 months	--
RCT on IPT before DR and PT vs. DR without prior IPT and PT								
Canada: Vancouver, Steinbok et al,⁸⁹ 2002; 9-month follow-up—diplegia (N = 28)^a								
IPT + DR + IPT 13 (NR)	GMFM-88 Total	62.5	--	At 9 months 5.7	--	--	--	10.2 NS
DR + IPT 13 (NR)	GMFM-88 Total	60.2	--	At 9 months 11.6	--	--	--	10.4

Abbreviations: DR, dorsal rhizotomy; EP, electrophysiologic; F, female; GMAE, Gross Motor Activity Estimator; GMFM, gross motor function measure; GMPM, gross motor performance measure; IPT, Intensified Physical Therapy; m, male; n, number; NR, not recorded; NS, not significant; post-op, post-operative; pre-op, pre-operative; PT, physical therapy; RCT, randomized controlled trial; SD, standard deviation TES, Therapeutic Muscle Electrical Stimulation; VMFM, Vancouver Motor Function Measure.

^aTotals do not add because some patients were lost to follow-up.

Comparative Non-Randomized Trials

Five comparative non-randomized studies assessed gross motor function after dorsal rhizotomy. However, two of these studies^{91,92} involved a different sampling frame for their study populations. Each involved retrospective selection of patients identified through a registry from their motion analysis laboratories to compare functional gait outcomes in ambulatory children with cerebral palsy undergoing different surgical procedures, rhizotomy or orthopedic surgery. Children undergoing gait analyses at the motion laboratories are often referred from different institutions and for different reasons. Neurosurgical procedures and orthopedic surgery are also complementary rather than competitive surgeries, and orthopedic surgery would not be a comparator for rhizotomy in treating spasticity.

Results of the other three comparative nonrandomized studies⁹³⁻⁹⁵ are detailed in Table 2. The study by Steinbok et al⁹⁵ compared the impact of using or not using electrophysiologic guidance during rhizotomy on gross motor outcomes. Gross motor function, however, was evaluated using a combination of the GMFM and the Vancouver Motor Function Measure (VMFM), a locally developed unvalidated outcome measure. The mean percentage of dorsal rootlets cut was similar in the two groups—53% ± 11.5% in the electrophysiologic-guided group and 57% ± 3.2 in the unguided group. Mean gross motor function at 1 year significantly increased in both groups over baseline for the blended motor function score.

The study by Buckon et al⁹³ essentially compared patients' preference for dorsal rhizotomy over orthopedic surgery among children with diplegic cerebral palsy judged suitable for both rhizotomy and orthopedic soft tissue procedures. Parents received information from both surgical specialists. Of the 25 families, 18 chose dorsal rhizotomy and 7 chose orthopedic surgery. All children were either independently or dependently ambulatory. This study was the only one to evaluate gross motor function as well as the quality of movement with a standardized measure—the Gross Motor Performance Measure (GMPM). The GMPM is an outcome measure based on five attributes of movement: alignment, coordination, dissociated movement, stability, and weight shift.⁷⁷

The mean change scores for total GMPM movement improved significantly over baseline at 6 months and consistently improved at 1-year and 2-year follow-up for both surgical groups. Improvements in the individual domains of the mean GMPM scores over time, however, differed between surgical groups. Although improvements over baseline in three attributes were similar between surgical groups, improvements in dissociation and coordination consistently improved only after dorsal rhizotomy. The mean changes in the total GMFM-88 scores at 12 months improved (5.9 ± 4.89 , $P = .018$) over baseline in the orthopedic group but not in the dorsal rhizotomy group (3.4 ± 7.82 ; $P = .084$). At 2-year follow-up, mean score changes improved in the rhizotomy ($P = .011$) and in the orthopedic surgery ($P = .048$) group. At 2-year follow-up, the mean change for Item D (standing) improved over baseline in both surgical groups, whereas Item E (walking/running/jumping) improved only in the dorsal rhizotomy group.

The study by Engsborg et al⁹⁴ compared gross motor outcomes (GMFM-66) in ambulatory children with spastic diplegia undergoing dorsal rhizotomy and intensive physical therapy with several groups. Patients receiving rhizotomy were recruited from a clinic offering selective dorsal rhizotomy while those in the comparison group receiving physical therapy only were recruited from local and national advertising campaigns. An additional comparison group of children without disability were recruited by contacting parents who had brought their children to the hospital and were age-matched to children in the cerebral palsy group. The post-operative mean change in GMFM-66 scores at 20 months was significantly higher in the group treated with dorsal rhizotomy and physical therapy than in the group treated with physical therapy alone

(7.0 vs. 3.0; $P < .05$). Post-operative hip, ankle, and knee spasticity were significantly reduced in the dorsal rhizotomy and physical therapy group and were not significantly different from spasticity in the comparison group of children without disabilities.

Table 2. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Comparative Studies

Study Groups Patient, n (M, F) Age, Mean ± SD (Range) ^a	GMFM Variant	Mean GMFM ± SD (Range)						
		Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change	12-month Post-op	Pre-op to 12-Month Post-op Mean Change	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change
Comparative Non-Randomized Studies								
Canada: Vancouver, Steinbok et al,⁹⁵ 2009; 12-month follow-up—DR with or without EP guidance								
DR + EP guidance 22 (NR) 5.4 ± 4.2	GMFM-88/VMFM	--	--	--	--	8.5 ± 10.7	--	--
	--	--	--	--	--	5.2 ± 10.6 <i>P</i> = .34 for difference	--	--
United States: Portland, OR, Buckon et al,⁹³ 2004; 2-year follow-up of DR vs. orthopedic surgery—diplegia (n = 25)								
DR + PT 18 (15 M, 3 F) 71.3 months (49–123 months) (All ambulatory, 11 independent, 7 dependent)	GMFM-88 Total	82.1 ± 13.2	84.0 ± 12.9	1.98 ± 5.22, 95% CI (-0.62 to 4.58) <i>P</i> = .127	85.4 ± 12.8	3.39 ± 7.82, 95% CI (-0.49 to 7.27) <i>P</i> = .084	89.5 ± 11.1	6.32 ± 8.38, 95% CI (1.76–10.88) <i>P</i> = .011
Orthopedic surgery + PT 7 (4 M, 3 F) 78.6 months (51–132 months) (n = 7, 3 independent ambulatory, 4 dependent ambulatory)	GMFM-88 Total	78.2 ± 13.0	79.2 ± 10.4	0.96 ± 4.45, 95% CI (-3.15 to 5.07) <i>P</i> = .589	84.1 ± 8.8	5.90 ± 4.89, 95% CI (1.37–10.43) <i>P</i> = .018	85.7 ± 7.1	7.51 ± 8.04, 95% CI (0.07–14.95) <i>P</i> = .048
DR + PT	GMFM-88 Item D	65.7 ± 25.1	69.2 ± 26.6	3.56 ± 13.88, 95% CI (-3.34 to 10.46) <i>P</i> = .292	71.8 ± 26.3	6.13 ± 17.68, 95% CI (-2.67 to 14.93) <i>P</i> = .160	80.0 ± 21.4	12.14 ± 18.38, 95% CI (2.14–22.14) <i>P</i> = .022
Orthopedic surgery + PT	GMFM-88 Item D	56.4 ± 27.5	64.8 ± 20.1	8.43 ± 10.85, 95% CI (-1.60 to 18.46) <i>P</i> = .086	76.9 ± 12.0	20.51 ± 16.49, 95% CI (5.27–35.75) <i>P</i> = .017	81.0 ± 9.6	24.54 ± 21.85, 95% CI (4.33–44.75) <i>P</i> = .025
DR + PT	GMFM-88 Item E	55.0 ± 29.3	57.3 ± 31.2	2.32 ± 7.91, 95% CI (-1.63 to 6.27) <i>P</i> = .232	59.9 ± 32.6	4.86 ± 12.80, 95% CI (-1.51 to 11.23) <i>P</i> = .126	70.7 ± 30.3	14.4 ± 16.38, 95% CI (5.51–23.37) <i>P</i> = .004
Orthopedic surgery + PT	GMFM-88 Item E	41.7 ± 35.5	38.9 ± 29.5	-2.78 ± 11.73, 95% CI (-13.59 to 8.03) <i>P</i> = .554	48.2 ± 29.3	6.55 ± 8.81, 95% CI (-1.60 to 14.70) <i>P</i> = .097	53.4 ± 25.2	11.71 ± 19.08, 95% CI (-5.93 to 29.35) <i>P</i> = .156

Study Groups Patient, n (M, F) Age, Mean ± SD (Range) ^a	GMFM Variant	Mean GMFM ± SD (Range)						
		Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change	12-month Post-op	Pre-op to 12-Month Post-op Mean Change	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change
DR + PT	GMPM	54.6 ± 7.0	59.5 ± 7.7	4.89 ± 6.48, 95% CI (1.66–8.12) <i>P</i> < .005	59.6 ± 8.5	5.00 ± 6.58, 95% CI (1.73–8.27) <i>P</i> < .005	63.4 ± 7.2	8.13 ± 7.44, 95% CI (4.09–12.18) <i>P</i> < .001
Orthopedic surgery + PT	GMPM	54.1 ± 7.8	58.0 ± 7.4	3.86 ± 3.63, 95% CI (0.51–7.21) <i>P</i> < .031	59.6 ± 9.0	5.43 ± 4.86, 95% CI (0.93–9.93) <i>P</i> < .025	60.7 ± 9.4	6.67 ± 7.55, 95% CI (–0.40 to 13.54) <i>P</i> < .061
United States: St Louis, Engsborg et al,⁹⁴ 2006; 2-year follow-up—DR vs. PT vs. age-matched, able-bodied controls—ambulatory diplegia, 55 independent, 8 dependent								
DR + PT 31 (15 M, 16 F) 9 ± 5.3 years	GMFM-66	87 ± 10	At 8 months, 88 ± 9	--	--	--	At 20 months, 92 ± 8 <i>P</i> < .05	--
PT 37 (19 M, 18 F) 9.7 ± 4.5 years	GMFM-66	89 ± 7	At 8 months, 90 ± 7	--	--	--	At 20 months, 91 ± 7 <i>P</i> < .05	--
DR + PT	GMAE	70 ± 12	At 8 months, 72 ± 12	--	--	--	At 20 months, 77 ± 13	--
PT	GMAE	71 ± 8	At 8 months, 73 ± 10	--	--	--	At 20 months, 74 ± 9	--

Abbreviations: DR, dorsal rhizotomy; EP, electrophysiologic; F, female; GMAE, Gross Motor Activity Estimator; GMFM, gross motor function measure; GMPM, gross motor performance measure; IPT, Intensified Physical Therapy; m, male; n, number; NR, not recorded; NS, not significant; post-op, post-operative; pre-op, pre-operative; PT, physical therapy; RCT, randomized controlled trial; SD, standard deviation; SE, standard error; TES, Therapeutic Muscle Electrical Stimulation; VMFM, Vancouver Motor Function Measure.

Nine observational pre-post cohort studies (Table 3) evaluated gross motor function with the GMFM, either as total scores or only for the higher skill levels of standing (Item D) or walking/running/jumping (Item E). Studies were grouped according to the presenting level of motor disability of study subjects. Six studies⁹⁶⁻¹⁰¹ involved ambulatory patients with diplegia and minimal motor disability (GMFCS Levels I, II, or III) and are referred to as Group A in Table 3. One study⁹⁹ targeted an older population of adolescents and young adults (mean age 26 years) with 4-month follow-up. Three other studies¹⁰²⁻¹⁰⁴ involved populations with moderate or mixed levels of motor disability and are referred to as Group B in Table 3. Nordmark et al¹⁰⁴ studied ambulatory and nonambulatory children with spastic diplegia (GMFCS Levels I–V); McLaughlin et al¹⁰³ studied children with either spastic quadriplegia or spastic diplegia; and Hodgkinson et al¹⁰² studied children with spastic quadriplegia.

In the four studies of patients with minimal motor disability that reported total GMFMs (Group A), three^{97,98,101} reported statistically significant improvements in mean total GMFMs at 12-month follow-up. Chan et al⁹⁷ reported changes in motor function for dependent (GMFCS Levels II–III) and independent (GMFCS Level I) ambulatory status. Although GMFM total scores increased in both groups at follow-up, the gains for only dependent ambulatory status were significant ($P = .02$) at 12-month follow-up. The study by Reynolds et al⁹⁹ was the fourth reporting GMFM total scores, but for an older population. Increased GMFM mean scores at the 4-month follow-up had not improved significantly for total or for subdomain GMFM scores for Item D (standing) or Item E (walking/running/jumping).

Three studies¹⁰²⁻¹⁰⁴ compared how various levels of gross motor disability affected gross motor gains after dorsal rhizotomy (Group B). In the study by Nordmark et al,¹⁰⁴ improvement in GMFM total scores over baseline was statistically significant ($P = .017$) and clinically important ($> 6\%$) for ambulatory children with diplegia and less motor disability (GMFCS Levels II and III) but was not significant ($P = .168$) for nonambulatory children with diplegia and more disability (GMFCS Levels IV and V). The same pattern was noted for the “goal-specific” GMFM totals, in which scores improved in both groups but improved significantly only in the ambulatory group.

The higher-level subdomains Item D (standing) and Item E (walking/running/jumping) were statistically significant and clinically important ($> 6\%$) for ambulatory patients at 12-month follow-up. However, nonambulatory patients (GMFCS Levels IV and V) whose pre-operative scores were close to zero for Items D and E had no gains. Although they gained lower-level skills reflected in Item A (lying/rolling) from 59.4 ± 23 to 73.9 ± 24.7 ; $P = .216$, in Item B (sitting) from 57.9 ± 19.8 to 66.3 ± 21.9 ; $P = .417$, and in Item C (crawling/kneeling) from 42.0 ± 23.4 to 48.8 ± 28.1 ; $P = .519$, none of these gains were statistically significant.

McLaughlin et al¹⁰³ compared gains in gross motor skills between higher-functioning ambulatory diplegia patients with nonambulatory quadriplegia patients. The gains in mean total GMFM scores were similar in the two groups (9% and 9.8%), and improvements were statistically and clinically significant in both spastic diplegia and quadriplegia. All subdomains (Items A–E) significantly improved among children with spastic diplegia. The largest gains were in the higher-order skills of Items D (13.3%) and E (13.7%). The mean pre-operative scores in the lower subdomains (Items A–C) were already high for this group. For quadriplegia, only the gross motor gains in the lower motor skills Item A (lying/rolling), Item B (sitting), and Item C (crawling/kneeling) were significant. The gains for these lower motor skills were much higher for spastic quadriplegia than for diplegia in each subdomain. The gain in Item B (sitting) was particularly high, a 23-point increase from a pre-operative score of 37 points.

In the study by Hodgkinson et al,¹⁰² baseline scores for the quadriplegia population were particularly low, especially for the higher-order motor skills of standing or running and jumping.

Small gains in motor skills for the individual GMFM subdomains at 1-year follow-up were not statistically significant.

Table 3. Short-Term Impact of Dorsal Rhizotomy on Gross Motor Function in Pre-Post Cohort Studies

Gross Motor Classification	GMFM Variant	Mean GMFM ± SD (Range)					
		Pre-op	6-Month Post-op	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change (95% CI)
Group A. Minimal Motor Disability							
Germany, Berlin, Funk et al,⁹⁸ 2015: 24-month follow-up—diplegia (n = 54)							
54 patients (29 M, 25 F), mean age ± SD = 6.9 ± 2.9 years							
GMFCS Levels I or II (n = 54)	GMFM-88	79	--	84	<i>P</i> < .001	86	<i>P</i> = .002
Hong Kong, Chan et al,⁹⁷ 2008: 12-month follow-up—diplegia (n = 20), quadriplegia (n = 1)							
21 patients (12 M, 9 F), mean age ± SD 8.6 ± 2.6 years							
GMFCS Level I (n = 8), Level II (n = 4), Level III (n = 2)	GMFM-88 Total for all	77.1 ± 18.8	--	80.9 ± 17.4	3.8 ± 4.5 (1.7–5.8) <i>P</i> = .001	--	--
	GMFM-88 Item E for all	48.6 ± 36.2	--	54.3 ± 37.4	5.8 ± 11.1 (0.7–10.8) <i>P</i> = .027	--	--
	GMFM-88 Total for Level I	94.3 ± 4.7	--	96.1 ± 2.9	1.9 ± 2.4 (–0.1 to 3.8) <i>P</i> = .059	--	--
	GMFM-88 Level I, Item E	84.6 ± 17.8	--	89.5 ± 11.4	4.9 ± 9.7 (–3.2 to 13.0) <i>P</i> = .199	--	--
	GMFM-88 Total for Levels II, III	70.2 ± 14.7	--	74.8 ± 14.6	4.6 ± 5.6 (0.9–8.4) <i>P</i> = .021	--	--
	GMFM-88 Levels II, III, Item E	31.1 ± 23.7	--	37.1 ± 31.2	6.0 ± 13.3 (–2.9 to 14.9) <i>P</i> = .164	--	--
Italy, Conegliano, Carraro et al,⁹⁶ 2014: 12-month follow-up—diplegia (n = 9)							
9 patients (6 M, 3 F), mean age ± SD 7.9 ± 3.2 years							
GMFCS Level II (n = 7), Level III (n = 2)	GMFM Item D	32 (21–39)	--	32 (22–39)	<i>P</i> = .547	--	--
	GMFM Item E	46 (12–67)	--	47 (19–72)	<i>P</i> = .438	--	--
Netherlands, Amsterdam, van Schie et al,¹⁰¹ 2005: 12-month follow-up—diplegia (n = 9, all ambulatory)							
9 patients (4 M, 5 F), mean age 65 months (range 43–82 months)							
GMFCS Level II (n = 1), Level III (n = 8)	GMFM-88	62.8 (51.2–84.6)	--	71.5 (60–90) <i>P</i> < .05	--	--	--

Gross Motor Classification	GMFM Variant	Mean GMFM ± SD (Range)					
		Pre-op	6-Month Post-op	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Mean Post-op Change (95% CI)
United States, Lexington, KY, Sacco et al,¹⁰⁰ 2000: 12-month follow-up—diplegia (n = 10, all ambulatory)							
10 patients (NR), mean age 8.5 years (range 5–16 years)							
--	GMFM Item D	47.2	--	52.5	<i>P</i> = .2858	--	--
--	GMFM Item E	22.0	--	31.2	<i>P</i> = .0324	--	--
United States, St Louis, Mo, Reynolds et al,⁹⁹ 2011: 4-month follow-up—diplegia (n = 21, all ambulatory, older ages)							
21 patients (15 M, 6 F), mean age 26 years (range 18–39 years)							
GMFCS Level I (n = 15), Level II (n = 28)	GMFM Total	87.1 ± 3.7	93.4 ± 1.7	<i>P</i> = .09	--	--	--
	GMFM Item C	94.9 ± 1.1	98.7 ± 0.69	<i>P</i> = .02	--	--	--
	GMFM Item D	78.6 ± 6.9	89.0 ± 2.7	<i>P</i> = .10	--	--	--
	GMFM Item E	65.6 ± 10.7	80.7 ± 5.8	<i>P</i> = .12	--	--	--
Group B. Moderate Disability or Mixed Disability Study Groups							
Sweden, Lund, Nordmark et al,¹⁰⁴ 2000: 12 month follow-up—diplegia (n = 18 nonambulatory, 10 ambulatory)							
18 patients (12 M, 6 F), mean age ± SD 4.3 ± 1.0 years							
GMFCS All Levels Level II (n = 3), Level III (n = 7), Level IV (n = 7), Level V (n = 1)	GMFM-88 Total for all	48.2 ± 18.9 (19–88)	52.1 ± 16.6 (17–72)	57.8 ± 20.1 (21–89)	9.6 ± 6.5 <i>P</i> = .003	--	--
	GMFM-88 goal for all	44.6 ± 14.4 (23–82)	48.5 ± 12.8 (21–76)	56.8 ± 15.7 (22–84)	12.2 ± 5.0 <i>P</i> = .001	--	--
	GMFM-88 Item D for all	24.9 ± 22.8 (0–69)	26.8 ± 24.6 (0–82)	33.0 ± 28.5 (0–90)	8.1 ± 8.6 <i>P</i> = .027	--	--
	GMFM-88 Item E for all	16.9 ± 21.8 (0–82)	17.5 ± 20.0 (0–76)	24.8 ± 28.2 (0–82)	7.9 ± 8.4 <i>P</i> = .06	--	--
GMFCS Levels II and III (n = 10)	GMFM-88 Total for Levels II and III	60.2 ± 16.4 (36–88)	64.3 ± 6.9 (49–72)	72.2 ± 9.2 (62–89)	12 ± 5.9 <i>P</i> = .02	--	--
	GMFM-88 goal for Levels II and III	46.8 ± 16.2 (28–82)	51.7 ± 11.8 (37–76)	63.5 ± 11.2 (46–84)	16.7 ± 6.3 <i>P</i> = .003	--	--
	GMFM-88 Item D for Levels II and III	40.4 ± 18.9 (18–69)	43.8 ± 20.4 (23–82)	53.2 ± 21.5 (28–90)	12.8 ± 9.1 <i>P</i> = .01	--	--
	GMFM-88 Item E for Levels II and III	29.3 ± 22.5 (14–82)	30.2 ± 18.6 (14–76)	43.0 ± 25.8 (15–82)	13.7 ± 10.8 <i>P</i> = .03	--	--
GMFCS Levels IV and V (n = 8)	GMFM-88 Total for Levels IV and V	33.3 ± 7.9 (19–42)	36.8 ± 11.1 (17–55)	39.8 ± 14.2 (21–59)	6.5 ± 5.7 <i>P</i> = .17	--	--

Gross Motor Classification	GMFM Variant	Mean GMFM ± SD (Range)					
		Pre-op	6-Month Post-op	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Mean Post-op Change (95% CI)
	GMFM-88 goal for Levels IV and V	41.8 ± 12.3 (23–55)	44.5 ± 13.6 (21–59)	48.4 ± 17. (22–69)	6.6 ± 7.4 <i>P</i> = .22	--	--
	GMFM-88 Item D for Levels IV and V	5.5 ± 5.2 (0–15)	5.5 ± 5.1 (0–13)	7.8 ± 8.1 (0–21)	2.3 ± 3.4 <i>P</i> = .80	--	--
	GMFM-88 Item E for Levels IV and V	1.4 ± 2.7 (0–7)	1.6 ± 2.4 (0–6)	2.1 ± 2.9 (0–8)	0.7 ± 1.4 <i>P</i> = .80	--	--
United States, Seattle, Wash, McLaughlin et al,¹⁰³ 1994: 12-month follow-up—diplegia and quadriplegia							
34 patients (21 M, 13 F): diplegia, mean age ± SD 8.0 ± 3.9 years (range 4.7–21.3 years); quadriplegia, mean age ± SD 7.2 ± 3.4 (range 3.3–13 years)							
Diplegia (n = 24: independent ambulatory (n = 10), assisted ambulatory (n = 12), nonambulatory (n = 2))	GMFM-88 Total for diplegia	68	--	--	9.8 ± 8.3 <i>P</i> < .0001	--	--
	GMFM-88 Item A for diplegia	97	--	--	2.4 ± 4.8 <i>P</i> = .02	--	--
	GMFM-88 Item B for diplegia	87	--	--	9.4 ± 11.4 <i>P</i> = .001	--	--
	GMFM-88 Item C for diplegia	83	--	--	8.6 ± 9.7 <i>P</i> < .001	--	--
	GMFM-88 Item D for diplegia	45	--	--	13.3 ± 9.9 <i>P</i> = .003	--	--
	GMFM-88 Item E for diplegia	32	--	--	13.7 ± 15.6 <i>P</i> < .0001	--	--
Quadriplegia (n = 10: assisted ambulatory (n = 2), nonambulatory (n = 8))	GMFM-88 Total for quadriplegia	32	--	--	9 ± 3.4 <i>P</i> < .0001	--	--
	GMFM-88 Item A for quadriplegia	81	--	--	7 ± 7.6 <i>P</i> = .02	--	--
	GMFM-88 Item B for quadriplegia	37	--	--	23 ± 13.0 <i>P</i> < .0001	--	--
	GMFM-88 Item C for quadriplegia	32	--	--	14.7 ± 9.0 <i>P</i> = .001	--	--
	GMFM-88 Item D for quadriplegia	8	--	--	1.2 ± 5.9 <i>P</i> = .54	--	--
	GMFM-88 Item E for quadriplegia	4	--	--	0.1 ± 4.2 <i>P</i> = .94	--	--

Gross Motor Classification	GMFM Variant	Mean GMFM ± SD (Range)					
		Pre-op	6-Month Post-op	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Mean Post-op Change (95% CI)
France, Lyon, Hodgkinson et al,¹⁰² 1997: 12-month follow-up—spastic quadriplegia and triplegia							
18 patients (12 M, 6 F), mean age 9 years (range 5.5–16.5 years); 15 spastic quadriplegia, 3 spastic triplegia							
	GMFM-88 Total	41 ± 18.5	--	44 ± 17.5	3.0 ± 6.0 <i>P</i> = .02	--	--
	GMFM-88 Item A	87 ± 12.1	--	91 ± 7.5	4.0 ± 3.4 <i>P</i> = .07	--	--
	GMFM-88 Item B	54 ± 31.1	--	59 ± 28.2	5.0 ± 9.9 <i>P</i> = .32	--	--
	GMFM-88 Item C	44 ± 33.9	--	46 ± 33.0	2.0 ± 11.2 <i>P</i> = .10	--	--
	GMFM-88 Item D	13 ± 16.2	--	15 ± 16.5	2.0 ± 5.5 <i>P</i> = .40	--	--
	GMFM-88 Item E	8 ± 11.2	--	7.2 ± 8.7	0.1 ± 3.3 <i>P</i> = .89	--	--

Abbreviations: CI, confidence interval; F, female; GMFCS, Gross Motor Function Classification System; GMFM, gross motor function measure; M, male; NR, not reported; post-op, post-operative; pre-op, pre-operative; SD, standard deviation.

Short-Term Effectiveness for Functional Independence and Caregiver Assistance

Ten reports, from nine institutions based in eight countries, evaluated short-term effectiveness of dorsal rhizotomy on functional independence of children with spastic cerebral palsy. Five studies evaluated short-term effectiveness of functional independence with the Pediatric Evaluation Disability Inventory (PEDI) assessments (Table 4), another five studies with the Functional Independence Measure for Children (WeeFIM) assessments (Table 5). All reports involved observational pre-post longitudinal cohort studies.

The PEDI is a multidimensional inventory that evaluates functional independence in activities of daily living and caregiver assistance for disabled children in three domains: self-care, mobility, and social function.¹⁰⁵ The PEDI is a reliable and validated inventory for both functional performance and caregiver assistance among children with cerebral palsy as well.¹⁰⁶⁻¹⁰⁸ Total scores for the PEDI subdomains are based on a scale from 0 to 100 in which higher scores represent increasing abilities or independence. Needs for caregivers' assistance with activities of daily living were also assessed (scores from 0 to 100) over the same three domains (self-care, mobility, and social function). For this measure there is an inverse relationship; increasing scores indicate less need for caregivers' assistance. A 10-point or greater change in PEDI scores has been estimated to represent a clinically important or relevant difference.¹⁰⁹ Much less is known about developmental milestones for functional independence than about motor function among children with cerebral palsy. An early follow-up study of children with cerebral palsy indicated that, even with intensive therapy programs, children who had not achieved independence in their activities of daily living by age 4 were unlikely to achieve it at any age.¹¹⁰

In this review, the five studies evaluating PEDI functional outcomes reported on different subdomains of the multidimensional inventory; only Nordmark et al¹⁰⁴ and Buckton et al⁹³ reported on all six subdomains (Table 4). Social function was omitted most often, in both functional independence and caregiver assistance domains. All studies involved 12-month follow-up of patients with diplegia except the study by Dudgeon et al¹¹¹ on both spastic diplegia and quadriplegia and the study by Buckton et al,⁹³ which involved 24-month follow-up.

All studies reported statistically significant and clinically important improvements in functional independence for both self-care and mobility among children with spastic diplegia, with mild or moderate (GMFCS Levels I–III), or with severe (GMFCS Levels IV–V) motor disability. In the study by Dudgeon et al¹¹¹ (on functional independence in spastic quadriplegia as well as in spastic diplegia), improvements seen for children with spastic diplegia were not seen for those with spastic quadriplegia. There were no improvements in functional independence for either their self-care or mobility, and caregiver assistance was not evaluated. Functional improvement in the upper limbs was also evaluated in this study, and no improvements were reported for children with either spastic diplegia or quadriplegia. Social function was evaluated in two studies.^{93,104} Both reported statistically significant improvement for children with spastic diplegia with mild to moderate or severe motor disability.

All studies evaluating caregiver assistance also reported less reliance in subdomains of both self-care and mobility for spastic diplegia with mild to moderate or severe motor disability. In the study by Dudgeon et al¹¹¹ evaluating both diplegia and quadriplegia, the need for caregivers' assistance was not reported. The burden related to caregivers' assistance for social function was evaluated in two studies,^{93,104} and both reported statistically significant improvement. In the study by Buckton et al⁹³ with 2-year follow-up, improvement continued in all domains for both functional independence and caregiver assistance over the 1-year to 2-year interval among mildly disabled children with spastic diplegia.

Chan et al⁹⁷ were the only researchers who evaluated outcomes based on the specific treatment goals for individual children in addition to PEDI assessments. They evaluated these goals with the Canadian Occupational Performance Measure, a family-centred or client-centred decision aid that guides children or their caregivers to identify problems in areas of self-care, productivity, or leisure.^{112,113} Respondents select up to five problems as treatment goals and rate them on a scale of 1 to 10 for their current performance of that item and then rate their satisfaction with performance of that item. In that study, many different skills were identified as treatment objectives in the self-care (9 skills were reported 28 times), functional mobility (12 skills were reported 35 times), recreation (9 skills were reported 13 times), and other problem areas (6 skills were reported 15 times). The mean total Canadian Occupational Performance Measure for the entire study group improved significantly at 6 months, in both performance score and satisfaction score.

Table 4. Short-Term Effectiveness of Dorsal Rhizotomy for Functional Independence and Caregiver Assistance Evaluated With PEDI

Outcome Measure	Mean PEDI ± SD (Range) ^a						
	Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change (95% CI)	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change (95% CI)
Hong Kong, Chan et al,⁹⁷ 2008; 12-month follow-up—diplegia (n = 20) and quadriplegia (n = 1)							
Study group: 20 patients (12 M, 9 F), mean age ± SD 8.6 ± 2.6 years							
GMFCS Level I (n = 8)							
PEDI-FS self-care	80 ± 19.0	--	--	85.4 ± 8.6	5.3 ± 4.6 (1.0–9.5) P = .023	--	--
PEDI-FS mobility	83.9 ± 14.2	--	--	94.4 ± 12.1	10.5 ± 11.1 (1.2–19.8) P = .032	--	--
PEDI-CA self-care	81.6 ± 8.8	--	--	96.1 ± 6.9	14.6 ± 10.8 (4.5–24.6) P = .012	--	--
PEDI-CA mobility	84.5 ± 13.4	--	--	91.9 ± 12.5	7.4 ± 9.7 (-0.7 to 15.5) P = .069	--	--
GMFCS Levels II–III (n = 11)							
PEDI-FS self-care	69.9 ± 11.0	--	--	78.8 ± 12.0	8.9 ± 6.1 (4.8–13.0) P = .001	--	--
PEDI-FS mobility	59.6 ± 10.4	--	--	65.5 ± 12.5	5.9 ± 7.9 (0.6–11.2) P = .032	--	--
PEDI-CA self-care	73.0 ± 14.9	--	--	84.5 ± 18.4	11.5 ± 9.1 (5.4–17.7) P = .002	--	--
PEDI-CA mobility	58.5 ± 6.8	--	--	63.7 ± 8.3	5.2 ± 5.5 (1.5–8.9) P = .011	--	--
COPM all (n = 21)							
COPM performance total	23.0 ± 8.7	28.9 ± 10.4	5.8 ± 7.8 (2.2–9.4) P = .003	--	--	--	--
COPM satisfaction total	23.4 ± 9.1	28.3 ± 11.0	4.9 ± 7.9 (1.3–8.5) P = .011	--	--	--	--

Outcome Measure	Mean PEDI ± SD (Range) ^a						
	Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change (95% CI)	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change (95% CI)
Netherlands, Amsterdam, van Schie et al,¹⁰¹ 2005; 12-month follow-up—diplegia (n = 9, all ambulatory), GMFCS Level II (n = 1) and GMFCS Level III (n = 8)							
9 patients (4 M, 5 F), mean age 65 months (range 43–82 months)							
PEDI-FS self-care	63.3	69.2	<i>P</i> < .005	--	--	--	--
PEDI-CA self-care	59.0	63.4	<i>P</i> < .05	--	--	--	--
Sweden: Lund, Nordmark et al,¹⁰⁴ 2000; 12-month follow-up—diplegia (n = 18, 10 ambulatory)							
18 patients (12 M, 6 F)							
PEDI-FS self-care	56.3 ± 11.9 (37.0–74.7)	62.2 ± 12.2 (43.6–81.4)	--	64.6 ± 12.8 (40.4–93.0)	8 ± 4.1 <i>P</i> < .001	--	--
PEDI-FS mobility	49.1 ± 15.9 (15.2–73.3)]	59.9 ± 15.7 (33.4–89.2)	--	61.7 ± 15.5 (33.4–89.2)	12.6 ± 5.2 <i>P</i> < .001	--	--
PEDI-FS social function	62.4 ± 12.3 (37.9–89.1)	67.4 ± 15.1 (37.0–89.1)	--	70.2 ± 13.2 (41.1–89.0)	7.8 ± 4.3 <i>P</i> < .001	--	--
PEDI-CA self-care	49.9 ± 15.4 (29.2–69.6)	57.1 ± 14.5 (29.2–76.7)	--	58.4 ± 17.9 (25.4–100.0)	8.5 ± 5.6 <i>P</i> = .001	--	--
PEDI-CA mobility	48.4 ± 21.6 (0–78.3)	57.1 ± 20.2 (20.3–82.5)	--	60.7 ± 18.4 (20.3–100.0)	12.3 ± 6.7 <i>P</i> < .001	--	--
PEDI-CA social function	64.3 ± 24.5 (11.3–100.0)	71.2 ± 24.3 (11.3–100.0)	--	74.6 ± 23.2 (11.3–100.0)	6.9 ± 8.1 <i>P</i> = .001	--	--
Mild or moderate disability, GMFCS Levels II or III (n = 10)							
PEDI-FS self-care	64.9 ± 7.1 (53.7–74.7)	71.2 ± 8.1 (59.9–81.4)	--	72.4 ± 10.4 (55.6–93.0)	7.5 ± 4.0 <i>P</i> = .004	--	--
PEDI-FS mobility	60.5 ± 9.6 (41.4–73.3)	71.1 ± 9.7 (58.2–89.1)	--	71.9 ± 10.6 (58.2–89.2)	11.4 ± 4.5 <i>P</i> = .007	--	--
PEDI-FS social function	70.2 ± 8.1 (62.3–89.1)	77.7 ± 9.7 (65.1–89.1)	--	78.0 ± 8.3 (66.2–89.1)	7.8 ± 3.7 <i>P</i> = .02	--	--
PEDI-CA self-care	61.8 ± 6.9 (45.9–69.6)	67.4 ± 7.0 (55.7–76.7)	--	69.0 ± 13.6 (53.4–100)	7.2 ± 4.8 <i>P</i> = .04	--	--
PEDI-CA mobility	64.3 ± 8.2 (53.6–78.3)	72.9 ± 7.4 (61.8–82.5)	--	72.5 ± 12.1 (56.1–100)	8.2 ± 4.6 <i>P</i> = .002	--	--

Outcome Measure	Mean PEDI ± SD (Range) ^a						
	Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change (95% CI)	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change (95% CI)
PEDI-CA social function	80.1 ± 13.2 (61.3–100)	87.1 ± 9.6 (70.0–100)	--	87.0 ± 10.1 [67.6–100]	6.9 ± 5.3 <i>P</i> = .07	--	--
Severe disability, GMFCS Levels IV and V (n = 8)							
PEDI-FS self-care	45.5 ± 6.2 (37.0–51.7)	51.9 ± 6.3 (43.6–64.6)	--	54.8 ± 8.1 (40.4–68.3)	9.3 ± 3.6 <i>P</i> = .001	--	--
PEDI-FS mobility	34.8 ± 8.8 (15.2–43.3)	46.0 ± 8.7 (33.4–57.3)	--	48.9 ± 10.0 (33.4–59.1)	14 ± 4.7 <i>P</i> = .001	--	--
PEDI-FS social function	52.6 ± 9.3 (37.9–68.9)	54.4 ± 9.5 (37.0–70.8)	--	60.5 ± 11.8 (41.4–82.2)	7.9 ± 5.3 <i>P</i> = .005	--	--
PEDI-CA self-care	35.1 ± 7.5 (29.2–48.6)	43.7 ± 9.3 (29.2–56.8)	--	45.2 ± 13.5 (25.4–64.5)	10.0 ± 5.5 <i>P</i> = .019	--	--
PEDI-CA mobility	28.5 ± 15.0 (0–12.8)	37.4 ± 10.8 (20.3–47.2)	--	45.9 ± 13.5 (20.3–65.0)	17.0 ± 7.1 <i>P</i> = .001	--	--
PEDI-CA social function	44.5 ± 20.8 (11.3–65.4)	51.4 ± 22.5 (11.3–100)	--	59.3 ± 26.3 (11.3–100)	14.8 ± 11.9 <i>P</i> = .005	--	--
United States: Portland (OR), Buckon et al,⁹³ 2004; 24-month follow-up—diplegia (n = 25, all ambulatory: 17 independent, 6 assisted)							
25 patients (15 M, 3 F), mean age 71.3 months (range 49–123 months)							
PEDI-FS self-care	73.7 ± 13.1	76.9 ± 12.2	3.27 ± 4.37 (1.10–5.44) <i>P</i> = .006	79.8 ± 13.5	6.18 ± 6.91 (2.74 to 9.62) <i>P</i> = .001	84.1 ± 14.2	11.89 ± 6.81 (8.18–15.60) <i>P</i> = .0001
PEDI-FS mobility	70.5 ± 10.1	71.9 ± 9.3	1.41 ± 3.80 (-0.49 to 3.31) <i>P</i> = .134	74.2 ± 9.9	6.18 ± 6.91 (-0.22 to 7.68) <i>P</i> = .063	77.8 ± 10.4	7.51 ± 7.11 (3.63–11.39) <i>P</i> = .001
PEDI-FS social	69.2 ± 8.8	72.4 ± 11.8	1.22 ± 5.95 (-1.73 to 4.17) <i>P</i> = .039	72.7 ± 11.8	3.19 ± 6.56 (-0.08 to 6.46) <i>P</i> = .055	75.0 ± 7.9	7.82 ± 6.63 (4.21–11.43) <i>P</i> = .0004
PEDI-CA self-care	74.0 ± 18.5	76.9 ± 15.4	2.82 ± 9.77 (-2.02 to 7.68) <i>P</i> = .236	77.1 ± 12.4	3.06 ± 10.73 (-2.28 to 8.40) <i>P</i> = .22	82.5 ± 14.1	10.53 ± 8.33 (5.99–15.07) <i>P</i> = .0002
PEDI-CA mobility	81.0 ± 17.5	81.8 ± 15.2	0.78 ± 5.15 (-1.77 to 3.33) <i>P</i> = .530	89.0 ± 13.1	8.0 ± 11.97 (2.06 to 13.96) <i>P</i> = .011	94.0 ± 10.0	13.58 ± 13.76 (6.09–21.07) <i>P</i> = .002

Outcome Measure	Mean PEDI ± SD (Range) ^a						
	Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change (95% CI)	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change (95% CI)	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change (95% CI)
PEDI-CA social	87.0 ± 12.8	88.1 ± 10.7	1.12 ± 13.56 (-5.63 to 7.87) P = .730	90.1 ± 10.4	3.07 ± 10.40 (-2.0 to 8.24) P = .228	92.1 ± 8.8	7.00 ± 10.31 (1.39–12.61) P = .02
United States: Seattle (WA), Dudgeon et al,¹¹¹ 1994; 12-month follow-up—diplegia (n = 20) and quadriplegia (n = 9)							
29 patients (NR), mean age ± SD 8.1 ± 4.1 years (range 3.7–22.0 years)							
Diplegia							
PEDI-FS self-care	70.41 ± 16.09	76.57 ± 16.58	6.01 (-1.2 to 22.7) P = .0005	78.88 ± 13.49	7.82 (0 to 21) P = .0051	--	--
PEDI-FS mobility	56.48 ± 11.23	63.93 ± 12.44	8.09 (-3.6 to 32.8) P = .0003	72.78 ± 15.18	16.8 (7.7 to 30.1) P = .0033	--	--
Upper limb—basic hand placement	12.95 ± 1.82	13.63 ± 8.3	0.79 (0 to 4) P = .0277	13.45 ± 0.93	0.82 (0 to 5) P = .0679	--	--
Upper limb—cube stacking	7.53 ± 3.88	9.11 ± 2.78	2.08 (-4 to 12) P = .0619	10.09 ± 1.87	3.91 (-2 to 12) P = .0469	--	--
Upper limb—scooping water	13.83 ± 8.38	17.28 ± 10.93	2.07 (-9 to 16) P = .0348	19.18 ± 11.45	3.55 (-7 to 22) P = .2135	--	--
Quadriplegia							
PEDI-FS self-care	49.74 ± 13.84	51.26 ± 14.56	1.51 (-5.4 to 6.0) P = .2489	47.46 ± 25.0	2.36 (-12.0 to 11.4) P = .715	--	--
PEDI-FS mobility	36.64 ± 9.04	35.64 ± 10.13	-1.08 (-9.3 to 3) P = .6858	41.98 ± 12.86	3.92 (0 to 11.1) P = .0679	--	--
Upper limb task—basic hand placement	7.78 ± 3.93	9.01 ± 4.80	1.57 (-4 to 7) P = .2945	10.6 ± 5.4	1.8 (-2 to 5) P = .2012	--	--
Upper limb task—cube stacking	5.44 ± 3.08	5.56 ± 2.78	-0.14 (-3 to 2) P = .675	6.00 ± 4.3	0.6 (-4 to 6) P = .715	--	--
Upper limb task—scooping water	5.89 ± 5.37	9.11 ± 8.39	4.14 (0–11) P = .0431	10.2 ± 7.95	3.6 (0 to 8) P = .0679	--	--

Abbreviations: CA, caregiver assistance; CI, confidence interval; COPM, Canadian Occupational Performance Measure; F, female; FS, functional status; GMFCS, Gross Motor Function Classification System; m, male; n, number; NR, no response; PEDI, Pediatric Evaluation of Disability Inventory; post-op, post-operative; pre-op; pre-operative; SD, standard deviation.

^aUnless otherwise indicated.

Functional independence in activities of daily living was also evaluated using the Functional Independence Measure for Children (WeeFIM). The WeeFIM is a reliable and validated multidimensional outcome scale that evaluates disability and focuses on performance.^{114,115} It does not provide a separate measure of caregiver assistance. The WeeFIM scale consists of 18 measures in 6 areas: self-care (6 items), sphincter control (2 items), transfers (3 items), locomotion (2 items), communication (2 items), and social cognition (3 items).¹¹⁴ These are summed into three subscales: self-care (self-care, sphincter control); motor (transfers and locomotion), and cognitive (communication and social cognition). Each item is rated on a 7-level ordinal scale from 1 (total assistance) to 7 (complete independence). Scores of 1 to 4 indicate some degree of assistance; a score of 5 indicates supervision is required; scores of 6 or 7 indicate no help is required. The WeeFIM total score ranges from 18 (total dependence) to a maximum of 126 (complete independence in all skills). Maximum total scores are 56 for self-care, 35 for motor, and 35 for cognitive subscales.

Results of the five studies employing the WeeFIM to evaluate functional independence in children with spastic cerebral palsy are outlined in Table 5. How WeeFIM scores were reported varied across the studies with various subscales reported and combined and various formats used for scoring. The self-care subscale was reported in only one study.¹¹⁶ Study populations differed; three studies¹¹⁶⁻¹¹⁸ included children with severe gross motor disabilities or quadriplegia. In the study by Tichy et al,¹¹⁸ older children (mean age 16.5 years) with spastic quadriplegia were included.

Studies reporting total scores for the WeeFIM showed statistically significant improvements in functional independence.^{96,117} The self-care subscale that was reported separately only in the one study¹¹⁶ was also the only study that compared changes in self-care between children with spastic diplegia and quadriplegia; both showed gains. This was also the only study to evaluate sphincter control; those with spastic quadriplegia had much greater gains in sphincter control than those with spastic diplegia. Children with spastic diplegia, however, had greater gains in mobility.

Along with functional assessments by WeeFIM, improvements in upper-limb function assessed by the Quality of Upper Extremity Skills Test (QUEST), were reported in two studies from the same centre.^{95,117} Significant improvements in total QUEST scores were reported for upper-limb function. The QUEST instrument is a reliable and validated outcome measure designed to evaluate upper-limb movement and function in children with spasticity-related neuromotor dysfunction.^{119,120} Validation studies, however, have shown that the four individual subdomains—dissociated movement, grasp, weight bearing, and protective extension—represent different traits. Experts recommend that individual scores rather than total scores be reported.

Table 5. Short-Term Effectiveness of Dorsal Rhizotomy for WeeFIM-Evaluated Functional Independence

WeeFIM Variant, Outcome Measure	WeeFIM Score, Mean ± SD (Range)						
	Pre-op	6-Month Post-op	Pre-op to 6-Month Post-op Mean Change [95% CI]	12-Month Post-op	Pre-op to 12-Month Post-op Mean Change [95% CI]	24-Month Post-op	Pre-op to 24-Month Post-op Mean Change [95% CI]
Canada: Vancouver, Loewen et al,¹¹⁷ 1998: 12-month follow-up—diplegia (n = 23), quadriplegia (n = 13)							
36 patients (17 M, 19 F), mean age 4.1 years (range 2.9–14.6 years)							
WeeFIM Total, median ± SD	66.0 ± 22.8	--	--	80.5 ± 24.7	11.0 ± 11.3 P = .0001	--	--
WeeFIM motor, median ± SD	33.5 ± 18.7	--	--	47.5 ± 21.7	9.5 ± 10.6 P = .0001	--	--
WeeFIM cognitive, median ± SD	31.5 ± 7.1	--	--	34.0 ± 5.8	1.0 ± 3.0 P = .008	--	--
QUEST, median ± SD	85.5 ± 15.4	--	--	89.8 ± 14.4	3.2 ± 4.7 P = .0001	--	--
Canada: Vancouver, Steinbok et al,⁹⁵ 2009: 12-month follow-up—NR							
22 patients (NR) mean age 5.2 ± 4.2 years							
WeeFIM Total	58.44	--	--	--	9.5 ± 11.8	--	--
QUEST	75.66	--	--	--	3.8 ± 4.8	--	--
Czech Republic: Prague, Tichy et al,¹¹⁸ 2003: post-operative diplegia (n = 2, 2 ambulatory), quadriplegia (n = 12, 12 nonambulatory)							
14 patients (10 M, 4 F) mean age 16.5 years (range 8–27 years)							
WeeFIM motor	19.5	25.5	31%	--	--	--	--
WeeFIM cognitive	7.7	9.5	26%	--	--	--	--
Italy: Torino, Carraro et al,⁹⁶ 2014: 12-month follow-up—diplegia (n = 9, all ambulatory) GMFCS Level II (n = 7) GMFCS Level III (n = 2)							
Diplegia 9 patients (6 M, 3 F) mean age ± SD 7.9 ± 3.2 years							
WeeFIM Total	107 (76–126)	--	--	114 (86–126)	P = .031	--	--
WeeFIM motor	57 (25–91)	--	--	63 (29–91)	P = .031	--	--
United States: Chicago, Nishida et al,¹¹⁶ 1995: 24-month follow-up—diplegia (n = 60, 52 ambulatory, 8 nonambulatory), quadriplegia (n = 36, 28 nonambulatory, 8 assisted ambulatory)							
96 patients (56 M, 40 F) mean age 5.4 years (range 1.1–12.8 years)							
Diplegia							
WeeFIM self-care	19.1	--	--	--	--	27.7	45%
WeeFIM mobility	15.7	--	--	--	--	29.6	88.5%

WeeFIM sphincter control (bowel and bladder)	13.2	--	--	--	--	18.3	39%
Quadriplegia							
WeeFIM self-care	11.5	--	--	--	--	16.9	47%
WeeFIM mobility	6.9	--	--	--	--	9.7	40.5%
WeeFIM sphincter control (bowel and bladder)	7.3	--	--	--	--	14.4	97%

Abbreviations: CI, confidence interval; F, female; GMFCS, Gross Motor Function Classification System; M, male; n, number; NR, not reported; post-op, post-operative; pre-op, pre-operative; QUEST, Quality of Upper Extremity Skills Test; SD, standard deviation; WeeFim, Functional Independence Measure for Children.

CONCLUSIONS: Short-Term Effectiveness for Functional Independence and Caregiver Assistance

- Statistically significant improvements in functional independence, self-care, and mobility were reported for children with spastic diplegia with various levels of gross motor disability.
- Social function was evaluated less often, but when it was assessed, significant improvements were reported.
- Children with spastic diplegia causing various levels of gross motor disability required less caregiver assistance.
- Children with spastic diplegia had additional gains in both functional independence and reduced caregiver assistance with longer-term follow-up.
- For children with spastic quadriplegia, functional independence was generally not evaluated. Where it was assessed, results were inconsistent. Caregiver assistance for these children was not evaluated in any study.
- The Canadian Occupational Performance Measure, a scale that measures the effectiveness of a treatment for individually declared treatment goals, demonstrated treatment effectiveness and satisfaction with goal attainment after dorsal rhizotomy for diverse declared treatment goals for children with spastic diplegia.
- Overall evidence: 10 studies, GRADE moderate quality (Table A5)

Treatment Satisfaction with Dorsal Rhizotomy at Short Term

Eight groups reported on treatment satisfaction of caregivers or parents of children with spastic cerebral palsy treated with dorsal rhizotomy by multi-disciplinary teams in five different countries: Canada,^{121,122} South Africa,^{123,124} the United States,^{125,126} the United Kingdom,¹²⁷ and Sweden.¹²⁸

Canadian Families

In two Canadian studies,^{121,122} Kinghorn et al¹²² evaluated upper-extremity function after dorsal rhizotomy in a small group (5 with quadriplegia and 2 with diplegia) of children with spastic cerebral palsy and Kim et al¹²¹ evaluated multi-disciplinary outcomes in a large group (102 with spastic diplegia and 72 with spastic quadriplegia) of children with cerebral palsy. In the Kinghorn et al¹²² study, all parents expressed satisfaction with their children's surgical outcomes. Parents' subjective reports included improvements in play skills, endurance, sitting and standing balance, and activities of daily living.

In the Kim et al¹²¹ study, treatment outcomes based on caregivers' opinions were divided into acceptable and unacceptable. Unacceptable or unsatisfactory outcomes were those judged by caregivers to show no improvement in lower-limb function, overall motor function, or ADLs or to show deterioration in overall motor function after 1 year. Other outcomes were then considered acceptable or satisfactory. Overall 94% (163/174) of treatments were judged to have acceptable outcomes and 6% (11/174) were poor or unacceptable. The rate of treatment outcomes judged to be acceptable was higher for children with spastic diplegia (99%; 101/102) than for those with spastic quadriplegia (86%; 62/72).

South African Families

Two studies^{123,124} in South Africa evaluated parents' or caregivers' reports on treatment outcomes in children with spastic cerebral palsy treated by dorsal rhizotomy. Peacock et al,¹²³ in one of the first clinical reports on dorsal rhizotomy, compared outcomes and parental reports for severely handicapped (7 children) and mildly or moderately handicapped (8 children) children. In the severely handicapped group, all parents reported children to be less irritable or easier to manage. For one child, improvements after dorsal rhizotomy in care, feeding, and toileting and in the child's well-being were so dramatic that the parents decided against institutionalizing the child. All parents, except one, reported general improvements in their children involving sitting, walking, and toileting. They also reported that their children were happier and more confident.

In the Peter et al¹²⁴ study, teenagers and young adults (26 with spastic diplegia and 4 with spastic quadriplegia) treated with dorsal rhizotomy had received physical therapy at specialized cerebral palsy schools. Most (77%, 23/30) reported that their motor function had improved and that they would recommend the surgery to others. Four respondents were ambivalent about their treatment outcomes, and three reported that treatment had not improved their function.

American Families

Two studies of practices in American centres^{125,126} involved different study populations. Montgomery et al¹²⁶ studied 14 children treated at five institutions who had spastic diplegia (5 children), spastic quadriplegia (8 children), or spastic hemiplegia (1 child). Of the 15 parents approached, 1 parent refused to participate because of lack of progress for her child, which she attributed to an inability to access the recommended intensity of physical therapy. Of the 14 parents interviewed, 9 reported that dorsal rhizotomy and post-operative rehabilitation was

highly stressful because of the hospitalization itself, time for therapies, and inadequate time to care for their other children. The majority (71%, 10/14) of parents reported that surgery had been beneficial and they would choose it again. The parents of four children, however, reported that, given the outcomes their children experienced, they would not have chosen the surgery.

Kaufman et al¹²⁵ studied treatment outcomes in 19 children with spastic diplegia. Patients and their families were asked to rate their satisfaction with surgical results. Their response options were graded with 4-level response options (unsatisfied, neutral, satisfied, very satisfied). One family was lost to follow-up; of the 19 families interviewed, 18 reported being very satisfied and 1 being satisfied.

United Kingdom Families

A recent and unique study in the United Kingdom by Ingale et al¹²⁷ reported on parent's satisfaction with outcomes for their severely disabled children (mean age 12 years) treated with dorsal rhizotomy after being managed with intrathecal baclofen. At their centre, more than 300 children with cerebral palsy classified as GMFCS Levels IV or V were treated with intrathecal baclofen. Ten of 130 caregivers or parents provided informed consent for dorsal rhizotomy as an alternative when intrathecal pumps needed replacement.

In the study, caregivers' and parents' opinions were solicited in structured interviews on their children's outcomes after dorsal rhizotomy. This feedback was compared to that after intrathecal baclofen therapy. Parents reported that spasticity improvement was greater (9 parents), nursing and day-to-day care was easier (7 parents), and children were less distressed or irritable (3 parents) with dorsal rhizotomy than with intrathecal baclofen. Only a few parents, however, reported functional gains for their children—four children (GMFCS Level IV) were able to crawl and stand with assistance; two children (GMFCS Level V) were able to sit with adaptive equipment. Results of this study led to a change in practice at the institution, where physicians now routinely offer dorsal rhizotomy to all GMFCS groups.

Swedish Families

The most detailed report on caregivers' and parents' observation of outcomes and opinions after dorsal rhizotomy was by Eliasson et al¹²⁸ on a small group (7 children) of Swedish children aged 4 to 8 years with spastic diplegia. Qualitative analysis of the interview data identified six themes relevant to parents—increased motion, normal-looking appearance, changed performance of activities, and changed independence of self-care. The importance of training and the negative consequences were also mentioned.

In that study, five parents reported changes in motion, smoothness, and control as important changes that were strongly satisfactory. Parents were emotional when they described how their child appeared more “normal” when walking or at rest or when children were carried or embraced. Parents also reported that their children had not learned new activities but performed tasks easier, faster, and for longer periods and were more adaptable in positioning. Only one parent reported no changes in self-care. The others all reported improvements in dressing, but few commented on feeding or toileting. Parents also commented on how the environment and the child's maturation affected these outcomes. Improved muscle control and range of motion after surgery enabled children to perform tasks better, and because they were getting older they became motivated by other children.

All families mentioned training after surgery, but their comments weren't always about physical therapy. Recreational activity and physical training had been a part of daily living before surgery, and several families continued to assume responsibility for physical activities after surgery. Families mentioned difficulties in their child's physical rehabilitation program caused by changes in physical therapists after dorsal rhizotomy, but all were uncertain about the effects of the frequency of physical therapy after surgery.

Negative effects, involving different events and to different degrees, were reported by all parents. Children who were ambulatory before dorsal rhizotomy all developed sudden fatigue, and many did not regain strength for lengthy periods after the surgery. For one parent, the year it took the child to get to the level before surgery was not worth it.

CONCLUSIONS: Treatment Satisfaction at Short Term

- Treatment satisfaction with dorsal rhizotomy was not routinely investigated in clinical studies, but reports were available from caregivers and parents of children with spastic cerebral palsy undergoing surgery by multi-disciplinary teams in different countries, including Canada.
- Generally parents reported improvement in their children after dorsal rhizotomy and expressed satisfaction with treatment of their children. Treatment satisfaction, however, was variously defined and measured in clinical studies.
- Treatment outcomes with which parents expressed satisfaction were rarely evaluated formally as outcomes in clinical studies—mood changes (happier, less distressed, less irritable), more normal-looking appearance, greater confidence, improved task performance (easier, faster, more adaptable).
- One study evaluated satisfaction among adolescents and young adults who had undergone dorsal rhizotomy. Most reported their motor function had improved and they would recommend the surgery to others.
- Parents in some studies reported that the surgery and post-operative rehabilitation had been highly stressful, been time-consuming, and interfered with care for their other children.
- Some parents also reported fatigue and loss of strength in their children after surgery and difficulties with changing therapists in the rehabilitation program.

Long-Term Effectiveness of Lumbosacral Dorsal Rhizotomy

The primary clinical studies and their characteristics evaluating long-term (> 2 years) effectiveness after dorsal rhizotomy are listed in Appendix 2, Table A3. All studies were long-term observational pre-post cohort studies. None of the earlier randomized controlled trials⁷³⁻⁷⁵ had included long-term follow-up. Studies in the Appendix reporting on long-term results using validated outcome measures for effectiveness are discussed under long-term effectiveness for gross motor function and under the results for long-term functional independence and Caregiver assistance. Those studies reporting on satisfaction in the long term with lumbosacral dorsal rhizotomy are discussed under treatment satisfaction at long-term.

Long-Term Effectiveness for Gross Motor Function

Ten studies evaluated gross motor function in children with spastic cerebral palsy undergoing dorsal rhizotomy. Of these, eight¹²⁹⁻¹³⁶ used the validated Gross Motor Function Measure (GMFM) and two employed outcome measures similar to the GMFM: the Berman scale¹³⁷ and the Vancouver Motor Function Measure (VMFM), a locally developed and unvalidated measure.¹³⁸

This review evaluates long-term gross motor function as evaluated by the GMFM in nine studies from three countries: Canada,^{130,133,138,139} Netherlands,^{129,136} and Sweden.^{132,134,135} The South African study by Langerak et al¹⁴⁰ employed the Berman scale, a gross motor outcome measure similar to the GMFM, which rated functional movement (sitting, kneeling, standing, rolling, crawling, walking) on a scale from 1 (normal) to 5 (most severe).

Canadian Cohorts

Canadian studies involved reports from two institutions, the Vancouver Children's Hospital^{138,139} and the Montreal Children's Hospital.^{130,133}

Vancouver Cohort, 5-Year and 10-Year Follow-Up

The Vancouver cohort was first reported by Gul et al¹³⁸ and involved a mean 5-year follow-up. Ailon et al¹³⁹ subsequently reported a mean 14.4-year follow-up for 44 children with average age 4.5 years (range, 2.9–7.7 years). The cohort included both ambulatory (GMFCS Levels I–III) and nonambulatory (GMFCS Levels IV–V) children with spastic cerebral palsy. The study cohort was assembled from 142 children having dorsal rhizotomy 10 years prior. Of these, 44 children had received formal assessments at 5 years and after more than 10 years. Gross motor function was assessed by two instruments: the VMFM for some children, and the GMFM for other children. The VMFM scores were subsequently converted to GMFM-66 values.

Mean scores in GMFM-66 scores significantly increased over baseline at 5-year follow-up in both ambulating (64.4 to 74.5; $P < .0001$) and nonambulating (36.7 to 45.0; $P = .01$) children. In both groups, mean scores were lower at the 10-year than the 5-year follow-up. Mean scores among ambulatory patients (71.0; $P = 0.042$), however, were still significantly higher than at baseline whereas scores among nonambulatory patients (31.8; $P = 0.3$) declined much more sharply and were no longer statistically significant over baseline.

Change in motor function appeared to parallel the differences in quadriceps strength. Mean muscle strength evaluated by the Medical Research Council scale increased over baseline in both groups, more for ambulatory patients (0.6 [95% confidence interval 0.3–0.9] $P = .001$) than for nonambulatory patients (0.1 [95% confidence interval –0.1 to 0.4] $P = .3$) at the early follow-

up. Muscle strength remained high at long-term follow-up in ambulatory groups but decreased sharply in nonambulatory groups. Muscle spasticity in both groups, however, remained significantly lower at both follow-up points than baseline.

Montreal Cohort, 5-Year and 15-Year Follow-Up

The Montreal cohort first reported by Mittel et al¹³³ had a 5-year follow-up and subsequently a 15-year follow-up reported by Dudley et al.¹³⁰ At the 5-year follow-up, Mittel et al reported on gross motor function (GMFM-88) and other developmentally relevant transitional movements (alignment, transition, and stability) graded on five levels of functional performance based on the Rusk Institute of Rehabilitation/NYU Rhizotomy form (Rusk/NYU). Ninety-three children met the study inclusion criteria and 71 completed the 3-year follow-up. The study cohort therefore consisted of 71 children (43 male and 28 female) of average age 5.2 years (range 3.0–10.7 years). Fifty of these children completed the 5-year follow-up; 21 had not reached that follow-up point.

Hip adductor spasticity was significantly reduced at 1-year follow-up and remained low at the 3-year and 5-year follow-up. The primary outcome measure of the Rusk/NYU form, the total alignment score, was significantly ($P < .001$) improved at 1-year, 3-year, and 5-year follow-up (line graphs only). Total GMFM-88 scores were significantly improved over baseline at all follow-up points, which included 1-year, 3-year, and 5-year follow-up. The greatest gains occurred in Item D (standing) and Item E (walking/running/jumping)—gains that continued to increase over the periods of 1- to 3-year follow-up and 3- to 5-year follow-up. In all subgroups the gains in GMFMs were equal to or greater than 6%, a clinically relevant difference.⁸¹

Dudley et al¹³⁰ reported GMFM-88 scores for long-term gross motor function at 1, 5, 10, and 15 years. Of the original cohort of 105 children with spastic cerebral palsy who were on average 5 years old (range 3.0–10.5 years), 102 had pre-operative assessments; 97, 62, 57, and 14 patients completed the 1-, 5-, 10-, and 15-year post-operative follow-up, respectively. Estimates of gross motor gains at the 15-year follow-up were limited by how few children reached this follow-up point.

Spasticity scores (hip adductors, ankle-plantar flexors, and hamstring) all remained significantly lower than baseline at all follow-up points. For all children, the total mean GMFM-88 scores and each of the five motor subdomains (Items A to E) significantly increased ($P < .05$) over baseline at each follow-up point. Between the 5-year and 10-year follow-up, however, all GMFM total scores and subscores appeared to level off, although they remained significantly improved over baseline. When mean total GMFM-88 scores were stratified by level of pre-operative motor disability over time, only the motor gains in the ambulatory group (GMFCS Levels I–III) remained significantly higher than baseline.

Netherlands Cohort, 6-Year and 10-Year Follow-Up

The first report on the Amsterdam cohort by van Schie et al¹³⁶ involved their 6-year follow-up; their subsequent report by Bolster et al¹²⁹ outlined their 10-year follow-up. The study by Bolster et al¹²⁹ included 36 ambulatory (GMFCS Levels I–III) children of median age 6 years (range 2 years, 10 months to 12 years, 1 month) with spastic diplegia who had undergone dorsal rhizotomy 5 years prior. Baseline motor assessments (GMFM-66) were obtained for 29 children, and 28 of these completed the 5-year follow-up and 20 completed the 10-year follow-up. Eight children had not reached the post-operative 10-year follow-up point.

The mean GMFM-66 scores at the 5-year (65.4 ± 13.3 ; $P < .001$) and 10-year follow-up (64.2 ± 14.9 ; $P < .001$) were significantly higher than baseline (57.8 ± 11.0). Mean gross motor scores after 5 years had levelled off; scores at 10 years were not significantly different from scores at 5 years.

The GMFM-66 scores were also transformed into percentiles according to the GMFCS and age of the child. The transformation of motor scores was based on an evaluation of GMFM-66 scores from a population-based cross-sectional longitudinal study of Ontario children with cerebral palsy (the Ontario Motor Growth Study).¹⁴¹ Percentiles based on age and GMFCS were used to estimate change outside normal developmental expectations. An increase of more than 20 percentiles between time intervals was judged to represent an increase in motor function beyond normal developmental expectations and a decrease of 20 percentiles to represent a loss in motor function. At the 5-year follow-up of the Amsterdam cohort, 36% (10/28) of children gained more than 20 percentiles. Percentiles for the other 18 children were within the 20-percentile range, essentially unchanged. At the 10-year follow-up, 6 of the 20 children had more than a 20-percentile increase; percentiles for 14 children were stable.

South African Cohort, 20-Year Follow-Up

The study by Langerak et al¹⁴⁰ involved a 20-year follow-up of 14 young adults (mean age 27 years, range 22–33 years) with spastic diplegia who were ambulatory before dorsal rhizotomy. The original cohort treated 20 years prior at the Red Cross Children's Hospital in Cape Town consisted of 29 children with spastic diplegia, 18 of whom were ambulatory and 11 of whom were nonambulatory.¹⁴²

Improvement in functional movement at 1 year for all nine items evaluated on the Berman scale (similar to the GMFM scale) was dependent on prior disability levels of the children. Improvements were reported mainly for ambulatory children. For the 14 ambulatory children evaluated at the 20-year follow-up, pre-operative median scores for functional movement were statistically significantly improved for eight of nine measures at 1-year follow-up. At the 20-year follow-up, the overall functional movement scores remained significantly ($P < .001$) improved over baseline and each of the seven sub-measures improved: long sitting ($P = .001$), side sitting ($P = .074$), prone kneeling ($P = .005$), kneel-standing ($P = .002$), half kneeling ($P = .003$), standing ($P = .028$), rolling ($P = .004$), crawling ($P = .008$), and walking ($P = .050$). Median motor scores did not change significantly between the 1-year and 20-year follow-up points.

Swedish Cohorts

Stockholm Cohort, 10-Year Follow-Up

The Stockholm cohort by Tedroff et al¹³⁵ included 19 children with spastic diplegia of average age 4 years, 7 months; range 2 to 9 years, who underwent dorsal rhizotomy at the Karolinska Hospital. Follow-up included complete assessments of 15 children at baseline and at 18 months, 3 years, and 10 years.

Median GMFM-88 scores were significantly improved over baseline (51; interquartile range [IQR] 31–75) at the 18-month (66; IQR 40–85, $P = .002$) and at the 3-year follow-up (76; IQR 51–91, $P < .001$). Between the 3-year and 10-year follow-up, motor function declined to a median of 62 (IQR 38–93). The GMFM-88 score at 10 years, despite the 11% reduction since the 3-year follow-up, was still significantly ($P < .001$) improved over baseline. Also at 10-year

follow-up, the Wilson gait score (a 9-level ordinal score), showed 63% (12/19) of children improved their mobility over baseline, 26% (5/19) worsened, and 10% (2/19) were unchanged.

Lund Cohort, 5-Year and 10-Year Follow-Up

The Lund cohort was first reported by Nordmark et al¹³⁴ for the 5-year follow-up and later by Josenby et al¹³² for the 10-year follow-up. The cohort initially included 35 children of average age 4.5 years, range 2.5 to 6.6 years, with spastic diplegia treated with dorsal rhizotomy at the University Hospital in Lund. The cohort included ambulating (GMFCS Levels I–III, 19 children) and nonambulating children (GMFCS Levels IV–V, 16 children). All children attended the follow-up interviews, but not all assessments were performed at 6 months, 12 months, 18 months, 3 years, and 5 years.

In the 5-year follow-up, gross motor function scores from the GMFM-88 and GMFM-66 were reported as mean change in scores over various follow-up periods. Individual motor function goals (GMFM-88 Goal Totals) established by the family and local therapists varied by children's motor disability and ambulatory status. For independent ambulators (GMFCS Levels I–II), goals were to improve balance, endurance, and flexibility in standing, walking, running, and jumping. For dependent ambulators (GMFCS Level III), goals were to improve stability and variability in sitting, to attain and maintain the ability to stand and walk, and to enable self-propelled wheeled transfers. For nonambulators (GMFCS Levels IV–V), goals were to attain independent sitting, to attain supported standing, and to enable wheelchair transfers.

All GMFMs (GMFM-66, GMFM-88, GMFM-88 Goal Totals) improved over baseline ($P < .001$) at 12-month, 3-year, and 5-year follow-up (Table 6). There were steady gains in motor scores throughout the follow-up period; statistically significant and clinically important gains over baseline were noted up to the 3-year follow-up. The gains in gross motor scores were also higher for the individual GMFM-88 Goal Total score than for the global total scores for both the GMFM-88 and the GMFM-66 at all follow-up periods. The ranges in motor gains for all outcome measures, however, reflected extensive variability in individual responses at all follow-up points.

The gains in gross motor function measures (GMFM-66) were also reported by the level of motor disability at baseline for interval changes over a 5-year follow-up period (Table 7). The overall gain in gross motor function scores from baseline to 5 years was inversely proportional to baseline disability—higher motor change scores were achieved by those with the least motor disability. A gross motor GMFC-66 overall change score (pre-operative to 5-year) for disability groups was 16.7 for independent ambulators (GMFCS Levels I–II) and 10 for assisted ambulators (GMFCS Level III). Both of these values represent clinically relevant change. A change score of 5.1 was achieved with nonambulators (GMFCS Levels IV–V), the most disabled group.

Table 6. Long-Term Changes in Gross Motor Function After Dorsal Rhizotomy for Children With Spastic Diplegia

Test Variant	Pre-Op to 12-Month Change			Pre-Op to 3-Year Change			Pre-op to 5-Year Change		
	Mean ± SD	Range	P Value	Mean ± SD	Range	P Value	Mean ± SD	Range	P Value
GMFM-66	3.5 ± 4.8	-3.8 to 19.5	< .001	8.0 ± 8.1	-9.9 to 29.4	< .001	9.5 ± 9.7	-7.4 to 38.4	< .001
GMFM-88	10.9 ± 10.3	-9.0 to 31.0	< .001	16.1 ± 12.9	-4.0 to 52.0	< .001	21.2 ± 17.9	-6.9 to 17.9	< .001
GMFM-88 Goal Total	13.6 ± 13.1	-10.0 to 42.2	< .001	23.6 ± 17.4	-17.0 to 59	< .001	25.2 ± 18.7	-21.3 to 59.0	< .001

Abbreviations: GMFM, Gross Motor Function Measure; pre-op, pre-operative assessment; SD, standard deviation.
Data from Nordmark et al.¹³⁴ 2008

Table 7. Long-Term GMFM-66 Interval Changes by Baseline Motor Disability After Dorsal Rhizotomy

GMFCS Level	Pre-op		Pre-op to 6-Month Interval Change		6-Month to 12-Month Interval Change		12-Month to 18-Month Interval Change		18-Month to 3-Year Interval Change		3-Year to 5-Year Interval Change	
	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range	Mean ± SD	Range
I–III	58.6 ± 8.9	47.3–71.2	2.8 ± 4.9	-5.5 to 10	5.5 ± 4.4	0 to 13.7	-1.2 ± 5.1	-13.0 to 3.2	7.5 ± 4.7	1.0 to 17.7	2.1 ± 4.4	-4.1 to 9.1
III	49.9 ± 3.0	43.3–3.1	1.0 ± 4.9	-6.5 to 9.8	1.8 ± 3.1	-5.0 to 6.6	1.3 ± 4.1	-3.0 to 8.5	2.2 ± 4.1	-2.2 to 8.7	3.7 ± 3.5	-2.5 to 7.4
IV–V	40.1 ± 5.8	31.2–51.1	1.4 ± 3.1	-4.5 to 6.3	1.0 ± 2.0	-2.8 to 4.8	1.2 ± 2.0	-3.4 to 4.0	1.2 ± 3.7	-2.6 to 11.9	0.3 ± 2.5	-3.0 to 6.5
All	47.6 ± 9.8	31.2–71.2	1.6 ± 4.0	-6.5 to 10.9	2.4 ± 3.5	-5.0 to 13.7	0.6 ± 3.5	-13.0 to 8.5	3.1 ± 4.8	-2.6 to 17.7	1.8 ± 3.6	-4.1 to 9.1

Abbreviations: GMFCS, Gross Motor Function Classification System; pre-op, pre-operative; SD, standard deviation.
Data from Nordmark et al.¹³⁴ 2008

In a further analysis of the 5-year gains in the Lund Cohort for individual GMFM-88 Goal Total scores, gains over baseline were reported for three groups with diplegia stratified by motor disability: independent ambulators, dependent ambulators, and nonambulators. In the ambulating group (GMFCS Levels I–II), seven of the nine children gradually increased their motor goal scores over follow-up by more than 10 points. In the assisted ambulation group (GMFCS Level III), nine of the ten children gradually increased their motor scores by more than 10 points. Overall, 84% (16/19) of the ambulating group significantly improved their individual motor goal scores.

In the nonambulating (GMFCS Level IV–V) group, 75% (12/16) also increased their individual motor goal total scores. Four children (ages 4.0, 4.8, 5.8, and 3.3 years) had minimal or no change at 5-year follow-up. Overall the highest goal total scores at final-year follow-up were achieved by independent ambulatory patients. Those achieving more than 70% of their goal total scores at the 5-year follow-up included 89% (8/9) of independent ambulatory patients, 30% (3/10) of assisted ambulatory patients, and 38% (6/16) of nonambulatory patients.

The 10-year follow-up of the Lund cohort by Josenby et al¹³² included 29 children of the original cohort of 35 children with spastic diplegia. Five children chose not to participate (three lived outside the region), and one was unable to participate because of poor health. At the 10-year follow-up, children (median age 15 years) were adolescents. At 10-year follow-up, the spasticity of major muscle groups, assessed by the Modified Ashworth Scale, remained normal for most children: hip flexors (70%, 19/27), abductors (83%, 24/29), knee flexors (46%, 13/28), and plantar flexors (79%, 23/29). The mean change of 10.6 ± 11.8 of GMFM-66 scores over baseline at 10-year follow-up (48.6 ± 9.7 to 59.2 ± 17.2) for the entire study group represents a clinically relevant change in gross motor function.

Development of gross motor function in children treated with dorsal rhizotomy in the Lund cohort was also compared with developmental motor patterns from the Ontario Motor Growth Study of 657 untreated children with spastic cerebral palsy.⁸⁷ Similar regression techniques for gross motor developmental curves employed in the Ontario population cohort study¹⁴¹ were used to construct developmental curves for the Lund cohort separately for the GMFCS classification (Levels II–IV) of motor disability. The GMFCS Levels I and V were not developed, as there were too few children in these categories for meaningful analysis.

Trends for GMFM-66 scores among children treated with dorsal rhizotomy in the Lund cohort were compared with scores among the control group of children with untreated cerebral palsy in the Ontario cohort. Key parameters from the analysis, age at which the maximum GMFM-66 score is achieved and the age-90 (age at which 90% of the maximum score is obtained) are detailed in Table 8. Children in the Lund cohort with GMFCS Levels II and IV achieved higher GMFM-66 scores than Ontario children with cerebral palsy not undergoing dorsal rhizotomy. Children at GMFCS Level III do not appear to achieve higher GMFM-66 scores, although they achieve their maximum scores at an earlier age.

Table 8. Maximum GMFM-66 Score and Age at Attainment of Maximum Scores for Lund Dorsal Rhizotomy Cohort and Ontario Control Cerebral Palsy Population

GMFCS	Population	Maximum GMFM-66 Score (95% CI)	Age-90
Level II	Lund	79.4	7 years, 8 months
	OMG	68.5 (66.0–71.0)	4 years, 11 months
Level III	Lund	54.7	2 years, 2 months
	OMG	53.5 (51.9–55.0)	3 years, 2 months
Level IV	Lund	45.4	3 years, 2 months
	OMG	39.5 (38.2–40.9)	3 years, 2 months

Abbreviations: Age-90, age at which 90% of maximum score is obtained; CI, confidence interval; GMFCS, Gross Motor Function Classification System; GMFM-66, Gross Motor Function test; OMG, Ontario Motor Growth Study.

Data from Josenby et al.¹³² 2012

CONCLUSIONS: Long-Term Effectiveness for Gross Motor Function

- Long-term cohort studies from four countries, two from Canada, showed significant improvements in gross motor function for children with spastic diplegia at 5-year follow-up.
- Interval analysis showed that gains in gross motor function were continuous throughout longer-term follow-up, suggesting that shorter-term cohort follow-up studies could underestimate motor gains.
- In general, gains in gross motor function level off 5 years after dorsal rhizotomy.
- There is an indirect relationship between long-term gross motor gains and level of pre-operative motor disability; greater total motor gains were achieved by less disabled children, particularly by independently ambulatory children.
- Gains in motor function were noted for nonambulatory children with spastic cerebral palsy when motor gains were evaluated as specific or “goal directed” subsets of the Gross Motor Function Measure.
- Compared with a control population of untreated children with spastic cerebral palsy, children with spastic diplegia treated with dorsal rhizotomy achieved greater gains in gross motor function at all levels.
- Overall evidence: 10 studies, GRADE moderate quality (Table A5)

Long-Term Functional Independence and Caregiver Assistance

Four studies^{130,134,143,144} evaluated functional independence, activities of daily living, or caregiver assistance using validated outcome measures. One study¹³⁷ evaluated children who were ambulatory and had spastic diplegia for long-term levels of activity and participation after dorsal rhizotomy.

Two centres, one in Montreal, Canada, and one in Lund, Sweden, reported on the long-term effects of dorsal rhizotomy on functional independence for children with spastic diplegia using validated outcome measures. All centres assessed functional independence with the Pediatric Evaluation of Disability Inventory (PEDI), an inventory based on three domains: functional independence, caregiver assistance, and social function. One centre^{130,144} reported on functional independence, and one centre^{134,143} reported on both functional independence and caregiver assistance. Neither centre reported on social function, which is often viewed as a more complex domain. Levels of activity and participation were evaluated by the Cape Town centre in South Africa by a different outcome assessment tool—the Life Habit questionnaire.¹³⁷

Montreal Cohort, 5-Year and 15-Year Follow-Up

Two studies reported how dorsal rhizotomy affected long-term function in the Montreal cohort: the 5-year follow-up by Mittal et al¹⁴⁴ and the 15-year follow-up by Dudley et al.¹³⁰ The original cohort consisted of 57 children completing the 6-month and 1-year follow-up. Of this cohort, 41 completed the 3-year and 30 completed the 5-year assessments. The children mainly had spastic diplegia (36 children) with mild motor disability classed at GMFCS Levels I to II (34 children) and at surgery were on average aged 4.8 years, range 3.0 to 7.5 years old.

Functional assessment, including self-care and mobility at follow-up, were evaluated with PEDI, but scores indicating need for caregivers' assistance with self-care and mobility were not reported. Functional scores for self-care significantly improved over baseline at the 1-year (59.0 to 67.9; $P < .001$), 3-year (59.0 to 81.6; $P < .001$), and 5-year (59.0 to 82.4; $P < .001$) follow-up. Self-care scores continued to improve after 1-year follow-up. Self-care scores improved ($P = .003$) between the 1-year and 3-year follow-up but not significantly ($P = .223$) between the 3-year and 5-year follow-up when scores plateaued. A similar pattern was observed for functional mobility scores. Scores improved over baseline (56) at the 1-year (64; $P < .001$), 3-year (77.2; $P < .001$), and 5-year (77.8; $P < .001$) follow-up. Mobility scores improved between the 1-year and 3-year assessment ($P = .002$) but not between the 3-year and 5-year ($P = .169$) follow-up—plateauing as the self-care scores did.

Dudley et al¹³⁰ reported on the longer-term follow-up (up to 15 years) until adolescence and early adulthood of a larger group of children from this centre. One hundred five children with a mean age of 5 years (range 3–10.5 years), of whom many had spastic diplegia (65 children) were treated by dorsal rhizotomy at the Montreal Children's Hospital. Afterward, children were assessed at 1-year (97 children), 5-year (62 children), 10-year (57 children), and 15-year (14 children) follow-up.

Functional scores for self-care and mobility over the follow-up period are outlined in Figure A4. Caregiver assistance for self-care and mobility subdomains was not reported. Mean PEDI scores for both self-care and mobility improved over baseline ($P < .05$) at each follow-up point. The gains in mean PEDI functional mobility independence scores appeared to level off after the 5-year follow-up point as they had with the GMFM gross motor scores. In comparison, mean

PEDI scores for self-care and functional independence appeared to continue to increase after 5 years.

Long-term functional independence in self-care and mobility was also examined across levels of pre-operative gross motor disability. However, pre-operative GMFCS classification was available for only 52 children, and no GMFCS Level V children were treated. For the PEDI mobility domain, only GMFCS Level III appeared to continue to increase after 5 years—the other levels (I, II, and IV) appeared to plateau. Mean PEDI self-care scores continued to improve after 5 years for all GMFCS levels except for Level IV, which appeared to level off.

Lund Cohort, 5-Year and 10-Year Follow-up

Two studies reported how dorsal rhizotomy affects long-term functional outcomes in the Lund cohort: the 5-year follow-up by Nordmark et al.¹³⁴ and the 10-year follow-up by Josenby et al.¹⁴³ The original cohort consisted of 35 children with spastic diplegia, mean age 4.5 years, range 2.5 to 6.6 years. All children attended the follow-up visits at 6 months, 12 months, 18 months, 3 years, and 5 years. Functional independence evaluated by PEDI and involving dimensions of both self-care and mobility as well as caregiver assistance were reported in both studies. However, baseline PEDI results were unavailable for five children who were treated before 1995, when the Swedish translation of the PEDI became available. The 5-year follow-up included 30 children. The 10-year follow-up included 24 children; four of the six children who did not participate lived outside the region.

Median PEDI scores for functional independence at 10-year follow-up, stratified by pre-operative ambulators (GMFCS Levels I–III) versus nonambulators (GMFCS Levels IV–V) for self-care and mobility and for caregiver assistance are listed in Table 9. The ambulatory group involved independent (8 children) and dependent (4 children) ambulators. The nonambulatory group consisted mainly of GMFCS Level IV (11 children); only one child was GMFCS Level V. In both the ambulatory and nonambulatory groups, children significantly improved their self-care and their mobility at 5-year follow-up (Table 9). However, further gains in median scores for mobility and self-care were no longer statistically significant in either group between from the 5-year to 10-year follow-up period.

Children in the ambulatory group in particular had already achieved near-complete or complete independence (score of 100 was complete independence) in these domains at the 5-year follow-up. Children in the nonambulatory group had plateaued at much lower independence scores for both mobility and self-care.

The trend in PEDI median scores for caregiver assistance was similar to that for functional independence (Table 9). Caregiver assistance for children in the ambulatory group continued to decrease (increasing scores) throughout the 10-year follow-up period. Some of these children already had no need (score of 100) for caregiver assistance for either self-care or mobility at the 5-year and 10-year follow-up, mainly those with less pre-operative motor disability (GMFCS Levels I–III).

Table 9. Long-Term PEDI Scores for Functional Independence in Children with Spastic Diplegia After Dorsal Rhizotomy

		PEDI Self-Care				PEDI Mobility Score			
GMFCS Level	Yr After Surgery	Median	25th Percentile	75th Percentile	Interval P Value 0 to 5 yr 5 to 10 yr	Median	25th Percentile	75th Percentile	Interval P Value 0 to 5 yr 5 to 10 yr
Child functional level									
Levels I–III (n = 12)	0	61.8	53.7	69.1	--	58.2	51.4	68.7	--
	5	85.1	70.0	100	.001	82.5	67.4	94.2	< .001
	10	96.5	79.4	100	.125	87.2	78.6	94.2	.039
Levels IV–V (n = 12)	0	46.0	40.4	51.7	--	37.1	32.0	41.4	--
	5	57.4	53.7	62.5	< .001	49.7	45.2	61.9	.007
	10	56.8	54.0	63.2	.774	56.5	46.1	60.0	1.00
Caregiver assistance									
Levels I–III (n = 12)	0	61.1	45.9	65.7	--	60.3	53.6	68.5	--
	5	79.5	69.9	100	.001	86.4	75.2	100	.001
	10	91.6	77.0	100	.031	100	89.4	100	.031
Levels IV–V (n = 12)	0	32.3	29.2	44.4	--	25.4	11.7	36.9	--
	5	54.6	44.4	60.1	.001	53.6	47.2	63.3	< .001
	10	57.9	50.8	61.1	1.00	58.8	49.8	63.4	1.00

Abbreviations: DR, dorsal rhizotomy; GMFCS, Gross Motor Functional Classification System; PEDI, Pediatric Evaluation of Disability Inventory; yr, years.

Data from Josenby et al.¹⁴³ 2015

South Africa, Cape Town, 17- to 26-Year Follow-Up

Langerak et al¹³⁷ evaluated the long-term effect of dorsal rhizotomy on activity and participation of children with spastic cerebral palsy by using the Life-Habit questionnaire. The questionnaire, consisting of 77 life habits related to activities of daily living and social roles, is a reliable and validated outcome measure in children and adults with cerebral palsy.^{145,146} The study cohort consisted of 31 of the 47 adults with cerebral palsy who had undergone dorsal rhizotomy at the Red Cross Children's Hospital in Cape Town between 1981 and 1991. Children had spastic diplegia and were all ambulatory (GMFCS Levels I–III).

Respondents rated five of the six Life-Habit categories for daily activities (nutrition [9.7 ± 0.9], fitness [9.0 ± 1.3], personal care [9.7 ± 0.5], communication [9.8 ± 0.5], and housing [9.3 ± 1.0]) as ≥ 8.0—representing independence without difficulties. Overall, most respondents (81%) reported having no difficulties with these items. They also rated their satisfaction with these items as high. However, the mean score for mobility was 7.1 ± 2.0—representing being independent but with difficulty. Respondents were also less satisfied with their mobility than with other daily activities. Respondents also rated five of the six Life-Habit categories for social roles (responsibilities [9.9 ± 0.3], interpersonal relationships [9.8 ± 0.5], community life [8.9 ± 1.5], education [9.1 ± 0.8], and employment [8.4 ± 2.2]) as ≥ 8.0—representing independence without difficulties. The mean score for recreation (7.4 ± 2.8), representing difficulty with that activity, was lower than the other items. Respondents' satisfaction with this item was also lower than for the other social activities.

CONCLUSIONS. Long-Term Effectiveness of Dorsal Rhizotomy for Functional Independence and Caregiver Assistance

- Information on functional independence and caregiver assistance in children with spastic cerebral palsy after dorsal rhizotomy is more limited than information on gross motor function.
- Long-term cohort studies involved mainly children with spastic diplegia.
- Functional independence for both mobility and self-care significantly improved over baseline in follow-up. Gains for mobility levelled off 5 years after dorsal rhizotomy, whereas gains for functional independence in self-care continued to increase after 5 years.
- Caregiver assistance was assessed in only one cohort; children with spastic diplegia at all levels of pre-operative gross motor disability needed significantly less assistance at 5-year follow-up.
- Caregiver assistance for children with less pre-operative motor disability continued to decrease from 5-year to 10-year follow-up after dorsal rhizotomy. Some children required no assistance at that point.
- Children with higher levels of pre-operative disability reduced reliance on caregiver assistance after dorsal rhizotomy, but improvements levelled off after 5 years. The need for assistance remained greater for them than for less disabled children.
- Overall evidence: 4 studies, GRADE low quality (Table A5)

Treatment Satisfaction at Long Term

Multi-disciplinary teams in three countries—Amsterdam in the Netherlands,¹³⁶ Cape Town in South Africa,^{137,147,148} and the states of Missouri^{149,150} and Michigan¹⁵¹ in the United States—evaluated various respondents' opinions about dorsal rhizotomy for children with spastic cerebral palsy and long-term treatment satisfaction. The seven studies involved several follow-up periods, various methods and measures of treatment satisfaction, and various respondents: parents, children, or therapists (Table 10).

Netherlands Families

In the van Schie et al¹³⁶ follow-up study, parents completed questionnaires and reported on their perceptions of their child's changes in overall function over baseline. Improvement was reported for 91% (20/22) of children and rated as slight (5 children), moderate (11 children), or very much (4 children). Two parents reported that their child's overall function was worse: slightly worse for one child and moderately worse for another child.

Cape Town, South Africa, Families

Researchers for the Cape Town cohort of children with spastic cerebral palsy treated with dorsal rhizotomy reported on treatment satisfaction at three time points for various patient groups involving different sets of respondents: patients or their parents and physiotherapists,¹⁴⁷ parents and children,¹⁴⁸ and children (adults at the time of follow-up interviews).¹³⁷ The hospital providing the surgery was a treatment centre for the entire country, so follow-up was reported to be unusually difficult.

In the Arens et al¹⁴⁷ report, all parents reported that the children were less stiff; 94% (33/35) reported that surgery had been worthwhile. Eleven parents reported that results were amazing, with vast improvements. Specific improvements noted were behavioural improvement (10 parents) and sleeping habits (2 parents). Three parents were uncertain about the value of the surgery—one child neither progressed nor deteriorated, a second parent was uncertain that improvements were due to surgery, and a third parent was not satisfied that treatment goals had been achieved. Nine parents reported unexpected benefits from surgery involving improvements in upper limb function, in balance, or in the ability to walk. Four parents reported that they had been led to expect more benefit from the surgery, and nine said that they had hoped for more. Physiotherapists noted improved motivation or confidence in 18 children. Although attempts were made to survey children, only 15 of the 51 children completed the questionnaire; of these, two were unable to recall their life before dorsal rhizotomy. All thought that the surgery had been worthwhile and that physical therapy still helped—five reported that it helped greatly and one desired more physical therapy.

Peter et al¹⁴⁸ assessed treatment satisfaction among a larger group of South African children (110 children) and reported generally positive responses from patients, parents, or physiotherapists. Overall, respondents for 78% (86/110) of children considered the operation to have been considerably beneficial and would recommend it to others. Among the others, three were ambivalent, therapists or parents of eight children thought the operation was not beneficial, and in 13 cases the outcome was unknown.

Langerak et al¹³⁷ reported the longest follow-up of the South African cohort—17 to 26 years after dorsal rhizotomy. A semi-structured questionnaire was used to interview subjects (young adults when interviewed) about their surgery. Most (74%; 23/31) reported that the surgery had been worthwhile. Seven did not know and one had no positive feelings. In terms of their level of exercise or sport after dorsal rhizotomy, 35% (11/31) of respondents were satisfied. However,

ten believed they should have done more; six regretted discontinuing physical therapy after school, and four did not know if they should have done something differently. When asked if they would undergo the operation, now that they could make independent decisions, 81% (25/31) reported they would undergo the surgery. Four were unsure and two (6%) would not choose to undergo the operation.

United States Families

O'Brien et al¹⁴⁹ at the St Louis Children's Hospital were the only researchers to evaluate parent's satisfaction with dorsal rhizotomy for children with spastic quadriplegia. Overall, 80% (41/51) of parents reported that their children had benefitted from surgery. Parents of younger children (2 to 5 years old) reported that their children benefitted from surgery less often than parents of older children (6 to 14 years old)—75% (27/35) versus 88% (14/16). A second report¹⁵⁰ from this centre examined parents' satisfaction with dorsal rhizotomy for children with spastic diplegia. Most reported that their children had benefitted from surgery, and their satisfaction was high for children of all ages: 59 children 2 to 3 years old (97%); 73 children 4 to 7 years old (93%), and 26 children 8 to 14 years old (96%).

Hurvitz et al¹⁵¹ studied children from two institutions, one in Ann Arbor, Michigan, and one in Chicago, Illinois. Children at the time of surgery were interviewed as adults or young adolescents almost 20 years after their surgery. Most respondents (92%) perceived their overall health to be good or better. Six percent rated their health as fair and 2% as poor. However those reporting their overall health to be very good or excellent varied by their mobility status: GMFCS Levels I to II (63%), GMFCS Level III (70%), and GMFCS Levels IV to V (50%). Most (65%; 57/88) believed that dorsal rhizotomy had increased their quality of life. Of the other respondents, 19 were unsure of the impact, 5 saw no change, and 7 (8%) thought that dorsal rhizotomy had decreased their quality of life. Sixty-five percent would recommend dorsal rhizotomy to others. Thirty-one respondents were unsure, and 5% (4/88) would not recommend the surgery.

Table 10. Treatment Satisfaction at Long Term With Dorsal Rhizotomy

Author, Report Year, Country	Follow-Up Mean \pm SD (Range)	Study Cohort	Response Rate	Treatment Satisfaction Measurement
Van Schie et al, 2011, ¹³⁶ Netherlands	6 years (3–8 years)	33 spastic diplegia: ambulatory (n = 14), nonambulatory (n = 19)	96% (22/23)	Questionnaire evaluation of parents' perception that their child's overall functioning had improved over baseline
Arens et al, 1989, ¹⁴⁷ South Africa	(3–7 years)	60 spastic cerebral palsy	69% (35/51)	Questionnaire evaluation of parents' (mother's, father's, or both) opinions about whether surgery was worthwhile with respect to movement and behaviour, surgical expectations, unexpected gains, and questionnaire evaluation of children's recall of life before surgery and changes after surgery and if physical therapy was helpful. Physiotherapists' case comments on improvements
Peter et al, 1993, ¹⁴⁸ South Africa	10 years	168 spastic cerebral palsy	88% (97/110)	Patient's, parents', and physiotherapist's perception of benefit and their confidence in recommendation
Langerak et al, 2011, ¹⁵² South Africa	(17–26 years)	47 spastic cerebral palsy	74% (23/31)	Patients' (as young adults) reports on worthiness of surgery, satisfaction with activity level after-surgery, willingness to repeat surgery now that they had independent decision
O'Brien et al, 2004, ¹⁴⁹ St Louis (MO) United States	7.5 years (5–9 years)	77 spastic quadriplegia, nonambulatory or requiring assistance	98% (51/52)	Mailed questionnaire, parents' opinion on surgical benefit to their children
O'Brien et al, 2005, ¹⁵⁰ St Louis (MO) United States	7.5 years (5–9 years)	158 spastic diplegia	NR/158	Mailed questionnaire, parents' opinion on surgical benefit to their children
Hurvitz et al, 2013, Michigan and Chicago ¹⁵¹ United States	19.6 \pm 3.0 years	271 spastic cerebral palsy all GMFCS levels (I–IV)	32% (88/271)	Telephone survey or clinic interview of patients (as adolescents and young adults) or their caregivers on perception of dorsal rhizotomy, their quality of life, willingness to recommend dorsal rhizotomy surgery

Abbreviations: GMFCS, Gross Motor Function Classification System; NR, not reported.

CONCLUSIONS: Treatment Satisfaction at Long Term

- Treatment satisfaction with dorsal rhizotomy in the long term was investigated at only a few centres; was inconsistently followed, evaluated, or reported; and did not always identify respondents with certainty: the mother, father, or physiotherapist.
- In studies where families were interviewed, most reported improvements in their children and satisfaction with treatment.
- Satisfaction with surgical treatment of children with spastic quadriplegia undergoing dorsal rhizotomy was investigated at only one centre. Most respondents thought that their children had benefitted from surgery.
- Opinions on treatment satisfaction by children themselves were investigated at two centres but were fully reported at only one centre. At that site respondents (as adolescents and young adults) reported that the surgery had improved their quality of life and they would recommend the surgery to others. However a large percentage (35%) of the respondents were unsure of how surgery had affected their quality of life, thought that there had been no change, or believed surgery had reduced their quality of life.

Safety of Dorsal Rhizotomy for Spastic Cerebral Palsy

Three randomized controlled trials (RCTs)⁷³⁻⁷⁵ that compared dorsal rhizotomy with physical therapy involved mainly ambulatory children with mild spastic diplegia. Safety was not the primary outcome in any of the trials, and trials were small with 30 or fewer participants in the surgical arm. Some surgical practices (from more than 20 years ago) were preliminary, and follow-up was short—1 year or less in two trials^{74,75} and 2 years in one trial.⁷³

No major adverse events were reported in any of the RCTs. All were minor transient peri-operative complications. The RCT by Wright et al⁷⁵ involved 12 children in the surgical arm, and no sensory changes, bladder dysfunction, or spinal abnormalities were noted. There were no complications in the physical therapy control arm. In the RCT by Steinbok et al,⁷⁴ 2 of 14 children in the surgical arm experienced complications—a spinal epidural abscess after surgery and transient urinary retention. There were no complications in the physical therapy control arm. In the RCT by McLaughlin et al,⁷³ almost all children (20 of 21 in the surgical arm and 17 of 17 in the physical therapy arm) had one or more minor complications. Most complications in both study arms involved back or leg pain, fatigue, or weakness. Complications of the urinary tract (3 children) and paresthesia in the lower extremity (4 children) were reported only for the surgical arm. Emotional and behavioural issues during physical therapy were similar: four children in each study arm.

Observational studies investigating complications involved larger study groups, more diverse patient groups, and longer follow-up (Table A4). The thirty-seven studies involved investigators and various clinical protocols from institutions in nine countries. In most (95%) clinical reports, a safety outcome was investigated as a primary study objective. Diverse safety outcomes were reported in the studies and were evaluated in this evidence review in the following categories: peri-procedural complications (4 studies); bladder dysfunction (9 studies); somatosensory alterations (five studies); spinal bony abnormalities (15 studies); and hip instability (8 studies).

Peri-operative Complications

Short-term or peri-operative complications were reported for practices from four institutions in three countries: Canada,¹⁵³ Korea,^{154,155} and the United States.¹⁵⁶⁻¹⁵⁸ All studies included operative and post-operative complications for study groups of more than 100 children with spastic diplegia or spastic quadriplegia undergoing dorsal rhizotomy (Table 11).

In the Canadian study by Steinbok et al¹⁵³ at the Vancouver Children's Hospital, complications were evaluated for 158 children treated in a 10-year period. Changes in surgical practices over time had involved various methods to determine the amounts and types of dorsal roots to be sectioned, reducing the number of dorsal rootlets sectioned, and sectioning less of the L3 and L4 spinal levels. Intra-operative complications were reported for 5 children. The mean length of hospital stay was 6.4 days; a longer than expected hospital stay was reported for one child who developed subdural hematoma

Post-operatively, back pain, constipation, and emesis were common side effects after surgery and were not considered complications. Many (44%, 69/158) children had one or more complications involving various systems (Table 10). Of these, hospital stay was longer than expected for seven children because of post-operative complications evaluated as major. *Overall, the peri-operative major adverse event rate, defined as having a longer than expected hospital stay, was 5.1% (8/158).*

In Korea, Kim et al^{154,155} at the Yonsei University College of Medicine in 2001¹⁵⁴ initially reported peri-operative complications for 208 children treated in their 10-year study. Intraoperatively, two patients experienced aspiration pneumonia. Post-operatively, most (83%) patients experienced temporary muscle hypotonia (floppy or low tone) that persisted in seven patients (3.5%). Back pain was common but was managed with opioids. Other complications included urinary retention in 20 patients, which was long-standing in two patients. Temporary sensory abnormalities were found in 15 patients; abnormalities persisted in five patients (2.4%) at follow-up. *Overall, the peri-operative major adverse event rate was 5.3% (11/208).*

In the American study by Abbott et al¹⁵⁶ at the New York University Medical Center, peri-operative complications were initially reported in 1992 for 250 patients. Follow-up and management of a subgroup of these patients was reported in 1993.¹⁵⁷ Peri-operative complications were reported as pulmonary, bowel and bladder retention, and altered sensations. Post-operative pain was severe in 145 patients (58%), and dysesthesia persisted for several weeks in 100 patients. Overall, 35 of the peri-operative complications (Table 10) were judged to be serious adverse events; the *peri-operative major adverse event rate was 14% (35/250).*

Protocols at that institute evolved to identify those at risk and to better manage complications. Peri-operative administration of intravenous aminophylline subsequently reduced bronchospasm to two children among the 190 treated with the protocol. Aspiration pneumonia was also reduced by administering histamine receptor antagonists to children with a history of gastric reflux or pneumonia. Since this protocol was introduced, no serious cases of aspiration pneumonia have occurred in 180 rhizotomy procedures. Earlier, three of five aspiration pneumonia cases required artificial ventilation. The three children who developed post-operative ileus required an additional intervention of 48 hours of nasogastric suctioning. Four children experienced persistent focal areas of sensory loss—three lost proprioception (awareness of position) in their toe—and one lost pain and temperature sensation.

In the other American study by Van De Wiele et al¹⁵⁸ at the UCLA Medical Center, peri-operative complications were reported for 105 consecutive children treated over a 6-year study period. Sixty-eight children had previous surgeries, and eight had prior complications related to general anesthesia. No major surgical complications, such as wound infection, dural leak, hemorrhage, altered lower extremity sensation, or motor dysfunction, were reported. The mean operating time was 4.4 ± 0.7 hours and several intra-operative cardiovascular events were reported (Table 11). Of the seven cases of transient dysrhythmia, four occurred with nerve root stimulation.

Post-operatively, nine patients required supplemental oxygen; eight of those patients had been born prematurely. All patients were treated with narcotics for post-operative pain; six patients became constipated and two had persistent nausea and vomiting. Urinary tract infections were the most common complication, occurring in 10 patients, six of whom had Foley catheters placed. No bladder dysfunction was noted, and this was attributed to the conservative approach to nerve rootlet dissection at the S2 spinal level. *Overall, no peri-operative events were reported to be major in the cohort.*

Table 11. Peri-Operative Complication Events

Author, Year, Country	Cohort	Intra-operative Adverse Events (n = Patients)	Post-operative Adverse Events (n = Patients)	Peri-operative Major Adverse Event Rate ^a and Events (n = Patients)
Steinbok et al, ¹⁵³ 1998, Canada	N = 159	Aspiration pneumonia (n = 2) Bronchospasm (n = 1) Subdural hematoma (n = 1) Hypothermia (n = 1)	Dermatologic (n = 33) Neurologic (n = 17) Urologic (n = 18) Pulmonary (n = 11) Other (n = 4)	5.1% (8/158) Subdural hematoma (n = 1) Dysesthesia (n = 3) Urinary retention (n = 7) Bronchopneumonia (n = 1) Wound infection (n = 1)
Kim et al, ¹³¹ 2001, South Korea	N = 208	Aspiration pneumonia (n = 2)	Muscle hypotonia (n = 173) Urinary retention (n = 20) Sensory abnormalities (n = 15)	5.3% (11/208) Aspiration pneumonia (n = 2) Urinary incontinence (n = 2) Muscle hypotonia (n = 7) Sensory abnormalities (n = 5)
Abbot et al, ¹⁵⁶ 1992, United States	N = 250	Bronchospasm (n = 13) Aspiration pneumonia (n = 5)	Urinary retention (n = 13) Constipation (n = 49) Ileus (n = 3) Proprioceptor loss (n = 3) Pain, temperature sensory alteration (n = 3) Severe pain (n = 145) Dysesthesia (n = 100)	14% (35/250) Major events not specified
Van de Wiele et al, ¹⁵⁸ 1996, United States	N = 105	Tachycardia (n = 10) Bradycardia (n = 1) Transient dysrhythmia (n = 7)	Supplemental oxygen (n = 9) Persistent nausea or vomiting (n = 2) Urinary tract infection (n = 6)	0 major events

^aPeri-operative major adverse event rate included major adverse events occurring intra-operatively or post-operatively.

CONCLUSIONS. Peri-operative Complications After Dorsal Rhizotomy

- Major peri-operative complications of dorsal rhizotomy were infrequently reported in large cohorts at four institutions.
- Over the course of lengthy experience, surgeons introduced protocols to mitigate surgical and medical complications, greatly reducing complication events.
- Overall evidence: 4 studies; *modified* McHarm 3 moderate, 1 low quality (Table A7)

Bladder Dysfunction

Nine investigators^{126,153,155,159-164} evaluated how dorsal rhizotomy affected bladder symptoms (frequency, urge, or incontinence) and function in children with spastic cerebral palsy (Table 12). In three studies^{126,153,155} bladder function was evaluated along with other complications. The other five studies^{159-162,164} focused only on how dorsal rhizotomy affected bladder function; three of these reports^{159,161,164} included pre-operative and post-operative urodynamic studies. Three investigators^{153,155,161} also evaluated the relationship between the extent that sacral nerve rootlets were sectioned and the occurrence of subsequent bladder complications.

Clinical Investigations Only

Several investigators from the New York University Medical Center published three reports on bladder dysfunction and sacral nerve rootlet sectioning in children undergoing dorsal rhizotomy at their institute.^{160,162,163} In the first report by Deletis et al,¹⁶⁰ 31 patients undergoing dorsal rhizotomy had additional monitoring of the afferent fibres of the pudendal nerves to guide sectioning of the sacral nerve rootlets. No post-operative urologic complications were reported using this technique. Before this technique was introduced, 13 of the 55 patients undergoing dorsal rhizotomy experienced urinary retention that was transient in 12 patients and persistent in one patient at 1-year follow-up. Of the 13 patients, 11 had the S2 roots sectioned bilaterally.

However, in the second report from the New York University Medical Center, Lang et al¹⁶³ reported that not sectioning any S2 roots resulted in residual spasticity of the plantar flexors (foot flexion). Of 13 patients in which only L2 to S1 were sectioned, five (38%) had functionally limiting residual lower limb spasticity. Of 72 patients in which L2 to S2 were sectioned, eight (11%) had functionally limiting residual spasticity.

In the third report from the New York University Medical Center, Huang et al¹⁶² reported on pudendal afferent monitoring to guide sectioning of the S2 rootlets in 114 children with spastic cerebral palsy. None had clinically relevant bladder dysfunction or recurrent urinary infection before dorsal rhizotomy. Monitoring was successful in most (92%, 105/114) patients, and 56% of pathologically responding rootlets were preserved during testing. A few patients (5%) had post-operative urinary retention, but all were able to void spontaneously within 1 to 2 weeks.

Steinbok et al¹⁵³ reported on post-operative complications including bladder dysfunction in 158 Canadian children with spastic cerebral palsy, both diplegia and quadriplegia, after dorsal rhizotomy. Twenty children (12.7%) had neurogenic bladder (impaired bladder control) complications including urinary incontinence (14 patients), urinary and fecal incontinence (1 patient), and fecal incontinence (1 patient). In 15 of these patients, urinary incontinence was new or worsening. Overall, of the eight patients (5%) with persistent neurogenic bowel or bladder problems attributed to surgery, four were incontinent prior to surgery.

Surgical protocols changed over time: decreasing the number of rootlets sectioned, reducing reliance on abnormal electrophysiologic responses, and increasing pudendal nerve monitoring to guide sectioning of sacral nerve roots to avoid potential bowel and bladder complications. Several outcomes were associated with these trends. The 19 patients whose S2 rootlets were not sectioned had no urinary or bowel dysfunction in follow-up. Among patients whose S2 rootlets had been sectioned, the urinary and fecal incontinence rate was lower for those who had pudendal nerve monitoring (7.7%) than for those who had not (13.8%). The median percentage of sacral nerve rootlet sectioning was also lower in cases with pudendal monitoring than in those without monitoring (31% vs. 54%; $P = .001$).

Kim et al¹⁵⁵ reported on peri-operative complications, including bladder dysfunction, in 200 Korean children with spastic diplegia or quadriplegia who had undergone dorsal rhizotomy a year prior. During the 10-year study period, 20 patients had post-operative voiding problems, mainly urinary retention due to decreased bladder tone and hyporeflexia. Most cases (18/20), however, resolved within 4 weeks after surgery. In two patients (1%) urinary incontinence persisted throughout follow-up. Voiding difficulties, both transient and permanent, correlated with the number of sacral nerve rootlets sectioned.

As with the study by Steinbok et al,¹⁵³ changing surgical protocols over the study period produced various patient subgroups depending on whether S2 nerve rootlets were sectioned unilaterally, bilaterally, or to different degrees. In the two cases of persistent urinary incontinence (1.3%; 2/159), S2 rootlets were sectioned bilaterally and more extensively. Temporary voiding difficulties were also mainly associated with bilateral sectioning (10%; 16/159) rather than unilateral sectioning (5%; 1/20) of sacral nerves or with preserved S2 rootlets (5%; 1/21).

Montgomery et al¹²⁶ reported on follow-up of 14 children with spastic cerebral palsy, mainly quadriplegia (8 children) for peri-operative complications, including symptoms related to the bladder. Bladder symptoms were noted in four children (29%) at follow-up: one lost bladder control (requiring catheterization) and three others had bladder difficulties related to initiating voiding, periodic incontinence, and urgency. Of the four children who developed bladder symptoms after surgery, three had established bladder control before surgery.

Clinical Investigations and Urodynamic Studies

Sweester et al¹⁶⁴ reported on bladder function in 34 children with spastic cerebral palsy (including both diplegia and quadriplegia) undergoing dorsal rhizotomy with rootlet sectioning performed from L2 to S2 without pudendal monitoring. Of the 24 diplegic patients, 14 were incontinent before and 6 were incontinent after surgery. Bladder function of the ten asymptomatic patients was not affected by the surgery. All but one of the nine quadriplegic children remained incontinent after surgery. One hemiplegic patient remained continent. Video urodynamics with fluoroscopy was selectively performed in 12 patients, but only two of them had post-operative urodynamic studies.

Houle et al¹⁶¹ reported on bladder function in 40 ambulatory children with spastic cerebral palsy undergoing dorsal rhizotomy with rootlet sectioning from L1 to S2 without pudendal monitoring. Sixty-six percent (23/35) of the children had abnormal results from pre-operative urodynamic studies, including abnormal total bladder capacity, pressure-specific volumes, and full resting pressures for age. Most of those with abnormal urodynamic studies (17/23) were asymptomatic. Of the seven children who were incontinent before surgery, five became continent afterward. Total bladder capacity ($P = .004$) and pressure-specific volumes ($P = .0004$) improved in the 13 children who had pre- and post-operative urodynamic studies.

Chiu et al¹⁵⁹ reported on bladder function in 54 children with spastic cerebral palsy, 49 with diplegia, 3 with quadriplegia, and 2 with triplegia. Dorsal rhizotomy was performed over a 7-year period, during which surgical protocols changed from multi-level laminectomy (33 patients) with sectioning from L2 to S1 (21 patients with S2 sectioning) to single-level laminectomy (21 patients) with sectioning from L2 to S1, none with S2 sectioning. Urinary symptoms of urgency and frequency improved ($P = .013$) after dorsal rhizotomy. Of the 22 patients with pre-operative urge or frequency, 12 (55%) had none post-operatively. Of the 32 patients without pre-operative

urge or frequency, two patients (6%) developed it post-operatively. Incontinence also improved significantly ($P = .013$) after dorsal rhizotomy. Of the 16 patients who were incontinent pre-operatively, 12 (75%) became continent post-operatively. Two of the 38 patients (5.3%) who were continent before developed incontinence after surgery.

For bladder function, the effect of S2 rootlet sectioning was more apparent on incontinence than on urge or frequency. Of the 16 patients with incontinence before surgery, 8 had S2 sectioning, and all became continent after surgery. Four of the other eight without S2 sectioning became continent. Among 22 patients with pre-operative urge or frequency, 6 of the 10 with S2 sectioning and 6 of the 12 without S2 sectioning were without these symptoms after surgery. Bladder symptoms also improved post-operatively in all three spastic quadriplegic children; two had urgency and incontinence and one had frequency only.

Urodynamic studies of the children included a range of parameters: bladder capacity; percentage of expected bladder capacity; neurogenic detrusor overactivity (i.e., uncoordinated bladder muscle activity); and incontinence. Studies were performed pre-operatively in 51 children and both pre-operatively and post-operatively in 20 children. The low completion rate for post-operative studies was attributed to patients' refusal of tests that would not have altered management. Pre-operative detrusor overactivity and incontinence were noted in 71% (36/51) and 28% (14/51), respectively, of the children. In the pre-operative and post-operative urodynamic studies obtained for 20 patients, only bladder capacity improved significantly ($P = .016$) from 69.8 ± 50.8 mL to 131.6 ± 71.4 mL. Recurrent or worsening bladder symptoms were not noted for any children in the median 6-year follow-up.

Table 12. Bladder Dysfunction Following Lumbosacral Dorsal Rhizotomy

Author, Year, Country	Cohort (N = Patients)	Pre-operative Bladder Status (n = Patients)	Post-operative Bladder Status (n = Patients)
Deletis et al, ¹⁶⁰ 1992, United States	N = 55	--	Urinary retention, transient (n = 12) persistent requiring intermittent catheterization (n = 1)
Huang et al, ¹⁶² 1997, United States	N = 114	No bladder dysfunction or recurrent urinary infection	Urinary retention, transient (n = 6)
Steinbok et al, ¹⁵³ 1998, Canada	N = 158 Hemiplegia (n = 86), quadriplegia (n = 66), hemiplegia (n = 2), triplegia (n = 2)	--	Neurogenic bladder and bowel complications (n = 20) (urinary incontinence (n = 14), urinary and fecal incontinence (n = 1), fecal incontinence (n = 1), urinary urgency (n = 3), urinary frequency (n = 3), urinary hesitancy (n = 2), incomplete bladder emptying (n = 1) Persistent neurogenic bladder symptoms (n = 8; of 6 involving incontinence, 3 were incontinent pre-operatively)
Kim et al, ¹⁵⁵ 2002, South Korea	N = 200	--	Voiding difficulty, transient (n = 18) Urinary incontinence, persistent (n = 2)
Montgomery et al, ¹²⁶ 1992, United States	N = 14, Quadriplegia (n=8)	Bladder dysfunction (n = 1)	Bladder dysfunction (n = 4) (urinary retention, initiating voiding, periodic incontinence, and urgency)
Sweetser et al, ¹⁶⁴ 1995, United States	N = 34 Diplegia (n = 24), quadriplegia (n = 9), hemiplegia (n = 1)	Incontinent (n = 23) Continent (n = 11)	Incontinent (n = 14) Continent (n = 120)
Houle et al, ¹⁶¹ 1998, Canada	N = 40	Incontinent (n = 7) No urinary symptoms (n = 33)	Incontinent (2/7)
Chiu et al, ¹⁵⁹ 2014, Hong Kong	N = 54 Diplegia (n = 49), quadriplegia (n = 3), triplegia (n = 2)	Urge or frequency (n = 22) No urge or frequency (n = 32) Incontinent (n = 16) Continent (n = 38)	Urge or frequency (10/22) No urge or frequency (30/32) Incontinent (4/16) Continent (36/38)

CONCLUSIONS. Bladder Dysfunction After Dorsal Rhizotomy

- Over time surgeons modified protocols to more carefully select and section spinal sacral rootlets; these changes greatly reduced the early incidence of bladder symptoms after dorsal rhizotomy.
- Urinary incontinence in children with spastic diplegia developed infrequently after dorsal rhizotomy.
- Children with spastic diplegia who had urinary incontinence before dorsal rhizotomy usually gained continence after surgery.
- Urodynamic studies confirmed a high pre-operative rate of urinary dysfunction, neurogenic detrusor overactivity, or detrusor incontinence in children with spastic diplegia.
- Among children with spastic quadriplegia, who were less often represented in studies and often had pre-operative bladder symptoms and incontinence, dorsal rhizotomy generally had a more neutral effect on bladder symptoms.
- Overall evidence: 9 studies; *modified* McHarm 3 moderate, 6 low quality (Table A7)

Sensory Abnormalities

Dorsal rhizotomy involves the sectioning of nerves in the lumbosacral regions and, depending on the degree of nerve root sectioning, can potentially cause nerve damage or altered somatosensory sensations. Five investigators evaluated sensory dysfunction in children with spastic cerebral palsy undergoing dorsal rhizotomy.^{126,148,153,165,166}

Peter et al¹⁴⁸ reported on complications that included sensory alterations in 110 South African children with diplegic or quadriplegic cerebral palsy. Post-operative sensory disturbances in the legs or sides of the feet were common. All who could communicate reported sensory effects that were described as dysesthetic: painful, persistent sensations aggravated by touch or bedclothes. Symptoms were disturbing for 25 patients up to 6 months or a year. Proprioceptive loss in the L3 to L5 dermatomes was noted for 13 patients, and seven (6.4%) had patchy areas of dysesthesia that persisted.

Steinbok et al¹⁵³ also reported on complications following dorsal rhizotomy, which included sensory alterations in 158 Canadian children who had either diplegic or quadriplegic spastic cerebral palsy. Neurologic complications were noted in 26 patients; 22 (13.9%) had sensory abnormalities that included paresthesia (sensation of tingling, pricking, or burning) in 10 children (6.3%), hyperesthesia or allodynia (abnormally increased sensitivity to stimuli, particularly touch) in 8 (5.1%), and hypesthesia or hypoesthesia (decreased sensation) in 5 (3.2%). Sensory abnormalities in most children resolved over time, with an average time to resolution of 19 months. In six children (3.8%) the sensory abnormalities persisted to last follow-up after more than 2 years—disturbances did not interfere with children's activities or require additional treatment.

The relationship between decreasing numbers of dorsal rootlets sectioned during dorsal rhizotomy and subsequent sensory abnormalities was examined over the 10-year study. The median number of spinal L5 and S1 rootlets sectioned was ($P = .02$) higher in patients with sensory abnormalities. The highest percentage of patients with sensory abnormalities (33%; 8/24) occurred in the initial phase of the study, where decisions to section nerves were made solely on electrophysiologic guidance. All cases with persistent hyperesthesia occurred in the first phase of the study.

Montgomery et al¹²⁶ and by Parise et al¹⁶⁶ investigated sensory abnormalities among two small cohorts. Montgomery et al¹²⁶ studied 14 children with cerebral palsy, mainly quadriplegia, over a mean 27-month follow-up after dorsal rhizotomy. Twelve of the children developed sensory abnormalities—two had had pre-operative abnormalities. Post-operative sensory abnormalities included hypoesthesia (4 children), paresthesia (4 children), and both hypoesthesia and paresthesia (4 children). Most symptoms were determined by functional observation, although one patient had hypoesthesia of the lower limb (right leg and foot) where a lack of response to neuromuscular stimulation was documented.

In the Parise et al¹⁶⁶ study, sensory abnormalities and nerve function were assessed in 10 children with spastic diplegia undergoing dorsal rhizotomy. Nerve function was evaluated by cortical somatosensory evoked potentials (SEP) from electrical stimulation of nerves in the lower extremities. Tests were performed pre-operatively and 6 months post-operatively. No pre-operative somatosensory deficits were noted on clinical examination, but most (7/10) patients had abnormalities in tibia nerve SEP components. Although some patients exhibited transient post-operative hyperesthesia of the lower limbs, no deficits were detected at 6-month follow-up. The abnormal SEPs or nerve transmission values were also significantly reduced after surgery.

McLaughlin et al¹⁶⁵ performed the most extensive evaluation of how dorsal rhizotomy affected lower extremity sensory function in children with cerebral palsy. Sensory function was evaluated in three groups of children: those with cerebral palsy (62 children) from specialty clinics; a comparison group of 65 children without cerebral palsy and no history of sensory deficits or medical condition associated with cerebral palsy; and children with spastic cerebral palsy undergoing dorsal rhizotomy (19 children). A complete sensory test battery was performed for lower extremity sensitivity function; test items included light touch, pain sensation, position sense, vibration sense, and direction of scratch.

There was a significant difference between children with cerebral palsy and the comparison group of children without cerebral palsy in their ability to complete sensory testing. Overall 85% (55/65) of children without cerebral palsy were able to complete the sensory testing compared with 52% (32/62) of children with cerebral palsy. Test performance also varied by age in both groups. Children younger than 5 years of age were less likely to complete sensory tests: no children with cerebral palsy and 50% (10/20) of children without cerebral palsy completed the tests. Children with cerebral palsy at all ages were less able to complete sensory testing than those without cerebral palsy. They were also significantly less able to correctly identify sensory stimuli than those without cerebral palsy—pinprick ($P = .02$), toe position ($P = .01$), vibration sense ($P = .01$), and direction of scratch ($P < .001$).

A subset of children in this study participated in an RCT of dorsal rhizotomy and physical therapy (19 children) versus physical therapy only (15 children).⁷³ In that study a conservative rate of dorsal tissue sectioning was performed (on average 34%), ranging from 20% to 56%. As the children in this study were younger, there was a high failure rate in the ability to complete sensory testing at baseline for many of the sensory tests—pinprick (26%), vibration sense (35%), toe position (76%), direction of scratch (56%). Of children who were able to complete the sensory tests at 1-year follow-up, there was no difference in the number of children in the study arms who correctly identified the sensory stimuli.

CONCLUSIONS. Sensory Abnormalities After Dorsal Rhizotomy

- Pre-operatively children with spastic cerebral palsy have abnormal lower limb sensory nerve transmissions and those younger than 5 years of age cannot complete sensory nerve testing.
- After dorsal rhizotomy, patchy areas of lower limb sensory disturbances were common but not persistent and without clinical sequelae.
- Long-term persistent sensory abnormalities after dorsal rhizotomy were uncommon.
- Overall evidence: 5 studies; *modified* McHarm 2 moderate, 3 low quality (Table A7)

Spinal Bony Abnormalities

Ten investigators from seven countries—Canada,^{167,168} Korea,¹⁵⁵ China,¹⁶⁹ Germany,⁶⁷ Netherlands,¹³⁶ United States,¹⁷⁰⁻¹⁷² and South Africa¹⁷³⁻¹⁷⁵—evaluated one or more spinal bony deformities or instabilities in twelve longitudinal follow-up studies of children with spastic cerebral palsy undergoing dorsal rhizotomy (Table 13).

Three other reports involved case studies^{176,177} or case series¹⁷⁸ detailing severe spinal deformity and outcomes of surgical correction after dorsal rhizotomy. A small case series by Mooney et al¹⁷⁸ was the first to describe spinal deformities progressing rapidly after dorsal rhizotomy for children with spastic cerebral palsy. In their cases, severe hyperlordosis (Cobb angles -80° to -112°) presented clinically on average 11 months after surgery. All cases affected nonambulating children with spastic quadriplegia. Pre-operatively, all children also had some clinically apparent lordosis, and three were known to have scoliosis. Surgical correction was required in five of six children. Crawford et al¹⁷⁶ reported two cases of extreme lordosis (Cobb angles -112° , -137°) in children with spastic quadriplegia after dorsal rhizotomy. Gooch et al¹⁷⁷ were the only researchers to document spinal stenosis appearing after dorsal rhizotomy. They reported this complication in 1.5% (2/130) of children with spastic diplegia undergoing surgery at their institution. At surgery, both cases were noted to have significant facet hypertrophy.

The results of the 12 clinical studies evaluating spinal abnormalities with radiologic follow-up after dorsal rhizotomy are detailed below: scoliosis (Table 14), kyphosis or lordosis (Table 15), or spinal instability of spondylolisthesis or spondylolysis (Table 16). The conduct and reporting of radiographic investigations in these studies varied and was often incomplete. Two studies^{170,171} demonstrated significant differences in sitting or standing (full weight-bearing) examinations; details of examination procedures were not always reported. In general, radiographic examinations performed in non-weight-bearing positions (rather than standing) underestimated incidence of spinal abnormalities.

Investigators evaluated different risks in children after dorsal rhizotomy—risk for progression of existing deformities as well as risk of developing new abnormalities. Pre-operative radiologic evaluations to establish existing spinal conditions were not always performed or reported. Paired pre-post radiologic examinations were often available only for subsets of the study cohorts, limiting conclusions on these risks. Studies that restricted their study cohorts to those without radiologically confirmed pre-operative spinal abnormalities provided stronger evidence on the risks that these conditions would develop or progress.⁶⁷

Table 13. Clinical Studies Evaluating Spinal Bony Abnormalities After Dorsal Rhizotomy for Children with Spastic Cerebral Palsy

Author, Year, Country	Study Population Mobility Status	Radiographic Follow-Up Mean \pm SD (Range)	Surgical Approach	Pre-operative Radiographic Examinations	Post-operative Radiographic Examinations	Paired Pre- and Post-operative Radiographic Examinations	Spinal Abnormality Reported)
Steinbok et al, ¹⁶⁸ 2005, Canada	N = 105 Ambulatory and nonambulatory	4.3 years (1–13.6 years)	Multilevel laminectomy or laminoplasty with replacement laminae	N = 105	N = 105 Sitting and standing	N = 44	S, K, L, SP)
Golan et al, ¹⁶⁷ 2007, Canada	N = 98 Ambulatory and nonambulatory	5.8 years (1.1–11.5)	Multilevel laminectomy with replacement laminoplasty	N = 39	N = 87 Standing	N = 35	S, K, L, SP
Li et al, ¹⁶⁹ 2008, China	N = 61 NR	6.3 years (5–9 years)	Multilevel laminectomy L2–S1	N = 61	N = 61	N = 61	S, K, L
Funk et al, ⁶⁷ 2016, Germany	N = 116 Ambulatory GMFCS Levels I–III	33 months (12–81 months)	Single-level laminectomy with lamina replacement	N = 72	N = 72	N = 72	S
Kim et al, ¹⁵⁵ 2002, Korea	N = 200 Ambulatory and nonambulatory	4.0 years (1–9 years)	Multiple laminectomy L1–S1 (n = 58) and laminotomy L1–L5 and sacral laminectomy (n = 142)	N = 200	N = 188	--	S, K, SP
Van Schie et al, ¹³⁶ 2011, Netherlands	N = 33 Ambulatory	3.0 years	Multiple laminectomy L2–L5	N = 33	N = 27	N = 27	S, SP
Gooch et al, ¹⁷⁷ 1996, Utah, US	N = 2 Dependent ambulatory	4 years, 3 years	Laminotomy	--	N = 2	--	SS
Crawford et al, ¹⁷⁶ 1996, Texas, US	N = 2 Nonambulatory quadriplegia	20 months, 5 years	--	--	N = 2	--	L
Mooney et al, ¹⁷⁸ 1999, Boston, US	N = 6 Nonambulatory quadriplegia	11 months (3–30 months)	--	N = 3	N = 6	--	L, S
Spiege et al, ¹⁷¹ 2004, Minneapolis (MN), US	N = 90 Ambulatory	4.2 \pm 2.0 years N = 79 for > 2 years	Multiple laminoplasty L1–L5 with lamina replacement	N = 74 Sitting (n = 50) and standing (n = 26) ^a	N = 77 Sitting (n = 51) and standing (n = 26)	--	S, K, L, SP
Johnson et al, ¹⁷⁰ 2004, Oregon, US	N = 34 Ambulatory	8.6 years (5–11.6 years)	Multiple laminectomy L1–L5 (n = 14) and multiple laminoplasty (n = 20) T12–L5, T12–L4, or L1–L5 (n = 20)	N = 34 Sitting or standing	N = 34	N = 34	S, T, L, SP
Turi et al, ¹⁷² 2000, Florida, US	N = 47 Nonambulatory and ambulatory	5.3 years (2–9 years)	Multiple laminectomy	N = 47	N = 47	--	S, K, L
Peter et al, ¹⁷⁵ 1990, South Africa	N = 55 Nonambulatory and ambulatory	4.5 years (1–7 years)	Multiple laminectomies without lamina replacement	--	N = 57	--	S, K, L, SP

Author, Year, Country	Study Population Mobility Status	Radiographic Follow-Up Mean ± SD (Range)	Surgical Approach	Pre-operative Radiographic Examinations	Post-operative Radiographic Examinations	Paired Pre- and Post-operative Radiographic Examinations	Spinal Abnormality Reported)
Peter et al, ¹⁷⁴ 1993, South Africa	N = 99 Nonambulatory and ambulatory	--	Multiple laminectomies	--	N = 99	--	SP
Langerak et al, ¹⁷³ 2009, South Africa	N = 30 Ambulatory	21.4 ± 3 years (17–26 years)	Multiple laminectomy L1/2–S1 (70%), L1–L5 (10%), L2–L5 (10%), L2–S2 (3%), or L3–S1 (7%)	N = 28 Sitting or standing	N = 30	N = 28	S, K, L, SP

Abbreviations: GMFCS, Gross Motor Function Classification System; L, lordosis; K, kyphosis; N, number; S, scoliosis; SP, spondylolysis/spondylolisthesis; SS, spinal stenosis.

^aTotals do not add because some patients were lost to follow-up.

Scoliosis

Children with cerebral palsy are more frequently affected by spinal abnormalities, particularly scoliosis, than the general population.¹⁷⁹ The prevalence of idiopathic scoliosis (no known cause) is 1% to 2% of the general population, whereas the prevalence of scoliosis in cerebral palsy ranges dramatically between 15% to 80%, depending on the diverse populations studied, the extent of functional impairment, radiographic methods, and the case definition.¹⁸⁰ Scoliosis has traditionally been defined as a 10° lateral curvature of the spine based on a Cobbs angle measured on radiograph.¹⁷⁹

Development of scoliotic patterns is related to the ambulatory ability and neurologic deficits of children with cerebral palsy. The incidence of scoliosis in spastic diplegia has been reported to be 5% compared with 65% to 74% in spastic quadriplegia.¹⁸¹ Several series have reported high prevalence (> 60%) of scoliotic patterns among residents with cerebral palsy in institutions.¹⁸²⁻¹⁸⁴ Occurrence and progression of scoliotic patterns are related to several factors—asymmetrical body positioning, trunk instability, muscular imbalance, and abnormal contractile patterns of axial muscles and secondary contractures.¹⁶⁷ Children with cerebral palsy with good ambulation and less severe neurologic deficits tend to have simpler or single thoracic or thoracic lumbar curves, whereas those with more neurologic deficits have more complex, longer, and stiffer spine curvature patterns.¹⁸⁵

During adolescent growth periods, scoliotic curves tend to progress at a rate of 2° to 4° monthly.^{186,187} Beyond skeletal maturity, the rate of progression in children with cerebral palsy is slower and has been estimated to vary by the severity of their curvature at maturity, progressing at a rate of 0.8° yearly for curves less than 50° and between 1.4° to 4° yearly for curves greater than 50°.¹⁸⁴ At skeletal maturity, scoliosis with curves greater than 40° was reported in 30% of spastic quadriplegia, 10% of spastic diplegia, and 2% of spastic hemiplegia cases.¹⁷⁹

Scoliotic patterns and neurologic impairment can combine to impair many functions including walking capacity, sitting tolerance, and cardiopulmonary performance.^{179,188} Progressive scoliosis can compromise lung function by deforming the chest cage and compressing lungs.¹⁸⁹ Pneumonia frequently causes repeated hospitalization among cerebral palsy patients, and respiratory failure is the most common cause of mortality.^{187,189,190} Surgical correction or stabilization is often considered for scoliotic angles above 40° or 45°.^{180,191,192}

In this evidence review, the results of the 11 studies investigating scoliosis in children with spastic cerebral palsy after dorsal rhizotomy are detailed (Table 14). Six studies involved study populations (mixed) of both ambulatory and nonambulatory children, and five studies involved only ambulatory populations. The pre-operative mean Cobb angle in three studies involving (on average) 5-year follow-up of mixed study populations (both ambulators and nonambulators)^{167,168} changed by 1.9° and 5.9° post-operatively and by 4° for a study population involving only ambulators.¹⁷⁰

The post-operative prevalence of scoliosis (defined as a Cobb angle \geq 10°) increased by 12%, 16%, and 26% over baseline in three studies of ambulators and nonambulators and increased by 17%, 23%, and 57% in three studies of ambulators. When a greater threshold for scoliosis (Cobb angle \geq 25°) was reported for ambulators and nonambulators, the increase in post-operative prevalence was 2%. At a threshold of > 20° for ambulators, increase in post-operative prevalence was 3%, 3%, and 9%. At a threshold of > 30°, increase in post-operative prevalence was 7%. Peter et al¹⁷⁵ at 5 years and Langerak et al¹⁷³ at 28 years reported on follow-up of the

same cohort. The 5-year follow-up showed a prevalence of scoliosis of 16% and the 28-year follow-up of 57%. The prevalence at 28 years (of a Cobb angle $\geq 30^\circ$) was 7%.

Three investigators^{167,168,172} evaluated change of scoliosis, either as worsening or improving, defined by $\geq 10^\circ$ change, after dorsal rhizotomy for individual children. Golan et al¹⁶⁷ reported that scoliosis (in their paired pre-post subsample of 35) worsened in 9 children, improved in 6, and was stable in 20 children. Steinbok et al¹⁶⁸ reported 4.8% (5/104) of children as improving with a median improvement of 15° (range 10° to 21°). A quarter (26/104) of cases became worse, with a median of 14° (range 10° to 48°). Turi et al¹⁷² reported that two of the three patients in the largely nonambulatory cohort (30/47) having pre-operative scoliosis significantly progressed after surgery—in one case from mild scoliosis to a curve of 42° and in the other case from a curve of 60° to 85° . Of the 44 children in this cohort who did not have pre-operative scoliosis, 27% (12/44) developed scoliosis with a mean Cobb angle of 36° (range 10° to 79°) during the 5-year follow-up.

Table 14. Scoliosis in Children With Spastic Cerebral Palsy After Dorsal Rhizotomy

Radiologic Measure	Pre-operative	Post-operative	Pre- to Post-operative Change
Ambulatory and Nonambulatory Spastic Cerebral Palsy Cohorts			
Golan et al,¹⁶⁷ 2007, 5.8-year follow-up			
Paired pre-post, Cobb angle Mean \pm SD (range)	6.4° (0 to 21°)	8.3° (0 to 30°)	1.9°
Paired pre-post, proportion Cobb angle $\geq 10^\circ$	31% (11/35)	43% (15/35)	12%
Paired pre-post proportion Cobb angle $\geq 25^\circ$	--	2% (2/87)	--
Paired pre-post, number worsening by $\geq 10^\circ$	--	9	--
Paired pre-post, number improving by $\geq 10^\circ$	--	6	--
All exams, Cobb angle Mean	--	8.5°	--
All exams, proportion with Cobb angle $\geq 10^\circ$	--	45% (39/87)	--
All exams, proportion with Cobb angle $\geq 25^\circ$ [Cobb angles]	--	7% (2/87) [26° , 30°]	--
Steinbok et al,¹⁶⁸ 2005, 4.3-year follow-up			
Cobb angle, mean \pm SD	6.6° \pm 6.2°	12.5° \pm 6.2°	5.9°
Proportion with Cobb angle $\geq 10^\circ$	--	55% (57/104)	--
Proportion with Cobb angle $\geq 35^\circ$	--	6% (6/104)	--
Proportion worsening $\geq 10^\circ$	--	25% (26)	--
Proportion improving $\geq 10^\circ$	--	4.8% (5) improved	--
Turi et al,¹⁷² 2000, 5.3-year follow-up			
Cobb angle, mean (range)	--	36° (10° to 79°)	--
Proportion with Cobb angle $\geq 10^\circ$ [Cobb angles]	6% (3/47) [42° , 85° , 45°]	32% (15/47)	26%

Radiologic Measure	Pre-operative	Post-operative	Pre- to Post-operative Change
Li et al,¹⁶⁹ 2008, 6.3-year follow-up			
Proportion with scoliosis	2% (1/61)	--	--
Kim et al,¹⁵⁵ 2002, 4.0 years			
Subset (laminectomy L2 to S), proportion with scoliosis	--	9% (5/58)	--
Peter et al,¹⁷⁵ 1990, 4.5-year follow-up^a			
Cobb angle (range)	--	(10° to 60°)	--
Proportion with Cobb angle ≥ 10°	0/28	16% (9/55)	16%
Ambulatory Spastic Cerebral Palsy Cohorts			
Langerak et al,¹⁷³ 2009, 21.4 years^a			
Proportion with Cobb angle ≥ 10°	0/28	57% (17/30)	57%
Proportion with Cobb angle > 30° [Cobb angles]	0/28	7% (2/30) [35°, 35°]	7%
Funk et al,⁶⁷ 2016, 2.8-year follow-up			
Proportion with Cobb angle ≥ 10°	--	10% (7/72)	--
Proportion with Cobb angle ≥ 20°	0/72	3% (1/72)	3%
Spiegel et al,¹⁷¹ 2004, 4.2-year follow-up			
Cobb angle, mean ± SD (range)	-	16° ± 4° (11° to 24°)	--
Proportion with Cobb angle ≥ 10°	0/74	17% (13/74)	17%
Johnson et al,¹⁷⁰ 2004, 8.6-year follow-up			
Cobb angle, mean (range)	7° (0 to 19°)	11° (0 to 26°)	4°
Proportion with Cobb angle ≥ 10°	21% (7/34)	44% (15/34)	23%
Proportion with Cobb angle ≥ 20°	0/34	9% (3/34)	9%
Van Schie et al,¹³⁶ 2011, 6-year follow-up			
Proportion with Cobb angle ≥ 20° [Cobb angle]	--	3% (1/33) [21°]	3%

Abbreviations: GMFCS, Gross Motor Function Classification System; L, lordosis; K, kyphosis; N, number; NR, not reported; S, scoliosis; SD, standard deviation; SP, spondylolysis/spondylolisthesis.

^aPeter and Langerak report on the same cohort at different follow-up periods.

Kyphosis and Lordosis Spinal Deformities

The normal spine has some degree of lordosis (swayback or arched back) at the lumbar and the cervical regions of the spine with various degrees of kyphosis (humpback) throughout the thoracic region, essentially allowing an equal distribution of forces across the spinal column.¹⁹³ Deformity in the sagittal plane can present as either exaggerations (hyperkyphosis or hyperlordosis) or deficiencies (hypokyphosis or hypolordosis) of these normal curves. Kyphosis

and lordosis are measured radiologically by various angle measurements (Cobb angle) of the vertebrae in various (standing or sitting) positions.¹⁷⁰

Normal ranges for kyphosis have been reported by several investigators: Probst-Proctor et al¹⁹⁴ reported a mean kyphosis of 27° (range 2° to 33°) and Boesker et al¹⁹⁵ reported a mean kyphosis of 33° (range 17° to 51°). Hyperkyphosis has been defined in some studies as curves with Cobb angles > 50° or > 55°. ^{170,171} Hyperkyphotic curves of 80° in the thoracic area or 60° to 70° in the thoracolumbar region are considered severe and are noticeable deformities. ^{181,196} Kyphotic deformities of this extent can create wide-ranging debilities resulting in severe pain, respiratory, digestive, and cardiovascular irregularities and neurologic compromise. ^{181,196}

Normal ranges for lordosis of lumbar curves have also been reported: Probst-Proctor et al¹⁹⁴ reported lordosis ranged from -22° to -54° and Voutsinas et al¹⁹⁷ reported it ranged from -47° to -65°. Hyperlordotic curves of -70° or above are more likely to cause bowel or bladder dysfunction, deteriorating balance, or back or leg pain and to require surgical correction. ¹⁹⁶

Children with cerebral palsy are more likely to develop sagittal deformities, either hyperkyphosis or hyperlordosis, if they have scoliosis but these deformities can occur independently. ¹⁹⁸ In a large surgical series of cerebral palsy patients, median age was 16 years, range 8 to 19 years, at surgery for spinal deformities; 7.6% (26/340) either for hyperlordosis (n = 8) or hyperkyphosis (n = 14). ¹⁹⁶ Of the 26 patients having surgery, all for seating problems or back pain, 23 had spastic quadriplegia. The mean pre-operative hypokyphotic curve of 94° was corrected to a mean 36°, and the mean pre-operative hyperlordotic curve of -92° was corrected to -49°. Six of the deformities (5 hyperkyphotic) were considered to be rapidly progressing: increasing ≥ 20° within 6 months.

In another surgical series, severe hyperlordosis was corrected in 27 cases (23 cerebral palsy and all spastic quadriplegia) in most cases to improve sitting position. ¹⁹⁸ In seven cases, the hyperlordosis was associated with thoracolumbar scoliosis. Pre-operative hyperlordosis ranged from -79° to -132° from L1 and S1, and post-operative lordosis was reduced by 8° to 77°. All children had improved function.

Children with spastic cerebral palsy who are untreated with dorsal rhizotomy can have pre-existing sagittal spine deformity, particularly when their neuromuscular abnormalities affect either the spine or the hips. Deformities, particularly lordosis, can be secondary to hip flexion contractures that can affect spinal sagittal alignment. The laminotomy procedure (removal of bone to access the spinal nerves) of dorsal rhizotomy can itself cause or affect the progression of kyphotic or lordotic curves.

In this review, the results of the nine studies investigating kyphosis and lordosis in children with spastic cerebral palsy undergoing dorsal rhizotomy are outlined in Table 15. The studies involve cohorts of both ambulating and nonambulating children (6 cohorts) or only ambulating children (3 cohorts).

The post-operative change in mean kyphotic curve involved minor changes in Cobb angles in the four studies reporting it: 3.0°, ¹⁷¹ 1.6°, ¹⁶⁷ 3.4°, ¹⁶⁸ and 5.0°. ¹⁷⁰ In the mixed ambulating and ambulating cohorts, post-operative prevalence of hyperkyphotic curves based on undefined Cobb angles was 2% (1/61), ¹⁶⁹ 5% (3/58), ¹⁵⁵ or 9% (4/47). ¹⁷² Another estimated post-operative prevalence of hyperkyphosis for a mixed ambulating group, based on a threshold Cobb angle ≥ 40°, was 5% (3/60), ¹⁷⁵ 12% (6/50), ¹⁶⁷ and 41% (18/42). ¹⁶⁸ In the two ambulatory cohorts, ^{170,171} post-operative prevalence of kyphosis, both based on higher Cobb angles, was 1.2% (1/77) ¹⁷¹

for an angle above 50° and was 9% (3/34)¹⁷⁰ for a Cobb angle above 55°. In one study¹⁷¹ the post-operative prevalence in an ambulatory cohort of both hypokyphosis (Cobb < 20°) and hyperkyphosis (Cobb > 55°) was reported to be 16% (12/77) and 1.3% (1/77), respectively.

The mean post-operative lordotic change was greater than that for kyphosis. In the four studies reporting them in mixed ambulatory/nonambulatory populations, mean changes in Cobb angles for lordosis were 10.4°,¹⁶⁸ 11.4°,¹⁶⁹ and 13.5°.¹⁶⁷ In ambulators, the post-operative change in mean lordotic curve was 1°¹⁷¹ and 35°.¹⁷⁰

In the mixed ambulating and ambulating cohorts, the post-operative prevalence of hyperlordotic curves, based on an undefined Cobb angle, was 15% (7/47)¹⁷² and 16% (10/61).¹⁶⁹ Based on a Cobb angle less than -54°, the post-operative hyperlordotic prevalence was 32% (17/53)¹⁶⁷ and 21% (10/47).¹⁶⁸ Post-operative prevalence of hyperlordosis in ambulators (defined as Cobb angle > -64°) was 27% (7/26)¹⁷¹ and (defined as Cobb angle > -60°) 59% (17/34).¹⁷⁰ The increase in prevalence of hyperlordosis from baseline to post-operative assessment was estimated in three studies^{169,172,173} (two based on an undefined Cobb angle) as an 11%¹⁷² and 14%¹⁶⁹ increase. A 19° increase in prevalence was based on Cobb angles for either hyperlordosis (> -54°) or hypolordosis (< -23°).¹⁷³

Table 15. Spinal Sagittal Deformities of Kyphosis or Lordosis After Dorsal Rhizotomy

Radiologic Measure	Pre-operative	Post-operative	Pre-op to Post-op Change
Ambulatory and Nonambulatory Spastic Cerebral Palsy Cohorts			
Golan et al,¹⁶⁷ 2007, 4.7-year follow-up			
Thoracic kyphosis, mean Cobb angle	28.0°	29.6°	1.6°
Thoracic kyphosis, proportion with Cobb AP angle > 39.5°	--	12% (6/50)	--
Thoracic kyphosis, proportion with Cobb AP angle < 11.5°	0/10	4% (2/50)	--
Lumbar lordosis, mean Cobb angle (range)	-34.2° (-16° to -48°)	-47.7° (-14° to -87°)	13.5°
Lumbar lordosis, proportion with Cobb AP angle > -22.5°	--	6% (3/53)	--
Lumbar lordosis, proportion with Cobb AP angle < -54°	--	32% (17/53)	--
Steinbok et al,¹⁶⁸ 2005, 3.6-year follow-up			
Thoracic kyphosis, mean Cobb angle ± SD	34.8° ± 13.3°	38.2° ± 13°	3.4°
Thoracic kyphosis, proportion with Cobb AP angle > 39.5°	--	41% (18/42)	--
Lumbar lordosis, mean Cobb angle ± SD	-30.8° ± 13.5°	-41.2° ± 15.2°	10.4
Lumbar lordosis, proportion with Cobb AP angle < -54°	--	21% (10/47)	--
Lumbar lordosis, proportion worsening (Cobb angle ≥ 15°) (range)	--	36% (17/47) (-17° to -62°)	--
Turi et al,¹⁷² 2000, 5.3-year follow-up			
Proportion with thoracic kyphosis	4% (2/47)	9% (4/47)	5%
Proportion with lumbar lordosis	4% (2/47)	15% (7/47)	11%
Lumbar lordosis, Cobb angles (pre-op individual hyperlordotic angles, post-op mean [range])	-55°, -58°	-79° (-60° to -90°)	--
Li et al,¹⁶⁹ 2008 6.3-year follow-up			

Radiologic Measure	Pre-operative	Post-operative	Pre-op to Post-op Change
Proportion with thoracic kyphosis	0	2% (1/61)	--
Lumbar lordosis, mean Cobb angle \pm SD (range)	17.9° \pm 4.5°	29.3° \pm 4.6° (23° to 44°)	11.4°
Proportion with lumbar lordosis	2% (1/61)	16% (10/61)	14%
Kim et al,¹⁵⁵ 2002, 4.0-year follow-up			
Proportion with thoracic kyphosis	--	5% (3/58)	--
Proportion with lumbar lordosis	--	0	--
Peter et al,¹⁷⁵ 1990, 4.5-year follow-up			
Thoracic kyphosis, proportion with Cobb angle > 40°	--	5% (3/60)	--
Lumbar lordosis, proportion with Cobb angle > -50°	--	7% (4/57)	--
Ambulatory Spastic Cerebral Palsy Cohorts			
Langerak et al,¹⁷³ 2009, 21.4-year follow-up			
Thoracic kyphosis, proportion with Cobb angle < 20° and > 40°	0/28	7% (2/28)	7%
Lumbar lordosis, proportion with Cobb angle < -23° and < -54°	21% (6/28)	40% (12/30)	19%
Spiegel et al,¹⁷¹ 2004, 4.2-year follow-up			
Thoracic kyphosis, mean Cobb angle \pm SD	36° \pm 9°	33° \pm 11°	3°
Thoracic kyphosis, proportion with Cobb AP angle < 20° and > 50°	9% (7/74) (5 hyperkyphotic, 2 hypokyphotic)	17% (13/77) (1 hyperkyphotic, 12 hypokyphotic)	8%
Lumbar lordosis, mean Cobb angle \pm SD	26° \pm 22°	27° \pm 23°	1°
Lumbar hyperlordosis, proportion with Cobb angle < -64°	1% (1/74)	27% (7/26)	--
Johnson et al,¹⁷⁰ 2004, 8.6-year follow-up			
Thoracic kyphosis, mean Cobb angle \pm SD	36° (20° to 55°)	41° (20° to 65°)	5%
Thoracic kyphosis, proportion with Cobb AP angle > 55°	0	9% (3/34)	9%
Lumbar lordosis, mean Cobb angle	-19°	-54°	35°
Lumbar hyperlordosis, proportion with Cobb angle < -60°	0	50% (17/34)	50%

Abbreviations: AP, anterior-posterior; NR, not reported; SD, standard deviation.

Spondylolysis and Spondylolisthesis

Dorsal rhizotomy involves opening the lamina (laminoplasty) in the lumbar region to allow access to the spinal nerves, removal of the lamina and spinous process (laminectomy) and—in some procedures—replacement or reinsertion of the lamina (laminotomy). Replacement of the lamina, particularly when multiple vertebrae levels have been opened for the procedure, is intended to minimize any destabilizing effects in the spine.

Spondylolisthesis, defined as the movement or slippage of one vertebrae over another toward the anterior or posterior, usually occurs in the lumbar region.¹⁹⁹ It has classically been divided into five subtypes²⁰⁰: Type I, dysplastic or congenital; Type II, isthmic or spondylolytic involving a lesion in the pars interarticularis; Type III, degenerative; Type IV, traumatic (involving fractures);

Type V, pathologic (involving lesions to pars or pedicle due to generalized bone disease). Only Types I and II are thought to occur in children and adolescents.²⁰¹ The severity of the condition has been rated according to the degree of slippage (i.e., degree that a vertebral body has slipped forward over the body beneath it): Grade 0 (no slippage), Grade 1 (1%–25%), Grade 2 (25%–50%), Grade 3 (50%–75%), or Grade 4 (75%–100%). Slippage has been described as low grade when it is below 50% and high grade when it is above 50%.¹⁹⁹ Most cases, particularly those involving low-grade slippage, are asymptomatic.²⁰² Surgery has been recommended for slippage of more than 50% in a growing child with or without symptoms.^{185,201-203}

Spondylolysis with or without spondylolisthesis is infrequently seen in children. The incidence of spondylolysis in a large population of unselected children at 6 years of age was 4.4% (22/500). This rate increased to 6% at adulthood. No unilateral spondylolysis resulted in spondylolisthesis. Although slippage occurred with bilateral spondylolysis, the progression of the slip was slow, and no subject in 45 years of follow-up developed slippage greater than 40%.²⁰⁴ In children with cerebral palsy, prevalence of spondylolysis appears to depend on ambulation. In a cohort of 143 nonambulators of average age 27 years, most of whom had cerebral palsy, no incidents of spondylolysis or spondylolisthesis in the lumbar spine were observed.²⁰⁵

Spondylolysis was reported in two studies^{206,207} involving ambulatory spastic cerebral palsy cohorts. In the Hennrikus et al²⁰⁷ study involving 47 spastic cerebral palsy patients (with quadriplegia, hemiplegia, and diplegia) of average age 16 years, two cases (4%) were reported on the basis of standing radiographs: one had asymptomatic Grade 1 spondylolisthesis and the other a spondylolysis without spondylolisthesis.

In the Harada et al²⁰⁶ study, these radiologic features in an unselected population of cerebral palsy patients with spastic diplegia and an average age of 20 years were compared with those in a control population of people undergoing routine physical examination. On the basis of standing radiographs, spondylolysis was reported for 21% (18/84), unilateral in 8 cases and bilateral in 10 cases, compared with 6% (3/50) in the control population.

Spondylolysis was also associated with higher degrees of lumbar lordosis, occurring in 29% of those with lumbar lordosis above -50° and in 7% of those with lordosis below -50° . Unlike the control population, the angle of lordosis also significantly increased with age in the patients with cerebral palsy. Spondylolisthesis, based on a slippage above 5%, was reported for 4% (3/84). The three patients were aged 20, 21, and 34 years with slippage of 3 mm, 4 mm, and 3 mm with corresponding angles of lordosis of -77° , -59° , and -59° . Osteoarthritis of the L5/S1 facet joints was noted in 67% of patients aged 20 years or older. Low back pain was commonly reported (44%) in the study group and was related to increasing age and degree of lordosis.

In this evidence review, nine studies^{136,155,167,169-174} evaluating radiologic spondylolysis or spondylolisthesis in children with spastic cerebral palsy undergoing dorsal rhizotomy are detailed in Table 16. The radiologic features were examined over an average 5-year follow-up—except for a cohort study¹⁷³ with 21 years of follow-up. Only three studies¹⁷⁰⁻¹⁷² reported pre-operative values, two for spondylolisthesis (0 and 6%) and one for spondylolysis (3%). Post-operative prevalence was reported either separately or jointly for spondylolysis or spondylolisthesis. In the Peter et al¹⁷⁴ study, the 19% (19/99) combined post-operative prevalence of spondylolysis or spondylolisthesis in their population was significantly higher than the 1% (1/100) of a sex- and race-matched cohort with untreated cerebral palsy.

Two studies,^{170,171} both involving ambulatory patients with spastic cerebral palsy, reported an increased post-operative prevalence of spondylolisthesis after dorsal rhizotomy. Most events involved low-grade slippage in the 4-year and 8-year study follow-up. In the Spiegel et al¹⁷¹ study, there was a 12% increase (0 to 9 events in 77 children) over baseline in prevalence of spondylolisthesis in the 4-year follow-up. In the Johnson et al¹⁷⁰ study, there was an 18% increase (2 to 8 events in 34 children) in prevalence in the 8-year follow-up. In both studies there was an association with lumbar lordosis. In the Johnson et al¹⁷⁰ study, three of the eight cases of spondylolisthesis were associated with hyperlordosis. In the Spiegel et al¹⁷¹ study, the pre-operative mean lordotic angle was $-41^\circ \pm 18^\circ$ in cases with spondylolisthesis and $-24^\circ \pm 22^\circ$ in cases without spondylolisthesis.

Table 16. Spondylolisthesis and Spondylolysis After Dorsal Rhizotomy

Radiologic Outcome Measure	Pre-operative	Post-operative ^a
Ambulatory and Nonambulatory Spastic Cerebral Palsy Cohorts		
Golan et al,¹⁶⁷ 2007, 4.7-year follow-up		
Proportion with spondylolisthesis at L5–S1	--	19% (18/94) Grade 1 (n = 14), Grade 2 (n = 4)
Proportion with post-operative spondylolisthesis and spondylolysis at L5 pars interarticularis	--	(11/18)
Proportion with post-operative spondylolisthesis and pre-operative spondylolisthesis at L5–S1	--	(4/18) Grade 1 (n = 4)
Proportion with post-operative spondylolisthesis and pre-operative spondylolisthesis and spondylolysis at L5–S1	--	(3/18)
Turi et al,¹⁷² 2000, 5.3-year follow-up		
Proportion with spondylolisthesis at L4–L5	0	2% (1/47) Grade 1 (n = 1)
Li et al,¹⁶⁹ 2008, 6.3-year follow-up		
Proportion with spondylolysis	3% (2/61)	--
Proportion with spondylolysis and spondylolisthesis	--	7% (4/61), Grade 1 (n=4)
Kim et al,¹⁵⁵ 2002, 4.0-year follow-up		
Proportion with spondylolysis	--	3% (2/58)
Peter et al,¹⁷⁴ 1993, follow-up not reported		
Proportion with spondylolysis or spondylolisthesis	--	19% (19/99)
Proportion with spondylolysis or spondylolisthesis in sex- and race-matched same-site untreated cerebral palsy cohort	--	1% (1/100)
Ambulatory Spastic Cerebral Palsy Cohorts		
Langerak et al,¹⁷³ 2009, 21.4-year follow-up		
Proportion with spondylolisthesis	--	3% (1/30) Grade 1, also with spondylolysis
Proportion with spondylolysis	--	37% (11/30), 5 bilateral
Spiegel et al,¹⁷¹ 2004, 4.2-year follow-up		
Proportion with spondylolisthesis	0/74	12% (9/77) Grade 1 (n = 8), Grade 2 (n = 1)
Van Schie et al,¹³⁶ 2011, 6-year follow-up		
Proportion with spondylolysis and spondylolisthesis	--	4% (1/27)
Johnson et al,¹⁷⁰ 2004, 8.6-year follow-up		

Proportion with spondylolisthesis	6% (2/34) Grade 1 (n = 2)	24% (8/33) Grade 1 (n = 8)
Proportion of spondylolisthesis with hyperlordosis	--	3/8

^aGrade 1 slippage = 1%–25%; Grade 2 slippage = 25%–50%.

CONCLUSIONS. Spinal Deformity and Instability After Dorsal Rhizotomy

- The prevalence of pre-operative scoliosis (Cobb angle $\geq 10^\circ$) varied greatly (0% to 31%) among study groups, and post-operative prevalence (defined as a Cobb angle of 20° or more) ranged between 3% and 9%.
- The risk of developing scoliosis was 27% for Cobb angle $\geq 10^\circ$ and 3% for Cobb angle $\geq 20^\circ$.
- Scoliosis remained radiologically stable ($\pm 10^\circ$) in most cases—5% improved, 70% had no change, and 25% worsened.
- Pre- and post-operative hyperlordosis was more common than hyperkyphosis. Post-operative mean lordotic angles increased over baseline in all reports, and prevalence of post-operative hyperlordosis ($> -55^\circ$) ranged from 21% to 50%.
- Prevalence of pre-operative spondylolisthesis was uncommon (0 to 5%) or unreported, and post-operative prevalence ranged from 2% to 27%; most events involved minor or Grade 1 slippage.
- Overall evidence: 15 studies; *modified* McHarm 5 moderate, 10 low quality (Table A7)

Hip Instability

All children with cerebral palsy have been shown to be at risk for developing progressive hip displacement or subluxation.⁴⁷ Children born with cerebral palsy usually have normal hips that deteriorate gradually because of various neuromuscular imbalances, such as hyperactive adductors and flexors and relatively weak extensors in their hips.²⁰⁸ The indirect relationship between increasing risk of hip displacement (subluxation or dislocation) with decreasing function in cerebral palsy patients has been detailed in several large population studies.²⁰⁹⁻²¹³

In a Norwegian population-based study of cerebral palsy by Terjesen et al,²¹³ 26% (89/335) of children had hip displacement (subluxation in 22% and dislocation in 4%) at their initial radiograph performed (on average) at 3 years of age. Children with spastic quadriplegia more frequently (81%; 56/69) presented with hip displacement than those with spastic diplegia (22%; 20/89). All dislocations occurred in quadriplegic children. The percentage of normal hips decreased with increasing motor disability from 99% in GMFM Level I to 28% in GMFM Level V.

In a UK study of patients with bilateral cerebral palsy by Scrutton et al,^{211,212} by age 5, 40% (71 children) of the children and 31% (110 hips) of the hips either had hip subluxation, hip surgery, or a hip orthotic device. In an American study by Lonstein et al,²¹⁰ prevalence of subluxated or dislocated hips in patients with cerebral palsy of average age 10.8 years varied directly with mobility status: independent ambulator (11%; 8/76); dependent ambulator (23%; 10/43); independent sitter (44%; 18/41), and dependent sitter (57%; 173/304). In the Australian study by Cooke et al²⁰⁹ involving children diagnosed with cerebral palsy before age 3 years, dislocation occurred in 10% of patients (47 patients) and 9% of hips (72 hips).

Progression of hip displacement in children with cerebral palsy is estimated by changes in the migration percentage score (MPS), a reliable radiographic measure^{214,215} of the adequacy of coverage of the femoral head by the acetabular roof. The MPS, first developed by Reimers,²¹⁶ defined cut-off MPSs of 33% for subluxation and 90% for dislocation. Recent guidance on hip morphology has defined a 6-grade classification scheme based on several radiologic parameters: Grade I, normal (MPS < 10%); Grade II, near normal (MPS 10%–15%); Grade III, dysplastic (MPS 16%–30%); Grade IV, subluxated (MPS 31%–99%); Grade V, dislocated (MPS ≥ 100%); Grade VI, salvage surgery, loss of hip joint.²¹⁷

Hips at risk have been defined as those with MPSs between 33% and 39%. A change of 10% or more of the MPS was considered to be clinically relevant.²¹⁸ Hips in children with cerebral palsy with severe subluxation (MPS 60%–90%) were reported to progress to dislocation at any age—the rate of progression, however, can vary from months to years.²¹⁹

The rate of annual progression of hip displacement in children with spastic cerebral palsy has been evaluated radiographically in two studies.^{213,220} Terjesen et al^{213,221} documented a mean annual hip MPS of 4% (range –9% to 49%) in a population-based longitudinal survey of 335 newly diagnosed unoperated children with cerebral palsy followed radiologically until 8 years of age. Annual hip MPSs were found to vary significantly with motor disability: GMFCS Level I (0.2 ± 3.7), GMFCS Level II (1.2 ± 3.2), GMFCS Level III (1.3 ± 3.1), GMFCS Level IV (3.9 ± 4.8), GMFCS Level V (9.5 ± 9.4). The rate of annual migration progression also varied between the types of cerebral palsy from 1.8% ± 4.2 in diplegia to 9.2% ± 8.4 in quadriplegia ($P < .001$). All of the children who developed hip dislocation were nonambulators.

In the Park et al²²⁰ study, 197 patients were prospectively followed radiologically for a mean duration of 2 years before any hip surgery. At initial examination, the mean MPS was 35% ±

24.6 and varied significantly by GMFCS level—27% for Levels I–III, 46% for Level IV, and 52% for Level V. The annual MPSs, adjusted for sex, duration of surveillance, and laterality for each motor disability level, increased for all GMFCS levels ($P < .001$). Annual mean score increases were 0.3% for GMFCS Levels I to III, 1.9% for GMFCS Level IV, and 6.2% for GMFCS Level V. There were significant annual changes with follow-up for the neck-shaft angle, another radiologic measure of hip displacement, but only for GMFCS Level IV. The other two radiologic measures evaluated, the acetabular index and pelvic obliquity, did not change significantly with annual follow-up. There were no sex differences for any of the radiologic follow-up measures.

Untreated hip displacement in children with cerebral palsy can increase morbidity. Indications for hip surgery vary but include interventions to prevent dislocation, improve perineal nursing care, decrease fractures of the femoral shaft, improve sitting balance, or decrease pain.²²² Hip pain in children with cerebral palsy and displaced hips has been reported inconsistently^{223,224} and has not always been the main indication for surgery.²²²

In this evidence review, eight studies^{155,225-231} evaluated how dorsal rhizotomy affected hip development in children with spastic cerebral palsy (Table 17). Six studies^{155,226-228,230,231} reported radiologic measures of hip instability using the MPS. The Hicdonmez et al²²⁹ study evaluated hip instability with the Wiberg centre-edge (CE) angle of the femoral head. The Chan et al study²²⁵ used both MPSs and CE angles. In the latter, MPSs were used for those younger than 8 years of age and the CE angle was used for those 8 years or older to define subluxation.²²⁵ Defined boundaries for the CE angle were normal (CE $> 20^\circ$), subluxation (CE 0 to 20°), severe subluxation (CE -40° to 0), and dislocation (CE $> -40^\circ$). The Floeter et al²²⁶ study used three radiologic measures to evaluate hip instability: the MPS, Hilgenreiner acetabular dysplasia index, and femoral anteversion angle.

Hip instability after dorsal rhizotomy was first reported by Greene et al²²⁷ in a small series of six cases, five of which involved quadriplegic spastic cerebral palsy. In that study, rapidly progressing hip instability was noted in six of the approximately 70 cases undergoing dorsal rhizotomy at the institute. The overall incidence of hip instability in their cases, either pre- or post-operatively, could not be evaluated, as radiographs were missing for many patients. However the cases did demonstrate that rapid progression of hip instability (an increase in the MPS ranging from 16% to 30%) could occur within a 1-year follow-up. It was noted however, that all patients already had severely subluxated hips before surgery.

Pre-operative hip instability status, evaluated by MPSs, varied significantly across the surgical clinical cohorts. In the two studies^{97,230} involving diplegic cerebral palsy, the proportion of patients with subluxated hips (MP $\geq 33\%$) was 58% (23/40) and 16% (22/134). In the study evaluating a large group of spastic diplegic and quadriplegic children by Hicdonmez et al,²²⁹ the proportion of subluxated hips (CE angle $< 20^\circ$) was 72% (118/164) and the proportion of patients with subluxated hips was 82% (67/82). The study by Heim et al²²⁸ involving spastic quadriplegic patients reported a pre-operative hip instability rate (MP $> 33\%$) of 29% (26/90).

Three studies^{155,226,229} reported pre- and post-operative mean MPSs or CE angles at various follow-up periods: 18 months (range 12–29 months),²²⁶ 4 years (range 1–9 years),¹⁵⁵ and 4 years (range 1–12 years).²²⁹ Two of the studies^{155,229} involved large cohorts or both spastic diplegia and quadriplegia. All three studies reported reduced lateral migration of hip displacement measured by both the migration percentage and CE angle change scores. Improved scores, however, were not clinically relevant with less than 10% change in MPSs and less than 5° in CE angle. The mean score changes reflected radiologic stability (no change) over the follow-up periods. The study by Floeter et al,²²⁶ involving only ambulatory diplegia, reported

significantly improved mean score changes for three radiologic measures. However the mean follow-up after dorsal rhizotomy was 18 months, suggesting that the changes support short-term radiologic hip stability.

How dorsal rhizotomy affects hip development was also evaluated by the proportion of children experiencing changes (improving, unchanging, or worsening) in their hip migration status with radiologic follow-up. Clinically relevant changes were defined as MPS changes of 10% or more or as CE angle changes above 5°. The proportion of hips with worsening MPSs ranged from 2% to 18% in various study populations: 7% (10/134²³⁰), 11% (10/90²²⁸), and 18% (33/188¹⁵⁵). The proportion of hips with worsening CE angle scores was 18% (29/164).²²⁹ Most patients in all studies, however, remained radiologically stable (unchanging or improving) after dorsal rhizotomy.

Two risks for hip development after dorsal rhizotomy were also evaluated in the studies—risk of developing hip abnormalities in those without prior hip abnormalities and risk of progression (or improving) of hip migration in those having subluxation before surgery. Three studies reported the proportion of hips with worsening (> 10%) MPSs among those with pre-operative hip MPSs that were normal (< 33% MPS): 12% (2/17²²⁵), 12% (13/112),²³⁰ and 14% (9/64).²²⁸ When pre-operative migration percentage cut-off scores of 0 to 14% were considered normal hips, the percentage of hips worsening was 19% (5/27) for diplegic patients²³⁰ and 56% (5/9) for quadriplegic patients.²²⁸

Two studies^{228,230} evaluated progression or worsening (> 10% MP change) of hips in those with pre-operative MPSs of 33% or more (representing subluxation). In the Park et al²³⁰ study of spastic diplegia, none of 22 hips worsened. In the Heim et al²²⁸ study of quadriplegia, 4% (1/26) of hips worsened. Most were radiologically stable in follow-up. Both of these studies also evaluated changes in hip displacement by pre-operative mobility status. In the study with 67 spastic diplegic patients, the proportion of hips worsening was 4% in independent ambulators, 9% in dependent ambulators, and 13% in nonambulators.²³⁰ In the study with 45 quadriplegic patients from the same site, the proportion of hips worsening was 4% in dependent ambulators (96% unchanged) and 14% in nonambulators (73% unchanged, 13% improved).

The study by Hicdonmez et al²²⁹ was the only one to evaluate hip progression by GMFCS level. On the basis of a 5° increase in CE angle, 10% (6/59) of hips in cases at GMFCS Levels II and III worsened and 37% of hips in cases at GMFCS Levels IV and V worsened with follow-up.

Dislocated hips were rarely reported, pre- or post-operatively. Only one case of hip dislocation was reported in the study by Hicdonmez et al.²²⁹ In a 10-year radiologic follow-up study by Silva et al,²³¹ incidence of hip dislocation and hip containment procedures after dorsal rhizotomy was reported for a study population of 69 children with spastic quadriplegia. Before surgery, none of the patients had dislocated hips and no orthopedic procedures, soft or hard, were performed. Final radiographs were available for 47 patients (94 hips) and medical/surgical records for 69 patients (138 hips). Hip dislocation was defined as a complete loss of contact between the articulating surfaces of the femoral head and the acetabulum; the cohort had a hip dislocation rate of 10.6% (10/94). Overall 36% (25/69) of children had either hip dislocation or hip containment surgery. The secondary hip containment rate was 25% (35/138).

Table 17. Radiographic Evaluation of Hip Instability After Dorsal Rhizotomy

Radiographic Outcome Measure	Pre-operative Hip Status	Post-operative Hip Status
Greene et al,²²⁷ 1991, United States, mean 1.0-year follow-up		
Individual MPS by laterality (worsened MPS, change > 10%)	Case 1. Left 23% Case 2. Left 41% Case 3. Left 41% Case 4. Left 16% Case 4. Right 12% Case 5. Left 27% Case 6. Right 26%	Case 1. Left 53% Case 2. Left 72% Case 3. Left 60% Case 4. Left 37% Case 4. Right 35% Case 5. Left 46% Case 6. Right 47%
Ambulatory and Nonambulatory Spastic Cerebral Palsy Cohorts		
Chan et al,²²⁵ 2013, Hong Kong, 5.3-year follow-up		
Proportion of patients with normal hips, MPS < 33% for those < 8 years old or CE angle \geq 20° for those > 8 years old	Normal 43% (17/40)	Normal 88% (15/17); 2 deteriorated, n = 2; 1 with poor coverage and 1 with both hips subluxated
Proportion of subluxated hips, MPS \geq 33%	58% (23/40): unilateral (n = 10), bilateral (n = 13); 20 followed up	Subluxated n = 18 Normal n = 2
Kim et al,¹⁵⁵ 2002, China, 4.0-year follow-up (range 1–9 years)		
MPS: mean (range)	29% (11%–78%)	27% (10%–68%)
Proportion of hips with MPS change over pre-operative, improve or worsen > 10%	--	Improved n = 60 (32%) Unchanged n = 95 (51%) Worse n = 33 (18%)
Hicdonmez et al,²²⁹ 2005, Canada, 4.0-year follow-up (range 1–12 years)		
CE angle, mean right-side CE angle, (mean left-side CE angle)	14.1° (13.6°)	17.2° (15.1°)
Proportion of patients with subluxation, CE angle < 20°	82% (67/82)	--
Proportion of hips with subluxation, CE angle: Normal > 20° Subluxation 0 to 20° Severe subluxation -40° to 0	Normal 28% (n = 46) Subluxation 67% (n = 110) Severe subluxation 4% (n = 7) Dislocated 0.6% (n = 1)	Normal 45% (n = 73) Subluxation 49% (n = 80) Severe subluxation 6% (n = 10) Dislocated 0.6% (n = 1)
Proportion of hips with change, improved as CE angle increased > 5° or worsened as CE angle decreased > 5°	--	Stable 44% (n = 72) Improved 38% (n = 63) Worsened 18% (n = 29)
Symmetry of hip joint change (> 10° difference from right to left)	--	Symmetric in 39 patients: both improved (n = 15), both unchanged (n = 19), both worse (n = 5) Asymmetric in 43 patients: improved in one, unchanged in other (n = 24); improved in one, worsened in other (n = 9); unchanged in one, worsened in other (n = 10)
For GMFCS Levels II and III, proportion of patients with change, improved as CE angle increased > 5° or worsened as CE angle decreased > 5°	--	Improved 34% (20/59) Unchanged 56% (33/59) Worsened 10% (6/59)
For GMFCS Levels IV and V, proportion of patients with change, improved as CE angle increased > 5° or worsened as CE angle decreased > 5°	--	Improved 60% (26/43) Worsened 37% (16/43)

Radiographic Outcome Measure	Pre-operative Hip Status	Post-operative Hip Status
Park et al,²³⁰ 1994, United States, 15- to 46-month follow-up (n = 47) and 6- to 10-month follow-up (n = 20)		
Change in hip MPS, improved or worsened > 10%	2–11 years 134 hips (67 patients)	Improved 17% (n = 23) Unchanged 75% (n = 101) Worsened 8% (n = 10)
Change in hip MPS by age group, improved or worsened > 10%	2–4 years 76 hips (38 patients)	Improved 21% (n = 16) Unchanged 68% (n = 52) Worsened 11% (n = 8)
	5–11 years 58 hips (29 patients)	Improved 12% (n = 7) Unchanged 85% (n = 49) Worsened 3% (n = 2)
Change in hip MPS from pre-operative hip MPS, improved or worsened > 10%	MPS = 0–14% 27 hips	Improved 0 Unchanged 81% (n = 22) Worsened 19% (n = 5)
	MPS = 15%–33% 85 hips	Improved 13% (n = 11) Unchanged 81% (n = 56) Worsened 11% (n = 8)
	MPS > 33% 22 hips	Improved 55% (n = 12) Unchanged 45% (n = 10) Worsened 0 (n = 0)
Change in hip MPS by pre-operative ambulation status, improved or worsened > 10%	Nonambulatory 16 hips (8 patients)	Improved or unchanged 87%, worsened 13%
	Dependent ambulatory 66 hips (33 patients)	Improved or unchanged 91%, worsened 9%
	Independent ambulatory 52 hips (26 patients)	Improved 96%, worsened 4%
Heim et al,²²⁸ 1995, United States, 20-month follow-up (range 7–50 months)		
Overall change in hip MPS, improved or worsened > 10%	90 hips (45 patients)	Improved 9% (n = 8) Unchanged 80% (n = 72) Worsened 11% (n = 10)
Change in hip MPS by age group, improved or worsened > 10%	2–4 years 46 hips (23 patients)	Improved 13% (n = 6) Unchanged 72% (n = 33) Worsened 15% (n = 7)
	5–9 years 44 hips (22 patients)	Improved 4% (n = 2) Unchanged 89% (n = 39) Worsened 7% (n = 3)
Change in hip MPS by pre-operative hip MPS, improved or worsened > 10%	MPS < 15% 9 hips	Unchanged 44% (n = 4) Worsened 56% (n = 5)
	MPS 15%–33% 55 hips	Improved 11% (n = 6) Unchanged 82% (n = 45) Worsened 7% (n = 4)
	MPS > 33% 26 hips	Improved 8% (n = 2) Unchanged 88% (n = 23) Worsened 14% (n = 1)
Change in hip MPS by pre-operative ambulation status: improved or worsened > 10%	Nonambulatory 32 patients	Improved 13% (n = 2) Unchanged 73% (n = 23) Worsened 4% (n = 1)
	Dependent ambulatory 12 patients	Unchanged 96% (n = 11) Worsened 4% (n = 1)
Silva et al,²³¹ 2012, United States, 10.8-year follow-up		

Radiographic Outcome Measure	Pre-operative Hip Status	Post-operative Hip Status
Radiographic hip dislocation rate as complete loss of contact between articular surfaces of the femoral head and acetabulum; radiographs were available for 47 (94 hips) of 69 patients	Nonambulatory 94 hips (69 patients)	Hip dislocation rate 11% (10/94)
Ambulatory Spastic Cerebral Palsy Cohorts		
Floeter et al,²²⁶ 2014, Germany, 66 hips (39 patients), 18-month follow-up (range 12–9 months)		
MPS, mean ± SD	23.6% ± 9.6	21.1% ± 9.0 2.5% ± 4.9 ($P < .001$) ^a
Hilgenreiner Acetabular Index for dysplasia, mean ± SD	18.9 ± 5.8	17.3 ± 6.3 1.6 ± 3.7 ($P = .001$) ^a
Femoral anteversion angle, mean ± SD	40.8° ± 7.6	38.1° ± 6.5 2.7 ± 5.3 ($P < .001$) ^a
GMFM-88, mean ± SD	80.0 ± 16.1	84.4 ± 14.2 4.7 ± 6.7 ($P < .001$) ^a

Abbreviation: CE angle, centre-edge angle; GMFM, Gross motor function measure; GMFM, Gross Motor Function Measure; MPS, migration percentage score; SD, standard deviation.

^aMean pre-post difference ± SD.

CONCLUSIONS: Hip Instability

- Prevalence of pre-operative hip subluxation, measured by migration percentage scores, varied significantly among various cerebral palsy cohorts undergoing dorsal rhizotomy.
- The rate of clinically relevant decline in hip stability (defined as a hip migration percentage change of more than 10%) varied from 2% to 18% of hips among children in surgical cohorts.
- In patients without prior hip subluxation, risk of developing hip subluxation was < 5% and risk of progression of hip subluxation was 14%.
- The risk of worsening hip stability after dorsal rhizotomy was higher in children with greater motor disability defined either by GMFCS level or by mobility status.
- Hip dislocation was rarely reported after dorsal rhizotomy, usually involving an average follow-up of less than 5 years. A 10-year follow-up study reported an 11% hip dislocation rate in nonambulatory spastic diplegia.
- For most children undergoing dorsal rhizotomy, hips remained radiologically stable during follow-up periods.
- Overall evidence: 8 studies, *modified* McHarm 4 moderate, 4 low quality (Table A7)

Discussion

Several randomized trials comparing the effectiveness of dorsal rhizotomy and physical therapy to physical therapy alone found that children with spastic cerebral palsy treated with surgery had significantly greater gains in their gross motor function than those treated with physical therapy alone. These trials, however, involved a limited assessment of short-term function, mainly of children with spastic diplegia. They also involved early and diverse surgical protocols. Given their small sample sizes, none of the trials had intended, or were able, to evaluate safety of the surgery or any long-term adverse effects.

Since these trials, a substantial number of observational cohort studies from pediatric centres in many countries have evaluated dorsal rhizotomy in diverse groups of cerebral palsy patients. Many of these studies reported broader and more comprehensive outcome measures, including motor function, functional independence, and caregiver assistance, that were based on reliable and validated outcome measures. Several observations about short- and long-term function can be made from these studies.

Results from earlier randomized trials on improved gross motor function after dorsal rhizotomy were replicated in these studies from diverse real-world clinical settings. Because patient groups in these studies were more diverse, more is known about the effects of the surgery for patients varying by their functional status (i.e., degree of ambulation) or by their degree of disability (i.e., diplegia or quadriplegia). In general motor gains involving higher-order skills, such as running or jumping, were more often achieved by children who were less disabled and had some ambulation abilities. However, more disabled children (those with spastic quadriplegia or nonambulatory) often had gains in motor function as well, but in lower-order skills such as crawling, sitting, or standing. Long-term follow-up studies also showed that there were continuous improvements in motor function for most children up to 5 years following surgery, after which gains often stabilized.

Functional independence, or children's ability to perform or improve their own basic activities of daily living, is particularly important for families potentially faced with life-long supportive care of their children. Gains in functional independence showed the same trend in as in gross motor function, although functional gains for many children continued beyond 5-year follow-up. Follow-up in several studies continued for 10 and 15 years and longer. Although results were consistent, longer-term progress is less certain because groups followed for long periods usually involved few children from a few centres.

Caregiver assistance was also assessed after dorsal rhizotomy and some improvements were reported, based both on informal parents' or caregivers' reports and on formal assessment tools. This outcome was less frequently evaluated and, when evaluated, usually involved short-term assessments of up to 2 years. The need for caregiver assistance was also not evaluated as a separate outcome but as a part of a multidimensional outcome scale that incorporated both functional independence and assistance. Many measurement outcome tools are specifically developed to assess caregiver assistance or burden (particularly for disabled children). It is uncertain which tool would be appropriate to measure the degree of caregiver assistance provided and be responsive to measuring changes over time in this disabled population. Given the importance of this outcome for children with spastic cerebral palsy and their families and caregivers, greater efforts are needed to more fully evaluate changes in what might be the primary outcome measure or treatment objective for some families.

Most studies, in addition to reporting improvements in either motor function or functional independence, reported extensive variability in individual responses to dorsal rhizotomy, even within different functional levels. Many factors could account for this variability. Cerebral palsy itself is an extremely heterogeneous condition. Evaluating neuromuscular abnormalities other than spasticity that might be affecting motor function and limiting treatment responsiveness, might not be straightforward and can be clinically challenging.

Physical therapy after surgery is essential for children to recover from surgery and in many cases to regain strength and learn how to use muscles that often were previously not used. Post-operative rehabilitation and practices may vary across centres and over time.

There are other limitations in this evidence. In general, most of these studies were observational cohort studies without comparison groups, making it difficult to evaluate change or improvement that might have occurred without treatment of spasticity by dorsal rhizotomy. Results from cohort studies were strengthened by findings not only of statistically significant improvement in gross motor function but also of clinically relevant improvements; these changes were also consistent across many centres. Findings in these studies were also consistent with those from the randomized clinical trials. Also expected developmental milestones in gross motor function could be compared with milestones in untreated children with cerebral palsy, given a large population-based control population of children with cerebral palsy treated with usual care. Those studies found that children with cerebral palsy on average achieve most of their motor function by age 5 years. More disabled children, those who are nonambulatory or have spastic quadriplegia, often reach their maximum potential at much younger ages—2 or 3 years of age. Children who receive dorsal rhizotomy often continue to make clinically relevant gains in motor function well beyond these ages. Children in the surgical cohorts also achieved on average higher total motor scores than those in the population control group.

Treatment satisfaction, an important consideration for major surgery with potentially life-altering effects, was not routinely evaluated in the cohort studies. When it was evaluated, who the respondents were and the format of the interviews were not always clear. Nevertheless, most respondents interviewed either at short-term or longer-term follow-up have been satisfied with the effects of their child's surgery and have said they would recommend it to others. However treatment outcomes with which some parents expressed satisfaction—improved mood, appearance, confidence, and task performance—were often not formally evaluated as outcomes in clinical studies. Parents also described difficulties after surgery (children were fatigued and lost strength) and during rehabilitation (which was stressful, was time-consuming, and interfered with care for their other children). Several investigators reported that treatment satisfaction depended on whether initial discussions with parents were focused on realistic goal setting and expectations for improvement that they might have for their child.

Many observational cohort studies also had safety as a primary study objective and, because of their larger sample sizes, were able to provide reliable estimates, particularly of the operative complications of dorsal rhizotomy.

Many of the safety studies evaluated the risks of causing or exacerbating existing bony abnormalities in either the spine or the hip after dorsal rhizotomy. Although radiologic investigations were performed in these studies, examinations were not always routine in study populations, or protocols were inconsistent or unreported. Evaluating risks of any of these abnormalities after surgery is made difficult by their common occurrence (particularly scoliosis or hip instability) in untreated children with spastic cerebral palsy. Children offered dorsal rhizotomy usually have more disability and greater muscle spasticity, making them even more

likely to have pre-existing bony abnormalities. The unbalanced muscular forces from spasticity in the spine or the hip are thought to be important contributing factors to these bony deformities or instabilities. Studies evaluating the rate of progression of these abnormalities after dorsal rhizotomy usually documented small and clinically irrelevant changes after surgery. However, study follow-up in most cases, often up to 5 years, was generally too short to conclude that stabilization of abnormalities would persist at longer-term follow-up. Nevertheless the high prevalence of bony spinal and hip abnormalities in children with spastic cerebral palsy make it advisable to document the status of these conditions before dorsal rhizotomy and to follow up closely after surgery for new or worsening orthopedic abnormalities.

Conclusions

Lumbrosacral dorsal rhizotomy and physical rehabilitation effectively reduces lower limb spasticity in carefully selected children with spastic cerebral palsy and improves their gross motor function and functional independence. Children with spastic diplegia and cerebral palsy treated with dorsal rhizotomy and physical therapy also have greater gains in gross motor function than those treated only with physical therapy (moderate GRADE).

In the surgical cohort studies, the degree and type of motor improvement varied by presenting levels of motor disability. Generally, less disabled children with some degree of mobility achieved gains in higher-order motor skills, and more disabled children without mobility achieved gains in lower-order skills. Functional independence and need for caregiver assistance also improved after surgery for spastic diplegic cerebral palsy at short-term (moderate GRADE) and long-term (low GRADE) follow-up. Information on caregiver assistance, however was limited, particularly for children with spastic quadriplegia.

Major operative complications for dorsal rhizotomy occurred infrequently (modified McHarm moderate). Children with cerebral palsy and untreated spasticity are at high risk for developing spine and hip bony abnormalities. Radiologic follow-up after dorsal rhizotomy suggests that most children are stable for at least these orthopedic abnormalities.

Parents generally reported improvements in their children after dorsal rhizotomy and expressed satisfaction with the treatment. Treatment satisfaction, however, was infrequently reported, and definitions and measurements in the surgical cohort studies varied. Treatment outcomes for which parents expressed satisfaction were not often formally evaluated as outcomes in clinical studies, and parents described difficulties with both surgery and post-operative rehabilitation.

ECONOMIC EVIDENCE REVIEW

Objectives

The primary objective was to review the published literature on the cost-effectiveness and cost-utility of lumbosacral dorsal rhizotomy in patients with spastic cerebral palsy.

Methods

Sources

We performed an economic literature search on December 3, 2015, using Ovid MEDLINE, Ovid MEDLINE In-Process, Ovid EMBASE, Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects (DARE), Centre for Reviews and Dissemination (CRD) Health Technology Assessment Database, and National Health Service (NHS) Economic Evaluation Database, for studies published from inception to December 3, 2015. We also reviewed reference lists of included economic literature for any additional relevant studies not identified through the systematic search. Appendix 1 provides details of the search strategy.

Literature Screening

We based our search terms on those used in the clinical evidence review of this report and applied economic filters to the search results. A single reviewer reviewed titles and abstracts and, for those studies meeting the inclusion and exclusion criteria, we obtained full-text articles.

Inclusion Criteria

- English-language full-text publications
- Studies published between 1946 and December 3, 2015
- Studies in patients with spastic cerebral palsy
- Studies comparing dorsal rhizotomy with usual care (physiotherapy alone)
- Cost-utility, cost-effectiveness, or cost-benefit analyses

Exclusion Criteria

- Studies evaluating treatment for nonspastic cerebral palsy

Outcomes of Interest

- Costs, cost per quality-adjusted life-year, cost per clinical effect

Data Extraction

We extracted relevant data on the following:

- source (i.e., name, location, year)
- population and comparator
- interventions
- outcomes (i.e., health outcomes, costs, cost-effectiveness)

Study Applicability Appraisal

We determined the usefulness of each identified study for decision-making by applying a modified methodology checklist for economic evaluations originally developed by the National Institute for Health and Care Excellence (NICE) in the United Kingdom. The original checklist is used to inform development of clinical guidelines by NICE.²³² We retained questions from the NICE checklist related to study applicability and modified the wording of the questions to remove references to guidelines and to make it Ontario specific.

Results

Literature Search

The database search yielded 45 citations published between 1946 and December 3, 2015 (with duplicates removed). We excluded a total of 28 articles based on article type and information in the title and abstract. We then obtained the full texts of 17 potentially relevant articles for further assessment. After screening and reviewing the full text, we found no studies met the inclusion criteria for our review. Figure 2 presents the flow diagram for the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA).

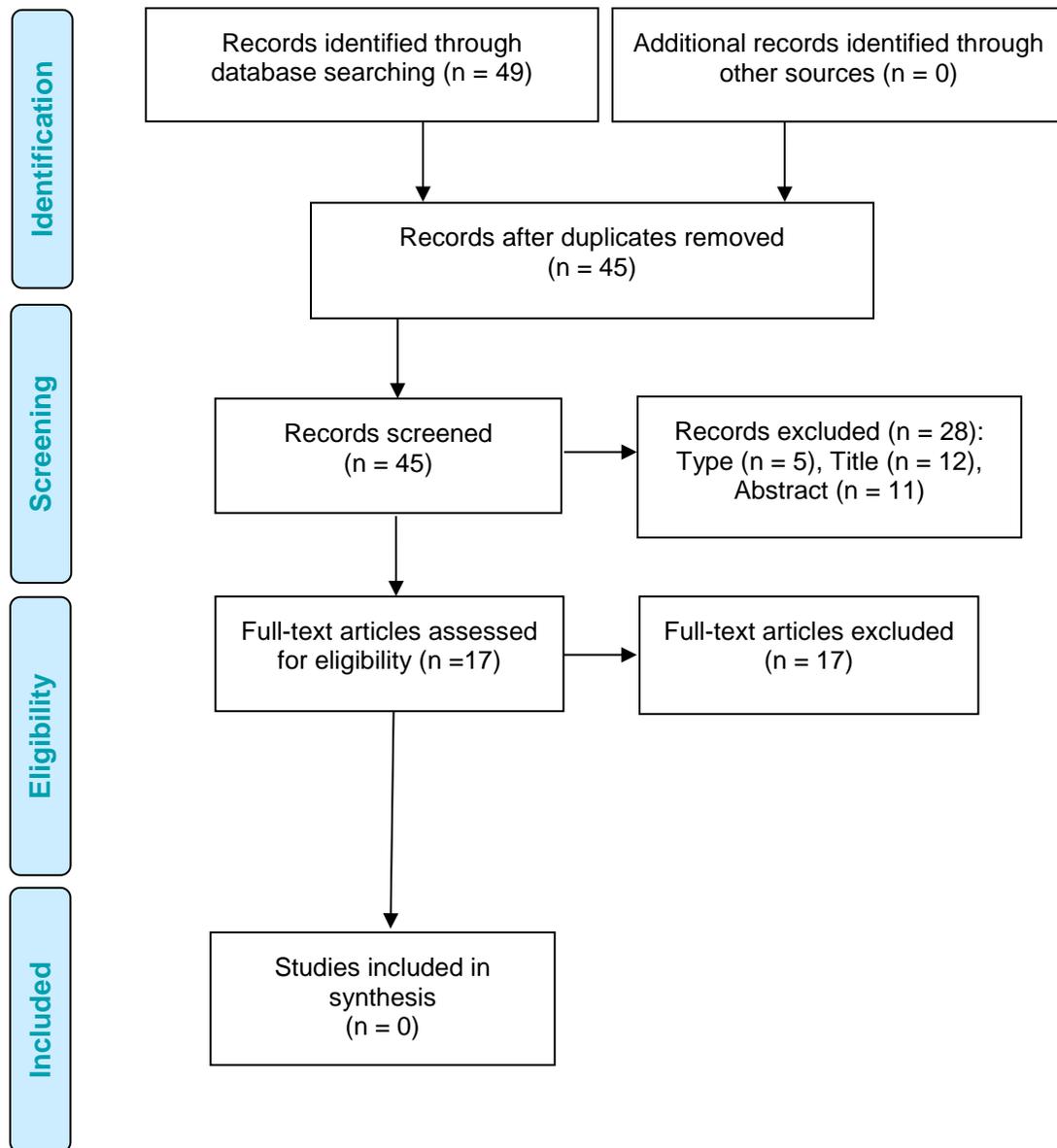


Figure 2: PRISMA Flow Diagram — Economic Evidence Review

Source: Adapted from Moher et al, 2009.²³³

Discussion

A search for economic evaluations of dorsal rhizotomy for children with spastic cerebral palsy yielded no studies that examined the cost effectiveness of this intervention. Therefore the cost-effectiveness of dorsal rhizotomy for spastic cerebral palsy is unknown.

Two reports were found that examined the cost of administering dorsal rhizotomy to treat spastic cerebral palsy. A cost-comparison study from the British Columbia Children's Hospital²³⁴ looked at the relative cost (in 1993 Canadian dollars) of selective functional posterior rhizotomy

and continuous intrathecal baclofen therapy in treatment of severe spastic quadriplegia related to cerebral palsy. In this analysis, nine children who received intrathecal baclofen were matched with ten patients who received selective functional posterior rhizotomy. The analysis showed that intrathecal baclofen was more expensive than dorsal rhizotomy. The first year after successful implantation of an intrathecal baclofen pump averaged \$64,100 versus \$16,900 after dorsal rhizotomy surgery. Most costs for both treatments were attributable to hospitalization cost, accounting for 72% among patients receiving an intrathecal baclofen implant and 65% among patients receiving dorsal rhizotomy therapy.

The second study is a 2006 Australian Medical Services Advisory Committee report²³⁵ that evaluated the financial implications of dorsal rhizotomy in Australia. The estimated total procedure cost was \$71,500 Australian dollars (AUD) per interstate patient and \$68,000 AUD per local patient who received dorsal rhizotomy. Costs were broken down into each component cost: assessment and selection (\$3,836), pre-operative care (\$280), surgical episode (\$10,567), postoperative care for the first 6 weeks (\$36,683), postadmission follow-up to 12 months (\$3,446), and indirect and other costs (\$16,667). Both cost analyses, although published more than a decade ago, provide some context for costs attributable to pre-operative screening, surgical procedures, and postoperative care for patients receiving dorsal rhizotomy.

Conclusion

The cost effectiveness of dorsal rhizotomy for treatment of spastic cerebral palsy is unknown.

BUDGET IMPACT ANALYSIS

We conducted a budget impact analysis from the perspective of the Ontario Ministry of Health and Long-Term Care to estimate the annual cost burden for dorsal rhizotomy. All costs are reported in 2016 Canadian dollars.

Objectives

The objective of this study is to determine the budget impact of funding dorsal rhizotomy for treatment of spasticity in children with cerebral palsy.

Methods

Target Population

According to Ontario administrative data, dorsal rhizotomy has not been performed in the province since 2001. The latest figures on the total number of patients who received dorsal rhizotomy in Ontario were between the years 1996 and 2001. A total of nine procedures was reported in 2001 but the number has decreased since then. Only one case in 2000 and two cases in 2001 have been treated with dorsal rhizotomy.

Table 18: Number of Patients Receiving Dorsal Rhizotomy Between 1996 and 2001 in Ontario

Year	Number of Patients Receiving Dorsal Rhizotomy ^a
1996	9
1997	7
1998	5
1999	4
2000	1
2001	2

^aData provided by Ontario IntelliHEALTH.

The decrease in volumes over time was due to several factors including the uncertainty of the clinical efficacy of this treatment according to expert opinion (content expert, personal communication, September 7, 2016). Assuming that the uncertainty of the treatment efficacy is resolved, all patients eligible for dorsal rhizotomy (nine total) are assumed to be treated per year. Prevalence of cerebral palsy among children in Ontario aged 5 years and under has been consistent over time with a rate of between 2.8 to 3.1 per 1,000 persons.²³⁶ Thus, as a base case we assumed that a total of nine children would receive dorsal rhizotomy yearly and that this number would be consistent each year.

An estimated 16 patients will be assessed at a tertiary referral centre to determine eligibility for dorsal rhizotomy yearly. If nine receive dorsal rhizotomy, seven patients a year would be assessed but would not continue on to receive this surgery.

Resource and costs

Some studies have examined how dorsal rhizotomy affects immediate health care utilization, but no studies have observed long-term health care use. No studies have explored how changes in GMFM, PEDI and WeeFIM scores (clinical outcomes of interest in dorsal rhizotomy randomized controlled trials [RCTs]) affect health care use. As a result, the long-term implications of dorsal rhizotomy treatment on health care resources are unknown. We limited costs in the budget impact analysis to the immediate treatment costs, including the costs for the surgical procedure and postsurgical rehabilitation.

Dorsal rhizotomy assessment, surgical procedure and postsurgical rehabilitation

According to experts, all patients who could benefit from dorsal rhizotomy are assessed by a team of clinicians for surgical eligibility (developmental pediatrician, neurosurgeon, orthopedic surgeon, physiotherapist, and kinesiologist) (content expert, personal communication, September 7, 2016). This team composition is similar to that in a clinical trial of dorsal rhizotomy from Holland Bloorview Kids Rehabilitation Hospital in Ontario by Wright and colleagues.²³⁷ The University of British Columbia's rhizotomy program,²³⁸ Seattle Children's hospital,²³⁹ and Cardinal Glennon Children's Hospital in St. Louis²⁴⁰ have similar members in their assessment teams. Physician costs were extracted from the Ontario Schedule of Benefits.²⁴¹ We calculated costs of health care professionals using the average Ontario hourly wage for physiotherapists and kinesiologists according to data from the Canadian government job bank.²⁴² In Ontario, the median wage for physiotherapists is \$36.06 per hour, \$23.32 per hour for kinesiologists. Unit costs are presented in Table 19. During the assessment period, children received magnetic resonance imaging (MRI) of the head and x-ray examination of the back and hips. Physiotherapists and kinesiologists would spend 2.5 hours and 5 hours, respectively, on assessment of surgical eligibility (content expert, personal communication, September 7, 2016).

Table 19: Dorsal Rhizotomy Eligibility Assessment Unit Costs

Variable	Unit Cost (\$)	Reference
Neurosurgeon (A935 Special surgical consultation)	160	Ontario Physician Schedule of Benefits ²⁴¹
Orthopedic surgeon (A935 Special surgical consultation)	160	Ontario Physician Schedule of Benefits ²⁴¹
Physiotherapist	36.06	Canadian Government job bank ²⁴²
Kinesiologist	23.32	Canadian Government job bank ²⁴²
Head MRI (X421 multislice sequence)	73.35	Ontario Physician Schedule of Benefits ²⁴¹
Hip x-ray (X037 pelvis and/or hip, 2 views)	36.95	Ontario Physician Schedule of Benefits ²⁴¹
Back x-ray (X205 lumbar or lumbosacral spine 4-5 views)	44.15	Ontario Physician Schedule of Benefits ²⁴¹

Abbreviations: MRI, magnetic resonance imaging.

The hospitalization cost for dorsal rhizotomy surgery was extracted from Ontario administrative data and from the Ontario Physician Schedule of Benefits.²⁴¹ Prior hospitalizations for dorsal rhizotomy were identified by collecting all acute hospital stays where "division of intraspinal nerve root" was a reported procedure (Canadian Classification of Diagnostic, Therapeutic and Surgical Procedures code 1610²⁴³) for children with "infantile cerebral palsy" aged 1 to 9 years

(ICD-9 diagnostic code 343.0 to 343.9²⁴⁴) between the years 1996 and 2001. A total of 28 cases were identified during this period. The average resource intensity weight, defined as the standardized measure estimating health care resource utilization by patients with the same disease profile versus average hospitalization, was calculated to be 2.2326. Using the most recent cost per typical hospital stay in Ontario of \$5,407,²⁴⁵ we estimated the average cost of hospitalization for dorsal rhizotomy as \$12,100 ($\$5,407 \times 2.2326$). Physician costs for surgical procedures were identified from the Ontario Physician Schedule of Benefits as posterior spinal decompression - lumbar (includes laminectomy) and intradural rhizotomy (Table 20).

Table 20: Dorsal Rhizotomy Physician Unit Costs

Variable	Unit Cost (\$)	Reference
Laminectomy (N511 posterior spinal decompression – lumbar, one level, unilateral)		
Primary physician	800.70	Ontario Physician Schedule of Benefits ²⁴¹
Physician assistant	96.32	Ontario Physician Schedule of Benefits ²⁴¹
Anesthetist	225.15	Ontario Physician Schedule of Benefits ²⁴¹
Intradural rhizotomy (N577 sympathectomy, any number of roots)		
Primary physician	714.00	Ontario Physician Schedule of Benefits ²⁴¹
Physician assistant	96.32	Ontario Physician Schedule of Benefits ²⁴¹
Anesthetist	150.01	Ontario Physician Schedule of Benefits ²⁴¹

There is currently no standard protocol for postsurgical rehabilitation. In our analysis, we used an Ontario-based rehabilitation protocol provided to us by a consultant. This protocol consists of inpatient rehabilitation for up to 3 months (content expert, personal communication, August 22, 2016), followed by outpatient rehabilitation with a physiotherapist twice weekly for 4 to 6 months and then with a physiotherapist one to two times weekly from 7 to 12 months. Postsurgical inpatient rehabilitation costs consisted of the hospital's per diem costs. For the 2015/2016 fiscal year, the per diem cost was estimated to be \$1,475.²⁴⁶ The per diem cost was assumed to include the cost of allied health professionals. The total cost for the inpatient rehabilitation is \$134,225 ($\$1,475 \times 91$ days). The cost of outpatient rehabilitation included physiotherapist time. For 4- to 6-month outpatient rehabilitation, we assumed individuals received two 1-hour sessions weekly for 13 weeks (52 weeks/4). From 7 to 12 months, outpatient rehabilitation was assumed to consist of 1.5 1-hour sessions weekly for 26 weeks (52 weeks/2). Our analysis assumed each child would receive 65 hours of physiotherapy (26 hours between 4-6 months, 39 hours between 7-12 months). Using an hourly wage of \$36.06 for physiotherapy, we calculated the total cost of outpatient rehabilitation would be \$2,343.90 ($\36.06×65 hours).

Standard care outpatient rehabilitation

The cost of standard care physiotherapy rehabilitation was also limited to the health care professional cost. Similar to the postsurgical inpatient rehabilitation calculation, the number of hours per week and duration of rehabilitation was based on expert opinion (content expert, personal communication, August 22, 2016). For our analysis, we assumed that standard care outpatient rehabilitation frequency was the same as 7 to 12 months' outpatient rehabilitation after dorsal rhizotomy. In other words, patients received 1.5 hours of physiotherapy weekly. For our analysis, we assumed that the frequency of physiotherapy would remain the same throughout the year. The total health care professional cost for standard care physiotherapy rehabilitation is \$54.09 per week or \$2,812.68 for 52 weeks.

Analysis

The net cost of dorsal rhizotomy per person was calculated by subtracting the total cost of dorsal rhizotomy from the cost of standard care physiotherapy and occupational therapy. The total cost burden of dorsal rhizotomy per year in Ontario was calculated by multiplying the net cost of dorsal rhizotomy per person by the number of cases per year (9). Given that the number of new cases per year is not expected to change and that only immediate costs are considered in our analysis, the annual cost burden is expected to be the same over time. As a result, only an annual estimate of the burden of illness was presented in our analysis.

Sensitivity analysis

In the absence of information about trends in the incidence of cerebral palsy over time, we estimated the number of cases of dorsal rhizotomy if the volume of cases were to increase in proportion to the increase in the population in Ontario since 1996. The population of Ontario has increased 30% in the last 20 years (10,754,000 in 1996²⁴⁷ to 13,792,000 in 2015²⁴⁸). Assuming that the number of dorsal rhizotomy procedures increases proportionally, the number of cases would be 12. The budget impact with 12 cases was calculated as a sensitivity analysis. To evaluate the range of rehabilitation protocols on the total budget impact, various frequencies and number of hours of physiotherapy and occupational therapy were examined. In the second sensitivity analysis, inpatient postsurgical rehabilitation was reduced to 6 weeks to match a protocol observed in an RCT conducted in Ontario many years ago. In the third sensitivity analysis, the rehabilitation protocol for an RCT on dorsal rhizotomy conducted by McLaughlin and colleagues²⁴⁹ was included in the budget impact analysis. In this protocol, both treatment and control arms had the same rehabilitation schedule. So, for this sensitivity analysis, we assumed there would be no additional costs for dorsal rhizotomy after hospital discharge.

Expert Consultation

In August and September 2016, experts were consulted on the use of dorsal rhizotomy for children with spastic cerebral palsy. Members of the consultation included clinical scientists who had conducted research in the topic area and physicians in the specialty areas of pediatrics. The role of the expert advisors was to place the evidence in context and to provide advice on treatment of cerebral palsy with dorsal rhizotomy. However, the statements, conclusions, and views expressed in this report do not necessarily represent the views of the consulted experts.

Results

Base Case

The net cost of dorsal rhizotomy, postsurgical rehabilitation, and physician follow-up for the first year is \$151,470 per person, while the cost of rehabilitation for the comparator (physiotherapy) is \$2,812. The breakdown of this cost is presented in Table 21. The net cost of dorsal rhizotomy is \$148,658 per person.

Table 21: Dorsal Rhizotomy and Standard Care Total Costs per Patient

Variable	Total Cost (\$)
Dorsal rhizotomy	
Pre-surgical assessment	620
Surgical	14,165
Postsurgical inpatient rehabilitation	134,225
Postsurgical outpatient rehabilitation	2,344
Follow-up physician visits	117
Total	151,470
Standard care	
Outpatient rehabilitation	2,813
Total	2,813

To annual net budget impact of dorsal rhizotomy including the cost of all patients assessed but deemed ineligible for this procedure is estimated to be \$1.3 million per year.

Table 22: Net Cost of Dorsal Rhizotomy per Year

Variable	Cost per Patient(\$)	Number of Patients	Total Cost
Patients receiving dorsal rhizotomy	148,657	9	1,337,917 ^a
Patients assessed but ineligible to receive dorsal rhizotomy	620	7	4,338
Total cost			1,342,255

^aNet cost of dorsal rhizotomy less standard care (\$151,470 – \$2,813).

Sensitivity Analysis

If the number of patients eligible to receive this procedure in Ontario were increased to 12 per year, the annual budget impact would increase to \$1.8 million. Reducing the postsurgical inpatient rehabilitation to 6 weeks followed by outpatient rehabilitation would reduce the cost per person to \$76,800 and the annual budget impact to \$696,000. Assuming that the postsurgical rehabilitation protocol for patients receiving dorsal rhizotomy would be the same as the standard rehabilitation protocol, the additional cost of dorsal rhizotomy would be \$14,900 per person. The total annual budget impact would be approximately \$138,500. Further details of these calculations are presented in Appendix 4.

Discussion

The total annual budget impact of dorsal rhizotomy in Ontario is estimated to be \$1.3 million. Even assuming that the number of patients eligible for dorsal rhizotomy increases with the increase in the Ontario general population, the total economic burden would be quite low.

A budget impact analysis of dorsal rhizotomy was conducted by the Medical Services Advisory Committee to the Australian Health Ministers' Advisory Council in 2006. The total cost observed

in this report was \$71,479 Australian dollars. Excluding the cost of gait analysis (not included in our analysis), the total cost was \$67,979 (\$71,479–\$3,500). Converting this result to Canadian dollars using 2006 purchasing power parities for GDP and inflating to 2016 dollars using the Bank of Canada's inflation calculator, the total would be \$68,900 ($\$67,979/1.404 \times 1.207 \times 1.179$). This is much less than the results from our analysis. The difference is likely due to the differences in the postsurgical rehabilitation protocol. In the analysis by the Medical Services Advisory Committee, patients remained in hospital for 6 weeks before returning to the community, while patients received 3 months of inpatient rehabilitation in our analysis. In our sensitivity analysis using a 6-week inpatient rehabilitation protocol, the total cost per person would be \$76,800, which is approximately \$8,000 more than the cost in the Australian analysis.

Our evaluation has several strengths. First, all cost inputs in our analysis were based on Ontario and Canadian sources. As well, the number of cases of dorsal rhizotomy was based on historical cases in Ontario. Second, analysis inputs, such as the postsurgical rehabilitation protocol, have been reviewed and confirmed by clinical experts in Ontario.

Our analysis also has several limitations. First, the estimate of hospitalization costs for dorsal rhizotomy covered the years 1997 to 2001. There have been no new cases since this time. Likely efficiencies in conducting this procedure have improved. For instance, dorsal rhizotomy formerly required multiple laminectomies (removal of the back of several vertebrae in the spine to access the spinal cord at multiple spots). In the last few years, this procedure has evolved to a less invasive single laminectomy technique (removing only a single vertebra). Thus, the results of our analysis could overestimate the true cost of hospitalization for dorsal rhizotomy. Second, several of our cost inputs were not specific to children receiving dorsal rhizotomy. Thus some of the costs might be inaccurate for this population. For instance, we were unable to estimate the cost of postsurgical inpatient rehabilitation specific for dorsal rhizotomy. Instead, our estimates were based on the mean per diem cost for an inpatient stay at the Bloorview Children's rehabilitation hospital. For this reason, our estimate might be inaccurate.

A clinical expert estimates the daily cost of inpatient rehabilitation for children receiving dorsal rhizotomy may be greater than the typical inpatient rehabilitation per diem cost (content expert, personal communication, September 8, 2016). Therefore, our analysis is likely an underestimate. In another example, physiotherapist costs were based on average wages. However, a clinical expert noted that physiotherapists treating patients receiving dorsal rhizotomy are likely to be specialized or experienced and receive a higher than average wage. Third, several immediate costs were not included in our analysis for various reasons. For instance, we did not incorporate the cost of purchasing new equipment for assessment and treatment of children receiving dorsal rhizotomy. Experts expect the one-time cost of equipment to assist in gait analyses to be substantial (content expert, personal communication, September 8, 2016). However, we do not know how the equipment will be funded, in particular whether it would be paid for by public health care or by another ministry. If new equipment is paid for by the Ministry of Health and Long-term Care, costs are underestimated.

Fourth, we could not estimate the downstream costs or savings for patients treated with dorsal rhizotomy. Review of the clinical evidence for this treatment shows statistically significant improvement in measurements of both short-term and long-term clinical outcomes. This improvement could result in downstream cost savings from fewer physiotherapy visits or additional treatments for spasticity. We were unable to quantify the changes in downstream costs because we lack information. Our results could overestimate costs if there are downstream cost savings.

Fifth, some of our sensitivity analyses include a rehabilitation protocol that assumes a similar frequency of outpatient rehabilitation visits between patients receiving dorsal rhizotomy and those receiving standard care. Experts believe this assumption underestimates expected clinical practice (content expert, personal communication, September 21, 2016).

Conclusions

The budget impact of funding dorsal rhizotomy for treatment of spasticity in children with cerebral palsy is \$1.3 million per year. If the number of patients eligible for dorsal rhizotomy were to increase in proportion to the general population, the annual budget impact could be as high as \$1.8 million.

PUBLIC AND PATIENT ENGAGEMENT

Background

Public and patient engagement explores the lived experience of a person with a health condition, including how the condition and its treatment affects the patient, the patient's family or other caregivers, and the patient's personal environment. Public and patient engagement is intended to increase awareness and build appreciation for the needs, priorities, and preferences of the person at the centre of a treatment program. Insights gained through public and patient engagement provide an in-depth picture of lived experience, through an intimate look at the values that underpin the experience.

Lived experience is a unique source of evidence about the personal impact of a health condition and how that condition is managed, including what it is like to navigate the health care system with that condition, and how technologies might or might not make a difference in people's lives. Information shared from lived experience can also identify gaps or limitations in published research (for example, outcome measures that do not reflect what is important to those with lived experience).²⁵⁰⁻²⁵² Additionally, lived experience can provide information or perspectives on the ethical and social value implications of technologies and treatments. Because the needs, priorities, preferences, and values of those with lived experience in Ontario are not often adequately explored by published literature, Health Quality Ontario makes an effort to reach out to, and directly speak with, people who live with the health condition, including those who have experience with the intervention in question.

The impact of cerebral palsy on patients and families was perceived at the outset of this project to have significant bearing on quality of life. To understand what the impact on quality of life truly was, we spoke directly with patients and families of children with cerebral palsy who may or may not have experience with dorsal rhizotomy. Understanding and appreciating their day-to-day functioning and experience of any treatments, including the intervention in question, helps to contextualize the potential value of the intervention from a lived experience perspective.

Methods

Engagement Plan

Engagement as a concept captures a range of efforts used to involve the public and patients in various domains and stages of health technology assessment (HTA) decision-making.²⁵³ Rowe and Frewer outline three types of engagement: communication, consultation, and participation.²⁵⁴ Communication constitutes a one-way transfer of information from the sponsor to the patient, while participation involves the sponsor and patient collaborating through real-time dialogue. Consultation, on the other hand, refers to the sponsor's seeking out and soliciting information (for example, experiential input) from the public, patients, and caregivers affected by the technology or intervention in question.²⁵⁵

The engagement plan for this HTA was consultation. Within this typology, the engagement design focussed on interviews to examine the lived experience of patients and of families of children with cerebral palsy, including those having undergone dorsal rhizotomy.

The qualitative interview was selected as an appropriate method because it allowed Health Quality Ontario (HQO) staff to deeply explore the meaning of central themes in the lived experience of participants. The main task in interviewing is to understand the meaning of what

participants say.²⁵⁶ Interviews are particularly useful for getting the story behind a participant's experiences, which was the objective in this portion of the HTA. The sensitive nature of exploring quality-of-life issues is another reason for using interviews for this project.

Recruitment of Participants

The recruitment strategy for this project pursued an approach called purposive sampling²⁵⁷⁻²⁶⁰ to actively recruit individuals with direct lived experience. Staff of the Patient, Caregiver, and Public Engagement staff contacted patients with cerebral palsy and families (including those with experience of the procedure) through a variety of partner organizations (including acute care and youth rehabilitation centres across the province), provincial cerebral palsy associations, and word of mouth as interview participants reached out to other families after they completed their interview.

Inclusion Criteria

A range of participants was sought, including those having experience with cerebral palsy and dorsal rhizotomy. A range of function in cerebral palsy was sought, as it was assumed that the values, needs, preferences, and priorities of patients and their families would evolve based on the severity of cerebral palsy's effects on mobility and independence. We also sought to speak with patients of various ages and their family members, as we assumed that the stage of development would affect the choices these patients and families faced in terms of treatments and outcomes they preferred. We sought a broad geographic representation, as we further assumed that access would vary from region to region. A final consideration was to speak with patients and their families who had dorsal rhizotomy performed at a variety of centres where procedures would vary.

Exclusion Criteria

No exclusion criteria were set.

Participants

Patient, Caregiver, and Public Engagement staff at HQO spoke to 13 patients with cerebral palsy and their families from across Ontario. Seven of the 13 families had undergone the procedure, five at the Children's Hospital in St. Louis, MO, one at the Shriners Children's Hospital in Montreal, and the other at the Hospital for Sick Children (SickKids) in Toronto. Two of these participants were patients themselves, one having had the procedure, and the other considering the procedure. All participants, whether they had had the procedure or not, were familiar with dorsal rhizotomy. Ages of patients ranged from 15 months to 34 years. Of the 13 patients, 11 were considered "spastic diplegic," while two had less severe hemiplegia.

Interview Approach

At the outset of the interview, Patient, Caregiver, and Public Engagement staff at HQO explained the purpose of the HTA process (including the role and mandate of HQO and the Ontario Health Technology Advisory Committee [OHTAC]), risks to participation, and protection of personal health information. Staff explained these attributes to participants orally and through a letter of information. Written consent was then obtained from participants before commencing the interview. The letter of information and consent form are attached as Appendix 5. Interviews were recorded and transcribed.

Interview questions focussed on how cerebral palsy affects patients' and families' quality of life, experiences with other health interventions used to manage the condition, experiences with the procedure itself, any postoperation rehabilitation that was required, and perceived benefits or limitations of the intervention. The interview guide is attached as Appendix 6.

The interview was semistructured, consisting of a series of open-ended questions. Interviews lasted from approximately 45 to 60 minutes, with questions for the interview based on a list developed by the Health Technology Assessment international's (HTAi's) Patient and Citizen Involvement Group (PCIG) to elicit lived experience specific to how a health technology or intervention affects lived experience and quality of life.²⁶¹

Data Extraction and Analysis

Patient, Caregiver, and Public Engagement staff at HQO selected a modified version of a grounded theory method to analyze transcripts of participant interviews, because it captured themes and allowed elements of the lived experience to be compared among other participants. The inductive nature of grounded theory follows an iterative process of eliciting, documenting, and analyzing responses while simultaneously collecting and analyzing data using a constant comparative approach.^{262,263} Through this approach, staff coded transcripts and compared themes by using NVivo, a qualitative software program that enabled identification and interpretation of patterns in the interview data about the meaning and implications of the lived condition from the perspective of what was important to participants in their daily life with cerebral palsy, before and after dorsal rhizotomy.

Results

Physical and Emotional Experience of Living With Cerebral Palsy

Given the spectrum of severity for the disorder, parents and those with cerebral palsy reported various degrees of burden of the disease. Age of the child as well as developmental progress also influenced level of impact. However, consistent mobility challenges were reported, affecting the child's ability to navigate the environment at home, at school, and in the community.

Even just simple things like when I do diaper changing, because her muscles are so tight, it is hard to get her changed. Everything from dressing, putting pants on, putting socks on, putting shoes on, all those kind of things affect daily living for her.

Parents consistently reported financial burdens associated with caring for their child. Many families by necessity became single-income households. At the same time purchasing equipment, covering travel expenses, and paying for the child's therapies increased stress in the household. Many out-of-pocket costs associated with caring for a child with cerebral palsy were reported. This burden of care was shared by other family members, who could spend less time together and had mutual caregiver duties.

After he was born I've never returned back to work full time. I just can't. With the amount of appointments that he has and the therapies that he requires, my husband and I actually work opposite shifts to each other. So when I'm at work my husband's home, and when I'm at home my husband's at work. So the only day that we actually see each other usually is Sundays.

Several parents reported emotional distress at forcing their children's consistent participation in therapy and making decisions on behalf of their children. They spoke of a need to balance insisting on therapy designed to improve the child's well-being against allowing the child simply to act as a "normal" child.

It's a constant fight of how do I manage being Mommy and caring for her, and at the same time she's looking at me like I'm this mean person that constantly [is] making her do all these things she doesn't want to do because it hurts.

But I know there's lots more out there; ... financially, it's just a huge burden. And [I am] constantly trying to make a balancing act and letting him be a kid as well and do things with his brother and not always having to do therapy after therapy.

Experience of Treatments for Cerebral Palsy

Following the initial diagnosis of cerebral palsy, families consistently reported quick initiation to children's rehabilitation centres and various therapies for cerebral palsy. Parents reported their physicians had treated their children with Botox, serial casting, physiotherapy, occupational therapy, speech therapy, conductive education, and orthopedic surgery at various stages of their development (subject to the level of severity for cerebral palsy).

In all cases of treatment, expectations and goals included decreased spasticity with improved mobility and independence, as well as decreased pain and dependence on others. Parents reported focusing on both short- and long-term goals. Despite the altruistic nature of these goals, parents reported emotional distress and uncertainty in pushing their children to accept these therapies, which could be time-consuming, stressful, or even painful for their children.

Some of the therapies we were looking at were pretty much trying to maintain what we have now, and [to increase] mobility as we go forward.

One of the main goals with therapy is that we want her to feel independent and confident.

We were looking at both short term and long term. Part of it is looking at those therapies as to prevent the condition from being worse, not necessarily making a radical change as to "He's not doing this today and he could do that tomorrow," but part of it was "He's doing this today; we don't want him to not be able to do that tomorrow."

Lots of kids, I think, that go through—that have different types of abilities, ...spend a lot of their childhood in and out of therapies. And it's interesting; in one of the classes I'm taking now, ...older adults with disabilities say one of the biggest regrets is they look back at their childhood and how much therapy they went through.

Access to these therapies varied. Those living closer to the greater Toronto area and other large urban centres had greater access and more resources available. Families who live in urban centres in close proximity to needed services acknowledged their fortunate circumstances. Families who live more rurally commented on the increased burden of travel and of choices that need to be made because of the consistent requirements for cerebral palsy therapies. Cost of therapies was another barrier reported by families, with out-of-pocket expenses, such as travel and purchasing private physiotherapy, mentioned consistently.

You have to travel everywhere, but it's the nature of living here. It's almost like an acceptance. Our pediatrician is a 45-minute drive. Most of our appointments are at SickKids in Toronto, which is a 1.5- to 2-hour drive. So it's just the nature of where you live, right?

Ongoing therapy—I don't know how a lot of parents of ... lower socioeconomic status ... do it. But for middle-class people, I mean, you end up spending a lot of funds on therapy.

Reported results from therapies contained several consistent themes. While Botox was typically helpful, it was also reported as painful, stressful, and of limited duration, with benefits lasting only a few months at most. Several families also reported viewing Botox with suspicion, given the chemical nature of treatment. Physiotherapy was universally viewed as essential and was highly encouraged by physicians and child specialists, but parents reported frustration with lack of insurance coverage, poor access, high cost, and uncertain long-term outcome projections.

They're very strict structures. They're older medical models that don't work. They're not flexible. It's on their schedule, not a child's. And I'd have to go because if I didn't I'd miss it. You don't get the time back.

We had to find a lot of private physiotherapy. So he did get—he only gets about an hour a week through our children's treatment centre and it's Grandview Children's Centre. So they offered him an hour a week of physiotherapy. And that was it.

Much more positive reactions were reported for conductive education, especially for its motivating and inclusive environment. Families stated that this environment—where multiple children can work together, motivate each other, and achieve goals together—led to much greater physical improvement and excitement about therapy for their child. However, this program is available only in the greater Toronto area and thus is of limited use across the province.

She's a kid; we want it to be play based; we want her to be engaged; we don't want it to be repetitive, like the same thing every single day. We were almost attacking the [cerebral palsy] to the point where she wasn't even like a kid any more.

[He] would come home and be so excited for his friends beside him. Because she walked today and that's all he would talk about. They would cheer for each other. And really that group therapy is so motivating for them. And I think that was another aspect that he loved, ...like he loved going. He couldn't wait to go, and he learned songs.

Exploring Dorsal Rhizotomy

Parents described dorsal rhizotomy as never the first option for treating their child's cerebral palsy. Rather, after months, and typically years of their child's undergoing various therapies, parents described feeling as though they were running out of options that provide relief. As part of seeking treatment for their child, parents spoke about coming across a procedure called dorsal rhizotomy. When they learned more, they asked about the risks and benefits, plus what is involved before, during, and after. Why parents explore dorsal rhizotomy was explained through a few different considerations: First, there were concerns that their child's condition would deteriorate coupled with frustration with existing therapies that provided only temporary relief.

Timing was the other major consideration, as the window for undergoing dorsal rhizotomy tends to close as the child ages. Finally, and most important, there continued to be concern over their child's quality of life, from both a pain-management and a mobility perspective.

Despite all of the therapies he was going through, it seemed like we were struggling just to maintain where he was at. There weren't really gains, which was frustrating for him.

After stretching our daughter, [we realized], wow, we're just constantly fighting against the spasticity. It doesn't matter how much physio we do. It might help her at certain times in the day but when it comes down to it, this is going to get worse.

Making the Decision

In terms of learning more about dorsal rhizotomy, even before deciding whether to pursue the surgery or not, many families talked about the desire to understand the procedure as a choice. In order to make this choice, families discussed the desire to become informed.

This should be an informed choice that people make with information that can help them decide. If you're diagnosed with a debilitating or a life-threatening disease, you should have choices. I think you need to have a choice to be able to say, "Well what's the best thing for you?"

Information Used to Make the Decision

Even before contemplating selective dorsal rhizotomy (SDR) as a potential procedure, parents described the strong desire to learn about it as they explored other procedures, in terms of its potential for help or harm as well as the logistical elements of what might be involved. Quickly after pursuing this information, most parents described their frustration in not being able to receive traditional medical expertise. Rather than see this as a complete barrier, parents described other outlets to gain understanding and certainty, the most important of which was to ask other families about their lived experience.

Research helped us make a decision: data and personal stories that focussed on outcomes. We talked to other families who went to the Cerebral Palsy Clinic in St. Louis and heard that Dr. Parks had been doing this surgery for 25 plus years.

We couldn't find information we were satisfied with about SDR. [The SDR procedure] had come onto our radar, but we wanted to know more about it. We needed to learn who had experience with it and who had performed it. We wanted to see someone who had seen more than this one kid this one time who had it, or who had heard things from other people. We just couldn't find anyone who had experience and the level of knowledge that made us comfortable in terms of whether our son was even in the realm of being an appropriate candidate.

We tried talking to doctors and physiotherapists in Ontario. And all of them ... didn't know what SDR was. So basically all of our research was just independent, trying to speak to other parents and other doctors. It was kind of a struggle to find information. I mean who wants to decide about surgery based on Facebook? It seems a little bizarre that this could be my only source of information.

Connecting with other families [is] really, really critical in your decision making.

It was hard to find information in Ontario. There is a huge void of information. [O]bviously nobody's doing the surgery in Ontario now. There was, from my understanding, only one doctor [who] did that type of surgery, but he didn't release very much information on the surgery. And that was the only source of information in Ontario.

Those who had not yet had dorsal rhizotomy, but were considering it, described similar searches for information both from a clinical and lived experience perspective. They found it difficult, if not impossible, to find information to help them make an informed decision.

Medically, I would like to see what the benefits could be, what the pros and the cons are. But I'd also like to see kids [who] are similar to my daughter and [their outcomes] after having that surgery. I feel like it's beneficial for me to know all of the medical aspects, as well as testimonies from other families that have had this done and what the outcomes were for them.

I know a lot about SDR now based on my own research and my own interest, but [no] conventional medical advice ... has been given.

It's hard to get a discussion going because a lot of doctors, old or new in the industry, are hush-hush about it.

Goals for the Procedure

Parents saw goals for dorsal rhizotomy as aligned with the goals of all other therapies in terms of how spasticity affected their child's pain and happiness, mobility, and independence. Parents whose children had severe cerebral palsy described more moderate goals that still affect day-to-day life.

If he's a candidate, maybe he still won't be able to walk, but maybe transfers will be easier. Maybe we can teach him how to get himself to get on the potty one day kind of thing.

Getting him to weight-bear just for a minute while we turn him in the bathroom or something or help get out of bed or things like that and as he gets bigger is really important.

Even if nothing happened, even if our son stayed in the wheelchair, the fact that he didn't have to have that 14-hour orthopedic operation and be 4 months in hospital would have been enough for me.

There are a lot of families that are pushing for increased independence and independent walking. For us, it's more if our son is happy and he could do more things than he could do before, then that's awesome. It's more about his quality of life.

For most, one of the other primary appeals of dorsal rhizotomy was its description as a more permanent solution in their ongoing battle against spasticity.

It's the spasticity that's causing the problem. And you [can] do orthopedic surgeries, you can do hip surgeries, you can do all of that, but all you're doing is ... prolonging the inevitable and increasing a whole lot of pain in the process. Whereas if you can remove that spasticity through a procedure like SDR, you prevent all the long-term complications and aging and pain that people have. You have to be prepared to do a lot of work in the short term though.

We didn't see SDR as a silver bullet. We saw it as a way of helping to manage spasticity that seemed to be longer lasting with more benefit than other short-term, painful interventions.

The flip side to the appeal of a more permanent route to manage spasticity was the awareness that once nerves in the spinal cord were severed, there was no reversal. This weighed heavily on parents' minds as they contemplated the benefits and risks of the procedure.

It's a really hard decision, and it's permanent. You're severing the nerve, and there's no going back from that, which is scary to hear as a parent because if it's the wrong decision, there's no going back.

We're not super gung-ho about SDR because it's final. There's no undoing once you cut; there's no sewing it back together. And so our concern while we're exploring it as a possibility to solve some of the problems that our son has is that it [could] create other problems. So we're very cautiously moving ... down the SDR path.

That being said, most families were not oblivious to the period of rehabilitation required post-surgery.

I know that it was going to be a long road to build our child's strength after the procedure. But when I considered how this procedure would improve [her ability] to gain independence and stop the problems that are going to eventually crop up as [she ages], to us it was worth it. I wasn't naive though; I was fully aware of the amount of hard work that needs to go into it after you have it done.

Financial Considerations

Most decisions about pursuing SDR were inextricably linked to the difficult question of money. After a short period of research, most families came across the St. Louis Children's Hospital. Most families quickly became aware that this out-of-country procedure would come at an out-of-pocket cost. Only some could go to great lengths to raise money privately, most often through fundraisers.

Knowing that I'd have to travel out of Canada, I can't afford to do it financially. I know that there is no government help for that kind of cost, so I'm not in a financial position to be able to have a surgery like that for my daughter.

We started saving because we don't have that kind of money. We're fortunate my husband has a good job, but we sold our house so that we could move our money into other priority areas. Finances are definitely a barrier. Paying for the surgery would swamp us, financially. Who has \$150,000? Nobody has that.

Surgical costs were a primary consideration, but other major considerations were families putting their lives on hold for over a month while they went out of country at a loss of income and covering the physiotherapy required after surgery (only a small portion of which is covered provincially). These factors compounded the financial burden, which raises equity and access as major considerations for this procedure.

Beyond the costs of the surgery, there is also the cost of all your lost time because you're basically taking a month off of doing anything else. And then you come home and then you have the equivalent of that cost for the next year in rehabilitation costs that aren't covered. From a financial standpoint, it's significant.

It's all out-of-pocket—not only the cost of the surgery, but the cost of the rehab—the long-term costs are significant because you're looking at least 2 years of expensive physio after you have SDR. You ask yourself, how can you make that possible?

Selective Dorsal Rhizotomy Procedure

Once children were accepted as candidates, families who pursued dorsal rhizotomy in St. Louis described the medical procedure and its associated experience as fairly straightforward for both

children and their families. Families did not mention any shortage of information about what to expect, not only of the procedure, but also for the rehabilitation period after surgery. The surgery was said to last approximately 3 hours. Children and their families observed improvements shortly afterward. Committing to be out of country at the Children's Hospital for upward of a month was described as likely the most challenging aspect of the journey (becoming much more complicated when other siblings had to accompany their family).

The hospital down there does a pretty good job preparing you for what to expect in terms of ... the process. For families like us that are out of country, they provide ... quite a bit of information. The day before the surgery, you do all of your assessment appointments; the surgeon then goes through; the surgery is the next day. The day of the surgery is fairly straightforward; not too dissimilar to any other type of surgery. After about 3 hours, ...our son came out of the procedure to the recovery room. We went down there for the month.

Rehabilitation After Selective Dorsal Rhizotomy

Upon completing the procedure, families described the need for a lot of intensive rehabilitation right away. Much of the month away in St. Louis was committed to completing a rigorous rehabilitation regimen after the procedure, a regimen that families described as having to follow once home in Ontario, despite the challenges of funded therapy. For families residing outside of larger centres, this complicated access to available therapies.

With SDR when you come home, you need 5 days of physio, so finding a pediatric therapist available for five days a week [was impossible] where I live. We have to travel everywhere, but it's the nature of living here. There would be much more opportunity for therapy and other things if we lived in an urban centre.

Risks and Benefits of Selective Dorsal Rhizotomy After Surgery

All of the families contacted as part of this HTA that pursued dorsal rhizotomy through the Children's Hospital in St. Louis described improvements in the quality of life of their child. These improvements stemmed from noticeable decrease in spasticity, thereby creating improvements in their child's mobility and independence. These improvements led to decreases in caregiving burden, through improvements as simple as enabling their child to be still and to care for some of their own functional needs.

Before the surgery he couldn't stay in a still position without falling out. So just from that standpoint [you could free] up two hands to do something else to whatever you're doing. You can be at the swimming pool, getting changed, and can put him on the bench, and he can sit there for a couple of minutes while you're getting something else out of the swim bag. You couldn't do that before. It was almost night and day in terms of the freedom it gave us in terms of being able to do things with him and his sister. It was a huge difference.

Our son was able to stand up from the floor and walk flat footed for the first time. He could walk backward and up and down the stairs. He wanted to play baseball. This was a big motivation. So now he can catch the ground ball or get in and out of the van without me having to lift him straight from the floor.

Our son thanks us for doing the surgery. I think when he notices it the most is when he's in the bathtub and he doesn't have the restriction of the pain of spasticity. He'll say to us, "I am so glad you got me new legs."

It has actually exceeded our expectations. We went into the surgery not necessarily wanting our son to be an independent walker, but to give him ... more independence. [T]he surgery ... has

opened doors for him. He does things that he couldn't do before—like going up and down the stairs was very challenging for him before. Now when he wants something in his room, he just goes up the stairs and gets what he wants.

Improvements did not come without effort, as families actively continued to pursue therapies that led to functional gains, primarily through physiotherapy at care facilities and at home. Because children having dorsal rhizotomy need 5 days a week of physiotherapy, it can be challenging to find access.

I think we'll continue to see that it's hard work and it's going to take hard work. But I believe if she wouldn't have had this surgery she would have been [regressing]. I think life would have become tougher for her.

A few of the families mentioned their child would still require orthopedic surgeries because of the severity of the cerebral palsy and age of the child who had the procedure.

As we got further into treatment protocol I can remember the physio telling us that a lot of children would need orthopedic surgery 2 and 3 years down the road. Our daughter had SDR at three and a half, but had to have her first round of orthopedic surgery on one side at seven. And then, she had to have the same thing on the other side [when] she was 13.

DISCUSSION

From speaking directly to families of children with cerebral palsy, they face enormous challenges. Physical, emotional, and financial hardships were described by every family interviewed as part of this HTA. Pursuit of therapies and interventions are simply all part of a journey that families take to bring any form of relief and improvement to their child's quality of life. The goals for all therapies and interventions are shared: to decrease spasticity. This spasticity was described as leading to pain, decreased mobility, decreased independence, and significant caregiving needs. These caregiving needs were said to be an immense burden for families, reducing the financial well-being of the family and placing great stress directly on parents and indirectly on other siblings. Trying to decrease spasticity through conventional therapies in Ontario was described as challenging and ultimately unfruitful. Lack of access to therapies, travel time and costs, and expenses of devices and home modification all weigh on families who typically find only temporary relief through conventional therapies.

Pursuit of other options and a more permanent resolution of spasticity symptoms were described as motivation for families' search for information. For a variety of reasons, even sometimes by fluke, this search for information led several families to dorsal rhizotomy. While families quickly learn the basics about the procedure, they often find little comprehensive information on clinical and lived experience, both described as key to making any decision about dorsal rhizotomy. Families described frustration with limited information from their Ontario health care providers, but continued their own research by speaking with other families and connecting directly to clinics in other provinces and in St. Louis at the Children's Hospital.

The decision for a child to undergo dorsal rhizotomy was described as weighing heavily on the minds of families. The permanence of the procedure has appeal after years of challenges associated with temporary interventions, but also raises concern about its irreversibility. After learning about clinical outcomes of the Children's Hospital in St. Louis and about the experience of other families and their children, families described having confidence in their own experience.

The procedure itself was not described as onerous; process and expectations were outlined clearly. Some minor pain was managed by medication, but the procedure was described as like any other surgical procedure. Out-of-pocket expenses for the month-long stay, for the procedure itself, and for intensive post-surgery physiotherapy mean that families are deeply affected by this experience.

The positive outcomes of the procedure were perceived improvements in spasticity. These outcomes were described as almost immediate and increasing in significance over time. These families see improved mobility and independence of their child, as long as they commit to intensive regular physiotherapy. Decreases in caregiving burden were noted, which meant being able to perform simple day-to-day tasks more easily, such as feeding and using toilets, transportation, and play and interaction with siblings and friends. Increased mobility aided children's movement around the house and in the community.

CONCLUSION

Families of children with cerebral palsy spend a lifetime facing complex challenges that require continual searching for treatments to help their children. After numerous attempts to manage spasticity, many families seek a more permanent resolution through dorsal rhizotomy. A range of families of children who undergo the surgery noted improvements in their child's quality of life. Despite successful procedures and improved outcomes, families have faced a lack of medical information upon which to make an informed decision, the enormous financial burdens incurred both at the point of surgery and after surgery, and lack of rehabilitation supports after surgery. Each of these factors made families' experience with dorsal rhizotomy challenging.

ABBREVIATIONS

CE angle	Centre-edge angle
GMFCS	Gross Motor Functional Classification System
GMFM	Gross Motor Function Measure
GMPM	Gross Motor Performance Measure
GRADE	Grading of Recommendations Assessment, Development, and Evaluation
McHarm	McMaster Quality Assessment Scale of Harms
MPS	Migration percentage score
OHTAC	Ontario Health Technology Advisory Committee
PEDI	Pediatric Evaluation of Disability Inventory
QUEST	Quality of Upper Extremity Skills Test
RCT	Randomized controlled trial
SD	Standard deviation
SEP	Somatosensory evoked potentials
TES	Therapeutic electrical stimulation
VMFM	Vancouver Motor Function Measure
WeeFIM	Functional Independence Measure for Children

GLOSSARY

Cost-Effective	Good value for money. The overall benefit of the technique or intervention justifies the cost.
Incremental cost	The extra cost associated with using one test or treatment instead of another.
Quality-adjusted life-year (QALY)	A measurement that takes into account both the number of years gained by a patient from a procedure and the quality of those extra years (ability to function, freedom from pain, etc.). The QALY is commonly used as an outcome measure in cost–utility analyses.
Randomized controlled trial	A type of study in which subjects are assigned randomly into different groups, with one group receiving the treatment under study and the other group(s) receiving a different treatment or a placebo (no treatment) in order to determine the effectiveness of one approach compared with the other.
Sensitivity analysis	Every evaluation contains some degree of uncertainty. Study results can vary depending on the values taken by key parameters. Sensitivity analysis is a method that allows estimates for each parameter to be varied to show the impact on study results. There are various types of sensitivity analyses. Examples include deterministic, probabilistic, and scenario.
Spastic Cerebral Palsy	Cerebral palsy is a condition, usually resulting from brain injury before, during, or after birth, characterized by delayed or abnormal development of muscle control. Spastic cerebral palsy, in which some muscles are subject to uncontrollable spasms, is the most common form of cerebral palsy.
Systematic review	A process to answer a research question by methodically identifying and assessing all available studies that evaluate the specified research question. The systematic review process is designed to be transparent and objective and is aimed at reducing bias in determining the answers to research questions.
Utility	The perceived benefit (value) placed on a treatment by a person or society.

APPENDICES

Appendix 1: Literature Search Strategies

Clinical Evidence Search

Search date: Dec 02, 2015

Databases searched: All Ovid MEDLINE, Embase, Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects, CRD Health Technology Assessment Database, Cochrane Central Register of Controlled Trials, and NHS Economic Evaluation Database

Database: EBM Reviews - Cochrane Central Register of Controlled Trials <October 2015>, EBM Reviews - Cochrane Database of Systematic Reviews <2005 to November 2015>, EBM Reviews - Database of Abstracts of Reviews of Effects <2nd Quarter 2015>, EBM Reviews - Health Technology Assessment <4th Quarter 2015>, EBM Reviews - NHS Economic Evaluation Database <2nd Quarter 2015>, Embase <1980 to 2015 Week 48>, All Ovid MEDLINE(R) <1946 to Present>

Search Strategy:

-
- 1 Cerebral Palsy/ (44893)
 - 2 ((cerebr* adj pals*) or CP).tw. (124687)
 - 3 Muscle Spasticity/ (21666)
 - 4 (spasticit* or (spastic* adj (dipleg* or hemipleg* or tetrap* or tripleg* or quadripleg*)) or little\$1 disease*).tw. (26225)
 - 5 or/1-4 (163989)
 - 6 exp Rhizotomy/ (3110)
 - 7 Spinal Nerve Roots/su [Surgery] (2154)
 - 8 Ganglia, Spinal/su [Surgery] (354)
 - 9 rhizotom*.tw. (3930)
 - 10 or/6-9 (6900)
 - 11 5 and 10 (1319)
 - 12 11 use pmoz,cctr,coch,dare,clhta,cleed (550)
 - 13 cerebral palsy/ (44893)
 - 14 ((cerebr* adj pals*) or CP).tw. (124687)
 - 15 spasticity/ (16900)
 - 16 (spasticit* or (spastic* adj (dipleg* or hemipleg* or tetrap* or tripleg* or quadripleg*)) or little\$1 disease*).tw. (26225)
 - 17 or/13-16 (161915)
 - 18 exp rhizotomy/ (3110)
 - 19 "spinal root"/su [Surgery] (2154)
 - 20 spinal ganglia/su [Surgery] (354)
 - 21 rhizotom*.tw. (3930)
 - 22 or/18-21 (6900)
 - 23 17 and 22 (1292)
 - 24 23 use emez (769)
 - 25 12 or 24 (1319)
 - 26 remove duplicates from 25 (869)
 - 27 limit 26 to english language [Limit not valid in CDSR,DARE; records were retained] (750)
 - 28 27 use pmoz (85)
 - 29 27 use emez (647)
 - 30 27 use cctr (1)
 - 31 27 use coch (4)
 - 32 27 use dare (7)
 - 33 27 use clhta (4)
 - 34 27 use cleed (2)

Economic Evidence Search

Databases searched: Ovid MEDLINE, Ovid MEDLINE In-Process, Ovid EMBASE, Cochrane Central Register of Controlled Trials, Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects (DARE), Centre for Reviews and Dissemination (CRD) Health Technology Assessment Database and National Health Service (NHS) Economic Evaluation Database

Database: EBM Reviews - Cochrane Central Register of Controlled Trials <October 2015>, EBM Reviews - Cochrane Database of Systematic Reviews <2005 to November 2015>, EBM Reviews - Database of Abstracts of Reviews of Effects <2nd Quarter 2015>, EBM Reviews - Health Technology Assessment <4th Quarter 2015>, EBM Reviews - NHS Economic Evaluation Database <2nd Quarter 2015>, Embase <1980 to 2015 Week 48>, All Ovid MEDLINE(R) <1946 to Present>

Search Strategy:

-
- 1 Cerebral Palsy/ (44928)
 - 2 ((cerebr* adj pals*) or CP).tw. (124747)
 - 3 Muscle Spasticity/ (21719)
 - 4 (spasticit* or (spastic* adj (diple* or hemiple* or tetrap* or triple* or quadriple*)) or little\$1 disease*).tw. (26306)
 - 5 or/1-4 (164127)
 - 6 exp Rhizotomy/ (3113)
 - 7 Spinal Nerve Roots/su [Surgery] (2155)
 - 8 Ganglia, Spinal/su [Surgery] (354)
 - 9 rhizotom*.tw. (3934)
 - 10 or/6-9 (6905)
 - 11 5 and 10 (1322)
 - 12 economics/ (250449)
 - 13 economics, medical/ or economics, pharmaceutical/ or exp economics, hospital/ or economics, nursing/ or economics, dental/ (711668)
 - 14 economics.fs. (376433)
 - 15 (econom* or price or prices or pricing or priced or discount* or expenditure* or budget* or pharmacoeconomic* or pharmaco-economic*).tw. (655807)
 - 16 exp "costs and cost analysis"/ (495890)
 - 17 cost*.ti. (224965)
 - 18 cost effective*.tw. (236192)
 - 19 (cost* adj2 (util* or efficacy* or benefit* or minimi* or analy* or saving* or estimate* or allocation or control or sharing or instrument* or technolog*).ab. (147519)
 - 20 models, economic/ (131045)
 - 21 markov chains/ or monte carlo method/ (115943)
 - 22 (decision adj1 (tree* or analy* or model*).tw. (31975)
 - 23 (markov or markow or monte carlo).tw. (95206)
 - 24 quality-adjusted life years/ (25352)
 - 25 (QOLY or QOLYs or HRQOL or HRQOLs or QALY or QALYs or QALE or QALEs).tw. (46567)
 - 26 ((adjusted adj (quality or life)) or (willing* adj2 pay) or sensitivity analys*s).tw. (91391)
 - 27 or/12-26 (2203643)
 - 28 11 and 27 (55)
 - 29 28 use pmoz,cctr,coch,dare,clhta (11)
 - 30 11 use cleed (2)
 - 31 29 or 30 (13)
 - 32 limit 31 to english language [Limit not valid in CDSR,DARE; records were retained] (11)
 - 33 cerebral palsy/ (44928)
 - 34 ((cerebr* adj pals*) or CP).tw. (124747)
 - 35 spasticity/ (16900)
 - 36 (spasticit* or (spastic* adj (diple* or hemiple* or tetrap* or triple* or quadriple*)) or little\$1 disease*).tw. (26306)

- 37 or/33-36 (162041)
- 38 exp rhizotomy/ (3113)
- 39 "spinal root"/su [Surgery] (2155)
- 40 spinal ganglia/su [Surgery] (354)
- 41 rhizotom*.tw. (3934)
- 42 or/38-41 (6905)
- 43 37 and 42 (1295)
- 44 Economics/ (250449)
- 45 Health Economics/ or exp Pharmacoeconomics/ (211572)
- 46 Economic Aspect/ or exp Economic Evaluation/ (382637)
- 47 (econom* or price or prices or pricing or priced or discount* or expenditure* or budget* or pharmaco-economic* or pharmaco-economic*).tw. (655807)
- 48 exp "Cost"/ (495890)
- 49 cost*.ti. (224965)
- 50 cost effective*.tw. (236192)
- 51 (cost* adj2 (util* or efficacy* or benefit* or minimi* or analy* or saving* or estimate* or allocation or control or sharing or instrument* or technolog*).ab. (147519)
- 52 Monte Carlo Method/ (49152)
- 53 (decision adj1 (tree* or analy* or model*).tw. (31975)
- 54 (markov or markow or monte carlo).tw. (95206)
- 55 Quality-Adjusted Life Years/ (25352)
- 56 (QOLY or QOLYs or HRQOL or HRQOLs or QALY or QALYs or QALE or QALEs).tw. (46567)
- 57 ((adjusted adj (quality or life)) or (willing* adj2 pay) or sensitivity analys*s).tw. (91391)
- 58 or/44-57 (1805287)
- 59 43 and 58 (54)
- 60 59 use emez (41)
- 61 limit 60 to english language [Limit not valid in CDSR,DARE; records were retained] (38)
- 62 32 or 61 (49)
- 63 remove duplicates from 62 (44)
- 64 62 use pmoz (5)
- 65 62 use emez (38)
- 66 62 use cctr (0)
- 67 62 use coch (3)
- 68 62 use dare (1)
- 69 62 use clhta (0)
- 70 62 use cleed (2)

Appendix 2: Evidence Tables

Appendices list studies reviewed for the health technology assessment and include A1. Alphabetical list of all studies reviewed; A2. List of all studies reviewed with short term outcomes grouped by country; A3. List of all studies reviewed with long term outcomes grouped by country; and A4. List of all studies reviewed with safety evaluations grouped by country.

Table A1: Evidence Base of Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Abbott et al, 1992 ¹⁵⁶ and 1993 ¹⁵⁷	New York, United States	Single pre-post cohort	Since 1986	250	Safety: To describe peri-operative and long-term complications after DR and protective measures implemented
Abel et al, 2005 ²⁶⁴	St Louis, United States	Control-matched pre-post cohort, eligible CP cases not undergoing DR	1999 to 2001	120	Comparative Short Term: To compare walking pattern of children with CP undergoing and not undergoing DR
Adams et al, 1995 ²⁶⁵	Northridge, Calif, United States	Single pre-post cohort	--	14	Short Term: To evaluate foot-floor contact pattern in children with CP after DR
Ailon et al, 2015 ¹³⁹	Vancouver, Canada	Single pre-post cohort	--	44	Long Term: To evaluate long-term effects of DR on hip spasticity, hip range of motion, quadriceps strength, and motor function
Albright et al, 1995 ²⁶⁶	Pittsburgh, United States	Single pre-post cohort compared with cohort undergoing continuous ITB	--	76	Comparative Short Term: To compare effects of DR and ITB on upper-extremity spasticity and range of motion
Arens et al, 1989 ¹⁴⁷	Cape Town, South Africa	Single pre-post cohort	1981 to 1984	60	Long Term: To determine degree and sustainability of functional improvement, occurrence of disadvantages, and factors to develop eligibility criteria
Beck et al, 1993 ²⁶⁷	San Antonio, Tex, United States	Single pre-post cohort	June 1987 to February 1990	21	Short Term: To evaluate change in upper-extremity function after DR
Berman et al, 1990 ¹⁴²	Cape Town, South Africa	Single pre-post cohort	--	29	Short Term: To evaluate functional outcomes in children with CP after DR
Bolster et al, 2013 ¹²⁹	Amsterdam, Netherlands	Single pre-post cohort compared with reference centiles	August 1998 to October 2007	36	Long Term: To evaluate long-term effects of DR on gross motor function compared with reference centiles
Boscarino et al, 1993 ²⁶⁸	St Paul, Minn, United States	Single pre-post cohort	--	19	Short Term: To compare independent and dependent ambulators for impact of DR on motion of pelvis, hip, and knee in 3 planes and transverse motion of foot using 3-D analysis
Buckon et al, 2004 ⁹³	Portland, Ore, United States	Single pre-post cohort	--	25	Comparative Short Term: To compare effects of DR and orthopedic surgery on multidimensional outcomes
Carraro et al, 2014 ⁹⁶	Conegliano and Torino, Italy	Single pre-post cohort	--	9	Short Term: To describe assessment and multidimensional outcome measures after DR
Chan et al, 2008 ⁹⁷	Hong Kong	Single pre-post cohort	--	21	Short Term: To evaluate effects of DR and PT on neuromotor performance, activity performance, and participation in daily context

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Chan et al, 2013 ²²⁵	Hong Kong	Single pre-post cohort	2003 to 2010	53	Safety, Hip: To evaluate effect of DR and other factors on hip development in Asian children
Chicoine et al, 1996 ²⁶⁹	St Louis, United States	Single pre-post cohort	--	90	Short Term: To assess correlation between foot dorsiflexion and ability to walk independently after DR
Chiu et al, 2014 ¹⁵⁹	Hong Kong	Single pre-post cohort	June 2003 to August 2010	56	Safety, Bladder: To evaluate effect of DR on bladder function and urodynamic findings after DR
Cole et al, 2007 ²⁷⁰	Oswestry, Shropshire, United Kingdom	Single pre-post cohort	--	19	Short Term: To evaluate impact of strict eligibility criteria on improving outcomes using gait analysis
Craft et al, 1995 ²⁷¹	St Louis, United States	Control and normal matched single pre-post cohort	--	49	Comparative Short Term: To evaluate changes in cognitive performance in children with spastic diplegic CP after DR
Crawford et al, 1996 ¹⁷⁶	Dallas, United States	Case reports	--	2	Safety, Spine: To describe cases of severe lumbar lordosis occurring after DR
Deletis et al, 1992 ¹⁶⁰	New York, United States	Single pre-post cohort	--	31	Safety, Bladder: To evaluate effect of intraoperative monitoring technique to reduce risk of bladder dysfunction
Dudgeon et al, 1994 ¹¹¹	Seattle, United States	Single pre-post cohort	--	29	Short Term: To evaluate upper-limb movement, self-care, and functional mobility after DR
Dudley et al, 2013 ¹³⁰	Montreal, Canada	Single pre-post cohort	October 1991 to August 2001	102	Long Term: To evaluate long-term functional benefits, protection against adolescent declines in function, and relationship between heterogeneity in DR response and baseline patient characteristics
Eliasson et al, 2000 ¹²⁸	Stockholm, Sweden	Single pre-post family cohort	--	7	Short Term: To determine if parents detected changes after DR not identified by previous research
Engsberg et al, 1998 ²⁷²	St Louis, United States	Normal group pre-post cohort	--	25	Comparative Short Term: To compare changes in hamstring muscle spasticity and strength between children with CP undergoing DR and nondisabled children
Engsberg et al, 2006 ⁹⁴	St Louis, United States	Pre-post DR cohort compared with CP group having PT only and with age-matched nondisabled children	--	117	Comparative Short Term: To compare multi-dimensional outcomes in children with CP undergoing DR vs. children with CP undergoing only intensive PT and vs. age-matched nondisabled children
Engsberg et al, 2004 ²⁷³	St Louis, United States	Pre-post DR cohort compared with CP group having PT only and with age-matched nondisabled children	--	46	Comparative Short Term: To quantify and compare ankle range of motion between children with CP and nondisabled children and before and after DR
Engsberg et al, 2002 ²⁷⁴	St Louis, United States	Pre-post DR cohort compared with nondisabled children	--	59	Comparative Short Term: To evaluate and compare changes in hip adductor spasticity and strength after DR with nondisabled children

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Engsberg et al, 2007 ²⁷⁵	St Louis, United States	Single pre-post cohort	--	33	Short Term: To evaluate relationship between pre-intervention objective measures of spasticity, strength, function, and gait and post intervention changes in gait and strength
Fasano et al, 1980 ²⁷⁶	Torino, Italy	Single pre-post cohort	--	80	Long Term: To evaluate effects of DR on functional consequences, suprasegmental effects, and complications
Feger et al, 2015 ⁹¹	Charlottesville, Va, United States	Single pre-post cohort	January 1993 to December 2008	94	Short Term: To compare effects of DR on functional and gait improvements with effects of osteotomies and muscle tendon surgery
Floeter et al, 2014 ²²⁶	Berlin, Germany	Single pre-post cohort	--	33	Safety, Hip: To evaluate changes in hip geometry after DR
Fukuhara et al, 2000 ²⁷⁷	Cleveland, United States	Single pre-post cohort	--	37	Short Term: To evaluate relationship between percentage of nerve rootlets sectioned and spasticity improvement
Funk et al, 2015 ⁹⁸	Berlin, Germany	Single pre-post cohort	--	54	Short Term: To evaluate relationship between pre-operative age, BMI, strength, spasticity, and motor function on post-operative changes in function
Funk et al, 2016 ⁶⁷	Berlin, Germany	Single pre-post cohort	--	132	Safety, Spine Scoliosis: To evaluate effect of single-level DR surgery on development of scoliosis in ambulatory children with CP
Galarza et al, 2001 ²⁷⁸	Los Angeles, United States	Single pre-post cohort	--	5	Short Term: To evaluate whether limited DR would reduce risk of post-operative hypotonia
Gigante et al, 2013 ²⁷⁹	New York, United States	Single pre-post cohort	2005 to 2011	42	Short Term: To evaluate effect of DR on upper-extremity tone
Golan et al, 2007 ¹⁶⁷	Montreal, Canada	Single pre-post cohort	September 1991 to June 2001	98	Safety, Spine: To evaluate frequency of spinal deformities and their risk factors
Gooch et al, 1996 ¹⁷⁷	Salt Lake City, United States	Case reports	1989 to 1990	2	Safety, Spine: To describe cases of spinal stenosis associated with abnormal gait after DR
Graubert et al, 2000 ²⁸⁰	Seattle, United States	RCT	--	43	Short Term: To compare effects of DR and PT to PT alone on ambulatory status and gait
Greene et al, 1991 ²²⁷	Chapel Hill, NC, United States	Case series	--	6	Safety, Hip: To describe rapid progression of hip subluxation after DR
Grunt et al, 2010 ²⁸¹	Amsterdam, Netherlands	Single pre-post cohort	August 1998 to March 2009	44	Short Term: To evaluate association between pre-operative characteristics of gait and gait improvement
Grunt et al, 2010 ¹³¹	Amsterdam, Netherlands	Single pre-post cohort	January 1998 to December 2007	36	Long Term: To evaluate relationship of MRI findings to gross motor function of patients before and after DR
Gul et al, 1999 ¹³⁸	Vancouver, Canada	Single pre-post cohort	--	33	Long Term: To evaluate long-term effects of DR on spasticity, range of motion, muscle strength, motor function, and adjunct orthopedic surgeries
Heim et al, 1995 ²²⁸	St Louis, United States	Single pre-post cohort	August 1987 to November 1990	45	Safety, Hip: To evaluate changes in lateral hip migration after DR and relationship to age and baseline measure of lateral migration
Hicdonmez et al, 2005 ²²⁹	Vancouver, Canada	Single pre-post cohort	1987 to 2001	82	Safety, Hip: To determine incidence of hip subluxation and risk factors after DR

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Hodgkinson et al, 1997 ¹⁰²	Lyon, France	Single pre-post cohort	April 1991 to April 1995	18	Short Term: To evaluate effects of DR on spasticity, range of motion, lateral extrusion of hips, and GMFM scores in spastic quadriplegia
Horinek et al, 2008 ²⁸²	Prague, Czech Republic	Case reports	--	4	Short Term: To evaluate effect of DR on pre-operative spasticity and pathologic eye movement in quadriplegia
Houle et al, 1998 ¹⁶¹	Montreal, Canada	Single pre-post cohort	January 1992 to September 1995	40	Safety, Bladder: To evaluate bladder function after DR
Huang et al, 1997 ¹⁶²	New York, United States	Single pre-post cohort	1991 to 1995	114	Safety, Bladder: To evaluate effectiveness of intraoperative technique to minimize risk of bowel, bladder, and sexual dysfunction after DR
Hurvitz et al, 2013 ¹⁵¹	Ann Arbor, Mich, and Chicago, United States	Single pre-post cohort	1987 to 1991	85	Long Term: To assess long-term outcomes of DR in adults and their (or family members') perceptions of procedure, and of surgery's effect on function, pain, and satisfaction with life and need for additional intervention for hypotonicity
Ingale et al, 2016 ¹²⁷	Nottingham, United Kingdom	Single pre-post cohort	--	10	Short Term: To evaluate effect of DR as alternative to ITB pump replacement
Johnson et al, 2004 ¹⁷⁰	Portland, Ore, United States	Single pre-post cohort	January 1990 to July 1997	34	Safety, Spine: To assess incidence of spinal deformity in ambulatory spastic diplegia and whether laminoplasty offers any advantages over laminectomy in DR
Josenby et al, 2012 ¹³²	Lund, Sweden	Single pre-post cohort	--	29	Long Term: To evaluate changes after DR in muscle tone, range of motion, and gross motor function, to identify factors associated with these changes and subsequent interventions (botulism toxin A and orthopedic surgery)
Josenby et al, 2015 ¹⁴³	Lund, Sweden	Single pre-post cohort	1995 to 1999	24	Long Term: To evaluate functional performance in self-care and mobility after DR
Kan et al, 2008 ²⁸³	Salt Lake City, Utah, United States	Single pre-post DR cohort compared with age- and motor function-matched pre-post cohort receiving ITB	DR before 1997, baclofen after 1997	142	Comparative Short Term: To compare outcomes of lower-extremity tone, range of motion, subsequent orthopedic surgeries, and parents' satisfaction with DR and ITB
Kaufman et al, 1994 ¹²⁵	Morgantown, WV, United States	Single pre-post cohort	--	19	Short Term: To evaluate muscle tone, range of motion, static postures, transitional movements, ambulatory ability, complications, and patients' and families' satisfaction with DR
Kim et al, 2001 ¹⁵⁴	Seoul, South Korea	Single pre-post cohort	1990 to 1999	208	Long Term: To evaluate long-term effect of DR on functional ability and adverse effects
Kim et al, 2002 ¹⁵⁵	Seoul, South Korea	Single pre-post cohort	1989 to 1998	200	Safety: To evaluate effect of DR rootlet sectioning techniques on adverse outcomes
Kim et al, 2006 ¹²¹	Vancouver, Canada	Single pre-post cohort	1987 to February 2002	174	Short Term: To determine predictors of poor performance after DR
Kinghorn et al, 1992 ¹²²	Calgary, Canada	Single pre-post cohort	March 1989 to February 1990	7	Short Term: To evaluate effect of DR on upper-extremity function

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Lang et al, 1994 ¹⁶³	New York, United States	Single pre-post	1986 to 1991	85	Safety, Bladder: To evaluate residual spasticity with and without inclusion of S2 dorsal rootlets in DR
Langerak et al, 2007 ²⁸⁴	Cape Town, South Africa	Single pre-post cohort	1985	14	Long Term: To evaluate long-term effect of DR on gait
Langerak et al, 2008 ²⁸⁵	Cape Town, South Africa	Single pre-post cohort compared with age-matched healthy controls	1985	14	Long Term: To evaluate long-term effect of DR on gait
Langerak et al, 2009 ¹⁴⁰	Cape Town, South Africa	Single pre-post cohort	--	14	Long Term: To examine effect of DR on functional status on basis of ICF model dimensions of body structure and function as well as activity
Langerak et al, 2009 ¹⁷³	Cape Town, South Africa	Single pre-post cohort	1981 to 1991	30	Safety, Spine: To evaluate incidence of spinal abnormalities in spastic diplegia after DR
Langerak et al, 2011 ¹⁵²	Cape Town, South Africa	Single pre-post cohort	1981 to 1991	31	Long Term: To examine level of activity and participation after DR and of patient satisfaction
Langerak et al, 2012 ²⁸⁶	Cape Town, South Africa	Single pre-post cohort compared with normal adults without history of orthopedic or neurologic pathology	--	74	Comparative Long Term: To evaluate long-term effects of DR on gait using 3-D gait analysis and of subsequent interventions
Lazareff et al, 1990 ²⁸⁷	Mexico City, Mexico	Single pre-post cohort	--	30	Short Term: To evaluate effect of limited DR procedure on spasticity of muscles in upper and lower limbs
Lazareff et al, 1999 ²⁸⁸	Mexico City, Mexico	Single pre-post cohort	--	71	Short Term : To compare effect of 2- vs. 3-level DR on lower-limb muscle tone and gait
Lewin et al, 1993 ²⁸⁹	Chicago, United States	Single pre-post cohort	1988 to 1990	36	Short Term: To evaluate self-help and upper-extremity changes after DR
Li et al, 2008 ¹⁶⁹	Guangzhou, China	Single pre-post cohort	October 1992 to December 2002	61	Safety, Spine: To evaluate incidence of lumbar spinal deformity after DR
Loewen et al, 1998 ¹¹⁷	Vancouver, Canada	Single pre-post cohort	November 1991 to March 1996	36	Short Term: To evaluate upper-extremity performance and self-care skill after DR
MacWilliams et al, 2011 ⁹²	Salt Lake City, United States	Single pre-post cohort compared with age- and function-matched children with CP receiving orthopedic surgery or no surgery	--	26	Comparative Short Term: To compare effects of DR on gait in older children with effects of either orthopedic surgery only or no surgery
Maenpaa et al, 2003 ²⁹⁰	Helsinki, Finland	Single pre-post cohort compared with active controls	April 1991 to September 1998	44	Long Term: To compare effect of DR and PT to PT only on spasticity, lower-limb strength, and function

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Marty et al, 1995 ²⁹¹	Chicago, United States	Single pre-post cohort compared with orthopedic surgery cohort	1987 to 1989	50	Comparative Long Term: To compare effects of DR to orthopedic soft-tissue surgeries on range of motion and ambulation
McFall et al, 2015 ²⁹²	Oswestry, United Kingdom	Single pre-post cohort	November 1996 to January 2007	17	Long Term: To evaluate sustainability of short-term effects and need for subsequent orthopedic surgery
McLaughlin et al, 1994 ¹⁰³	Seattle, United States	Single pre-post cohort	June 1988 to October 1991	34	Short Term: To evaluate changes in spasticity (tone, deep tendon reflexes, clonus) and function (joint range and independent movement) after DR
McLaughlin et al, 1998 ⁷³	Seattle, Washington	Single-site RCT	July 1991 to September 1994	38	Short Term: To compare efficacy and safety of DR and PT to PT only in spastic diplegia
McLaughlin et al, 2002 ⁸⁸	Seattle, Washington	Meta-analysis of 3 RCTs	--	90	Short Term: To analyze short-term outcomes in 3 RCTs comparing DR and PT to PT in spastic diplegia
McLaughlin et al, 2005 ¹⁶⁵	Seattle, United States	Single pre-post cohort compared with group without CP	--	127	Comparative Safety, Sensory: To evaluate feasibility of lower-extremity sensory testing, compare differences between children with and without CP, compare sensory changes after DR
Mittal et al, 2002 ²⁹³	Montreal, Canada	Single pre-post cohort	--	45	Long Term: To evaluate effect of DR on fine motor skills, adjunct orthopedic surgeries, botulinum toxin A injections, and complications
Mittal et al, 2002 ¹⁴⁴	Montreal, Canada	Single pre-post cohort	--	71	Long Term: To evaluate sustainability of DR on lower-limb spasticity, passive range of motion ADL, mobility, and use of complementary interventions
Montgomery et al, 1992 ¹²⁶	St Louis, United States	Single pre-post retrospective cohort	--	14	Short Term: To evaluate changes in function (motor skills and spasticity), sensation, and bladder function; orthopedic surgery; and family perception of DR
Mooney et al, 1999 ¹⁷⁸	Boston and Salem, NC, United States	Case series	--	6	Safety, Spine: To describe cases of spinal deformity occurring after DR
Morota et al, 2003 ²⁹⁴	Tokyo, Japan	Case series	March 1996 to May 2002	3	Short Term: To evaluate outcomes of DR for children with mixed type (spastic and dystonia) of CP
Moroto et al, 1995 ²⁹⁵	New York, United States	Single pre-post cohort	1986 to 1990	109	Short Term: To evaluate relationship between various approaches to nerve sectioning and residual spasticity after DR
Nishida et al, 1995 ¹¹⁶	Chicago, United States	Single pre-post cohort	--	96	Short Term: To evaluate range of motion and ambulation skills, and to measure overall self-care and toileting
Nordmark et al, 2000 ¹⁰⁴	Lund, Sweden	Single pre-post cohort	February 1994 to April 1997	18	Short Term: To compare outcomes based on GMFM and PEDI between mild to moderate impairment vs. severe impairment after DR and individualized PT
Nordmark et al, 2008 ¹³⁴	Lund, Sweden	Single pre-post cohort	--	35	Long Term: To evaluate long-term functional outcomes, safety, and side effects after DR
O'Brien et al, 2004 ¹⁴⁹	St Louis, United States	Single pre-post cohort	1990 to 1994	77	Safety, Surgery: To evaluate whether rate of orthopedic surgery in spastic quadriplegia varies by aided or independent ambulation status or age at DR and to evaluate parental views on DR

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
O'Brien et al, 2005 ¹⁵⁰	St Louis, United States	Single pre-post cohort	1990 to 1994	158	Safety, Surgery: To evaluate whether rate of orthopedic surgery in spastic diplegia varies by aided or independent ambulation status or age at DR and to evaluate parental views on DR
Olree et al, 2000 ²⁹⁶	St Louis, United States	Single pre-post cohort compared with normal control	--	27	Comparative Short Term: To evaluate effects of DR on synergistic movement patterns
Ou et al, 2010 ²⁹⁷	Vancouver, Canada	Single pre-post cohort	--	27	Comparative Short Term: To compare peri-operative outcomes of single-level vs. multi-level laminectomy DR
Park et al, 1993 ²⁹⁸	St Louis, United States	Single pre-post cohort	April 1991 to October 1992	66	Short Term: To evaluate outcomes after introducing single-level DR
Park et al, 1994 ²³⁰	St Louis, United States	Single pre-post cohort	July 1987 to April 1990	67	Safety, Hip: To evaluate effect of DR on hip stability
Parise et al, 1997 ¹⁶⁶	Lyon, France	Case series	--	10	Safety, Sensory: To evaluate effect of DR on somatosensory function of lower limbs
Peacock et al, 1987 ²⁹⁹	Cape Town, South Africa	Single pre-post cohort	--	60	Short Term: To describe and compare outcomes of tone, power, positional function (side sitting, locomotion in prone, standing, ambulation), upper-limb function, and speech after DR in 3 groups of children varying in degree of disability
Peacock et al, 1982 ¹²³	Cape Town, South Africa	Single pre-post cohort	--	15	Short Term: To describe and compare outcomes of tone, power, positional function (side sitting, locomotion in prone, standing, ambulation), upper-limb function, and speech after DR in 2 groups of children varying in degree of disability
Peter et al, 1990 ¹⁷⁵	Cape Town, South Africa	Single pre-post cohort	--	55	Safety, Spine: To determine incidence of spinal deformity after multiple-level laminectomy for DR
Peter et al, 1993 ¹⁷⁴	Cape Town, South Africa	Single pre-post cohort	Since 1981	99	Safety, Spine: To determine incidence of spondylolysis and spondylolisthesis after 5-level DR
Peter et al, 1993 ¹⁴⁸	Cape Town, South Africa	Single pre-post cohort	1981 to 1991	110	Long Term: To describe long-term outcomes after DR
Peter et al, 1994 ¹²⁴	Cape Town, South Africa	Single pre-post cohort	--	30	Short Term: To describe outcomes of tone, power, sitting and standing, ambulation, sensation, spinal abnormalities, and patient response in teenagers and young adults who had DR as children
Reynolds et al, 2011 ⁹⁹	St Louis, United States	Single pre-post cohort	--	21	Short Term: To describe joint range of motion in lower extremity, GMFM, muscle tone, ADLs in adult patients
Ross et al, 2001 ³⁰⁰	St Louis, United States	Single pre-post DR compared with nondisabled children	--	39	Comparative Short Term: To compare change in strength of quadriceps and hamstrings after DR and compare pre-post measures with nondisabled children
Sacco et al, 2000 ¹⁰⁰	Lexington, Ky, United States	Single pre-post cohort	--	10	Short Term: To evaluate changes in gait using 3-D gait analysis after nonselective DR rootlet section
Schijman et al, 1993 ³⁰¹	Buenos Aires, Argentina	Single pre-post cohort	April 1989 to October 1991	27	Short Term: To evaluate self-care, ambulation, and upper-limb spasticity after DR

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Schwartz et al, 2004 ³⁰²	St Paul, Minn, United States	Single pre-post cohort, within-group surgical comparison	January 1994 to January 2002	135	Short Term: To evaluate effects of DR and orthopedic surgery on gait
Silva et al, 2012 ²³¹	Ann Arbor, Mich, United States	Single pre-post DR cohort compared with ITB	1988 to 2002	119	Comparative Short Term Safety: To compare hip dislocation and containment procedures between DR and ITB in nonambulatory CP patients
Spiegel et al, 2004 ¹⁷¹	Minneapolis, United States	Single pre-post cohort	Since 1991	79	Safety, Spine: To evaluate frontal and sagittal spinal alignment to determine prevalence of scoliosis, thoracic hyperkyphosis, lumbar hyperlordosis, and spondylolisthesis in ambulatory CP after DR
Steinbok et al, 1998 ¹⁵³	Vancouver, Canada	Single pre-post cohort	1987 to 1996	158	Safety: To evaluate frequency and type of complications during and after DR
Steinbok et al, 1992 ³⁰³	Vancouver, Canada	Single pre-post cohort	February 1987 and first 50 cases	50	Short Term: To describe evolution of DR surgery and outcomes of function, suprasegmental effects, and complications
Steinbok et al, 1997 ⁷⁴	Vancouver, Canada	Single-site RCT	--	30	Short Term: To compare effectiveness of DR and intensified PT to intensified PT in spastic diplegia
Steinbok et al, 1997 ⁹⁰	Vancouver, Canada	Single-site RCT	--	44	Short Term: To compare effect of therapeutic electrical stimulation to no stimulation after DR on functional outcome
Steinbok et al, 2002 ⁸⁹	Vancouver, Canada	Single-site RCT	--	28	Short Term: To evaluate whether intensified PT before DR improves outcomes at 2 years over no pre-operative intensified PT
Steinbok et al, 2005 ¹⁶⁸	Vancouver, Canada	Single pre-post cohort	1987 to 2001	105	Safety, Spine: To evaluate incidence of scoliosis, kyphosis, and hyperlordosis after DR
Steinbok et al, 2009 ⁹⁵	Vancouver, Canada	Single-site cohort compared with age-matched control	--	24	Comparative Short Term: To evaluate utility of electrophysiologically guided versus non-electrophysiologically guided DR on number of dorsal roots cut, outcomes, and complications
Subramanian et al, 1998 ³⁰⁴	Cape Town, South Africa	Single pre-post cohort compared with age-matched healthy children	--	123	Comparative Long Term: To evaluate long-term effect of DR on gait
Sweetser et al, 1995 ¹⁶⁴	New York, United States	Single pre-post cohort	November 5, 1990, to January 20, 1994	36	Safety, Bladder: To evaluate effects of DR on urinary tract symptoms and function
Tedroff et al, 2011 ¹³⁵	Stockholm, Sweden	Single pre-post cohort	1993 to 1997	19	Long Term: To evaluate sustainability of spasticity reduction and its correlation with gross motor function, gait development, range of motion, and need for orthopedic surgery
Thomas et al, 1997 ³⁰⁵	Portland, Ore, United States	Single pre-post cohort	--	27	Short Term: To evaluate effect of DR on gait using 3-D gait analysis
Tichy et al, 2003 ¹¹⁸	Prague, Czech Republic	Single pre-post cohort	2001 to 2002	14	Short Term: To evaluate effect of DR on spasticity and spasms, range of motion, functional change in self-care and mobility, and complications

Author, Year	Location	Study Design	Surgical Intervention Period	Patients, N	Study Objective
Trost et al, 2008 ³⁰⁶	St Paul, Minn, United States	Single pre-post cohort	--	136	Short Term: To evaluate outcomes of spasticity, gait, functional mobility, and complications for patients undergoing rigorous selection process for DR
Turi et al, 2000 ¹⁷²	Gainesville, Fla, United States	Single pre-post cohort	1987 to 1995	47	Safety, Spine: To evaluate type and rates of spinal deformities after DR and etiology of deformities and suggested treatment
Van De Wiele, 1996 ¹⁵⁸	Los Angeles, United States	Single pre-post cohort	1986 to 1991	105	Safety: To evaluate incidence and clinical significance of adverse peri-operative events and their potential risk factors after DR
Van Schie et al, 2005 ¹⁰¹	Amsterdam, Netherlands	Single pre-post cohort	--	9	Short Term: To evaluate effect of DR on spasticity, range of motion, functional abilities of gross motor function, self-care, caregiver assistance
Van Schie et al, 2011 ¹³⁶	Amsterdam, Netherlands	Single pre-post cohort	August 1998 to December 2005	33	Long Term: To evaluate short- and long-term effect of DR on gross motor function and spasticity and to compare effects between children ambulating with or without aids. Secondary aims were to investigate occurrence of side effects, need for additional treatment (botulinum type A or orthopedic surgery) and parental satisfaction
Vaughan et al, 1988 ³⁰⁷	Los Angeles, United States	Single pre-post cohort	--	14	Short Term: To evaluate effect of DR on gait (cadence, stride length, and walking speed)
Vaughan et al, 1991 ³⁰⁸	Los Angeles, United States	Single pre-post cohort	1985	29	Short Term: To compare effect of DR on gait patterns at 2- and 3-year follow-up with pre-operative values
Wang et al, 2005 ³⁰⁹	Dalian, Liaoning Province, China	Single pre-post cohort	January 2001 to December 2002	30	Short Term: To evaluate effects of DR and adductor tenotomy on motor ability
Wong et al, 2005 ³¹⁰	Taipei, Taiwan	Single pre-post cohort compared with control group	--	62	Comparative Short Term: To compare effect of DR with botulism toxin A and rehabilitation only on gait performance
Wong et al, 2000 ³¹¹	Taipei, Taiwan	Single pre-post cohort with matched control group	--	79	Comparative Short Term: To determine whether lower motor control assessments could predict improved motor capability after DR
Wright et al, 1998 ⁷⁵	Toronto, Canada	Single-site RCT	--	24	Comparative Short Term: To compare effectiveness of DR and PT/OT with PT/OT on outcomes including passive and active range of motion, spasticity, mobility, and function
Yang et al, 1996 ³¹²	Taipei, Taiwan	Single pre-post cohort	August 1991 to June 1993	17	Short Term: To evaluate effect of DR on trunk stability and sitting balance

Abbreviations: ADL, activities of daily living; BMI, body mass index (measured as kg/m²); CP, cerebral palsy; DR, dorsal rhizotomy; GMFM, Gross Motor Function Measure; ICF, International Classification of Functioning, Disability, and Health; ITB, intrathecal baclofen; MRI, magnetic resonance imaging; N, number; NR, not recorded; OT, occupational therapy; PEDI, Pediatric Evaluation of Disability Inventory; PT, physiotherapy; RCT, randomized controlled trial.

Table A2. Short-Term Effectiveness of Lumbrosacral Dorsal Rhizotomy for Spastic Cerebral Palsy

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Argentina						
Schijman et al, 1993, ³⁰¹ Buenos Aires	Pre-post cohort (1–30 months)	30 (20 M, 10 F) (3–20 years)	Diplegia (n = 8): assisted ambulatory (n = 2), nonambulatory (n = 6) Quadriplegia (n = 19)	Spasticity (clinical observation)	ADL (clinical observation) Ambulation (clinical observation)	No
Canada						
Kinghorn et al, 1992, ¹²² Calgary	Pre-post cohort 6 months	7 (7 M) (7–16 years)	Diplegia (n = 2: 2 ambulatory) Quadriplegia (n = 5: 4 wheelchair, 1 walker)	Upper-extremity spasticity (Modified Ashworth Scale)	Upper-extremity function Fine motor skills (Developmental Test of Visual-Motor Integration) ADL score for dressing, eating, and brushing teeth Parents' satisfaction	No
Loewen et al, 1998, ¹¹⁷ Vancouver	Pre-post cohort 12 months	36 (17 M, 19 F) 4.1 years (2.9–14.6 years)	Diplegia (n = 23) Quadriplegia (n = 13)	--	Upper-extremity performance (QUEST) Independence, self-care, or ADL (WeeFIM)	No
Kim et al, 2006, ¹²¹ Vancouver	Pre-post cohort 12 months	174 (96 M, 78 F) 5.3 years (2.2–18 years)	Diplegia (n = 102) Quadriplegia (n = 72) Triplegia (n = 3) Ambulatory (n = 74) Wheelchair (n = 35) Crawl (n = 65)	--	Predictors of treatment success (based on caregiver opinion at 1 year)—prior orthopedic surgeries, older age, intellectual or speech delay, seizure history, gross motor function	No
Ou et al, 2010, ²⁹⁷ Vancouver	Pre-post cohort Group 1: Single-level DR (post-2005) Group 2: Multi-level DR (1987–2005)	Group 1: 9 (6 M, 3 F) 6.1 ± 1.4 years Group 2: 18 (10 M, 8 F) 5.8 ± 1.7 years	--	--	Peri-procedural outcomes (length of surgery, post-operative pain [FLACC scale, Faces Pain Scale, VAS]) Time to mobilization Length of hospital stay	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Steinbok et al, 1992, ³⁰³ Vancouver	Pre-post cohort 6 months	50 (NR) 5.9 years (2.2–8.1 years)	Diplegia (n = 29) Quadriplegia (n = 21)	Spasticity in lower limbs (Penny and Giles mometer) Passive range of movement (goniometer)	Ambulation status Suprasegmental effects Upper limb function, speech (clinical observation and caregiver report)	Yes Spinal subdural hematoma (n = 1), transient post-operative headache (n = 1), sensory loss (n = 1), transient urinary dysfunction (n = 4), prolonged hypersensitivity of feet (n = 1)
Steinbok et al, 1997, ⁷⁴ Vancouver	Single-site RCT Group 1: DR 15 (NR) Group 2: PT 15 (NR) 9 months	Group 1: 50 months (35–75 months) Group 2: 47 months (35–77 months)	Diplegia (n = 30) Ambulatory (n = 30)	Spasticity (Ashworth Scale) hip adductors, knee flexors, ankle-plantar flexors Range of motion (goniometer), hip adductors, knee flexors, ankle-plantar flexors Muscle strength (knee extensors, hip abductors, hip extensors, ankle dorsiflexors)	GMFM Physiologic cost index Peabody Fine Motor Scale	Yes PT group (no complications), DR post-operative infection (n = 1), spinal abscess (n = 1), transient urinary retention (n = 1), transient back pain (n = 1)
Steinbok et al, 1997, ⁹⁰ Vancouver	Single-site RCT Group 1: DR 22 (NR) Group 2: DR +TENS 22 (NR)	Group 1: 7.2 years Group 2: 7.2 years (4.3–10 years)	Diplegia (n = 44, all upright ambulatory)	Spasticity (Modified Ashworth Scale), hip adductors, knee flexors, ankle-plantar flexors Range of motion (goniometer) in hip, knee, ankle) Muscle strength	GMFM Seated postural control measure	No
Steinbok et al, 2002, ⁸⁹ Vancouver	Single-site RCT Group 1: IPT + DR + IPT Group 2: 2 DR +PT 2 years	Group 1: 13 (NR) Group 2: 13 (NR)	Diplegia (n = 26)	Lower-limb spasticity (Ashworth Scale) Passive range of motion Muscle strength quadriceps (MRC)	GMFM	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Steinbok et al, 2009, ⁹⁵ Vancouver	Pre-post age-matched cohorts with and without EPG during DR Group 1: EPG DR Group 2: DR, no EPG 12 months	Group 1: 22 (NR) 5.2 ± 4.2 Group 2: 22 (NR) 5.7 ± 4.8	--	Spasticity (Modified Ashworth Scale) Passive range of motion Muscle strength (MRC)	GMFM VMFM Functional skills in self-care, mobility, cognition (WeeFIM) Upper-extremity performance (QUEST)	No
Wright et al, 1998, ⁷⁵ Toronto	Single-site RCT Group 1: DR + PT Group 2: PT 12 months	Group 1: 12 (7 M, 5 F) 57.8 months Group 2: 12 (7 M, 5 F) 58.3 months	Diplegia (n = 24)	Spasticity (Modified Ashworth Scale) Active and passive range of motion	Ambulation (timed walking distance 60 seconds) GMFM-88 2-D Gait (VICON videotape system and EMG signals) Additional orthopedic surgeries	Yes DR: No major adverse effects, no sensory changes or bladder dysfunctions, urinary tract infection (n = 1)
China						
Wang et al, 2005, ³⁰⁹ Dalian, Liaoning Province	Pre-post cohort (DR + adductor tenotomy) 11 months (6 months to 3 years)	30 (NR) 5.3 years (4–13 years)	Spastic CP (all upright ambulatory)	Spasticity (Ashworth Scale) Muscle strength (Abott method)	Gait (clinical examination)	No
Czech Republic						
Tichy et al, 2003, ¹¹⁸ Prague	Pre-post cohort	14 (10 M, 4 F) 16.5 years (8–27 years)	Diplegia (n = 2: 2 ambulatory) Quadriplegia (n = 12: 12 nonambulatory)	Lower-limb spasticity (Ashworth Scale), Spasms Passive range of motion	Functional independence (WeeFIM) Gross motor function (Peacock grading system)	Yes Transient urinary infection (n = 3), moderately aggravated extrapyramidal symptoms (n = 1), acute pneumothorax (n = 1). No CNS leak or infection or sensory loss
Horinek et al, 2008, ²⁸² Prague	Case reports	4 (3 M, 1 F) 4–16 years	Quadriplegia (n = 3: 3 nonambulatory) Spastic paraparesis (n = 1: 1 assisted ambulatory)	Lower- and upper-limb spasticity (Ashworth Scale)	Visual function (eye movement control by video-oculography)	No
France						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Hodgkinson et al, 1997, ¹⁰² Lyon	Pre-post cohort 12 months	18 (12 M, 6 F) 9 years (5.5–16.5 years)	Spastic quadriplegia (n = 18)	Lower-limb spasticity, Range of motion	GMFM Parents' satisfaction	Yes Immediate and transient: meningocele (3 cases), wound complication (1 case), hyperesthesia in dorsal region of feet (2 cases), urinary infection (2 cases)
Germany						
Funk et al, 2015, ⁹⁶ Berlin	Pre-post cohort 2 years	54 (29 M, 25 F) 6.9 ± 2.9 years	Diplegia (n = 54: all ambulatory) GMFCS Levels I and II	Lower-limb spasticity (Modified Ashworth Scale); Muscle strength in hip, knee, ankle (MRC scale)	GMFM-88 Anthropometric data	Yes No changes in urinary function or sensitivity in dermatomes
Hong Kong						
Chan et al, 2008, ⁹⁷ Hong Kong	Pre-post cohort 12 months	20 (12 M, 9 F) 8.6 ± 2.6 years	Diplegia (n = 20) Quadriplegia (n = 1) GMFCS Level I (n = 8), Level II (n = 4), Level III (n = 2)	Lower-limb spasticity (Modified Ashworth Scale) Lower-limb passive range of motion (goniometry) Lower-limb muscle strength (MRC scale)	GMFM Gait (3-D VICON 624 system) Gait performance (Observation Gait Score) Efficiency, oxygen consumption Participation (COPM) PEDI Intelligence (Hong Kong Wechsler Intelligence Scale) Hip migration (Reimers Migration Index)	Yes 19 children had a urodynamic study before DR; 11 had bladder dysfunction suggestive of neurogenic bladder. None reported deteriorating urinary signs. Half of patients with prior detrusor instability had improved urinary signs after DR
Italy						
Carraro et al, 2014, ⁹⁶ Conegliano	Pre-post cohort 12 months	9 (6 M, 3 F) 7.9 ± 3.2 years	Diplegia (n = 9: 9 ambulatory) GMFCS Level II (n = 7), Level III (n = 2)	Spasticity (Modified Ashworth Scale) Passive range of motion (goniometry) Muscle strength in lower limbs (MRC scale)	Selective motor control of lower limb (SMCS) GMFM Functional independence (WeeFIM) Gait analysis (3-D motion analysis), not done for GMFCS Level III Gait efficiency by energy expenditure (breath by breath metabolimeter)	No
Japan						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Morota et al, 2003, ²⁹⁴ Tokyo	Case series 12 months	3 (2 F, 1 M): 3-year-old female, 4-year-old female, 10-year-old male	Quadriplegia, mixed-type CP (n = 3)	Spasticity (Ashworth Scale)	Care assistance required (parental report) Upper-extremity movement (clinical observation)	--
Mexico						
Lazareff et al, 1990, ²⁸⁷ Mexico City	Pre-post cohort 18 months	30 (17 M, 13 F) (4–12 years)	Spastic cerebral palsy (n = 30): independent ambulatory (n = 3), assisted ambulatory (n = 9), nonambulatory (n = 18)	Spasticity of upper and lower limbs (Ashworth Scale) Range of motion Muscle strength Sensory level (pain, touch, temperature, proprioception, 2-point discrimination)	Ability to balance while sitting Upper limb function (self-feeding, telephone dialing, hand grasp, handwriting) Subject well-being (self-report)	No
Lazareff et al, 1999, ²⁸⁸ Mexico City	Pre-post cohort 18 months	71 (38 M, 33 F) (4–12 years)	Diplegia (n = 35) Quadriplegia (n = 36)	Spasticity of lower limb (Sindou scale)	Gait (6-level score)	No
Netherlands						
Van Schie et al, 2005, ¹⁰¹ Amsterdam	Pre-post cohort 12 months	9 (4 M, 5 F) 65 months (43–82 months)	Diplegia (n = 9: 9 ambulatory) GMFCS Level II (n = 1), Level III (n = 8)	Spasticity (Ashworth Scale) Range of motion (potentiometer-based goniometer)	GMFM-88 Self-care (PEDI) 2-D gait pattern, SYBAR System (Edinburgh Visual Gait Score)	No
Grunt et al, 2010, ²⁸¹ Amsterdam	Pre-post cohort 24 months	30 (19 M, 11 F) 6.5 ± 1.96 years (2.8–13.2 years)	Diplegia (n = 30: 30 ambulatory) GMFCS Level I (n = 3), Level II (n = 9), Level III (n = 18)	Spasticity (Ashworth Scale) Range of motion (potentiometer-based goniometer), knee, and angle kinematics	GMFMS 2-D gait pattern, SYBAR System (Edinburgh Visual Gait Score, surface EMG)	No
South Africa						
Peacock et al, 1982, ¹²³ Cape Town	Pre-post cohort 12 months	15 (12 M, 3 F) (4–10 years)	Diplegia (n = 4) Quadriplegia (n = 10) Hemiplegia (n = 1)	Spasticity (clinical observation)	Gross motor function (clinical observation) Ease of care (parent report) Unexpected events (seizure reduction, mood status)	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Peacock et al, 1987, ²⁹⁹ Cape Town	Pre-post cohort Group 1: Spastic CP and moderate disability Group 2: Spastic CP and severely disabled Group 3: Mixed spastic CP and athetoid (1–5 years)	60 (42 M, 18 F) (20 months–19 years)	Group 1: (n = 40) Group 2: (n = 16) Group 3: (n = 4)	Spasticity at rest and with activity (clinical observation) Muscle strength (clinical observation)	Upper-limb function (clinical observation) Speech (clinical observation) Gross motor function (clinical observation) Unexpected events (seizure reduction, bladder control, emotional status, personality change)	Yes No early surgical complications; spondylolisthesis due to undetected pars interarticularis defect of 5 th lumbar vertebra (n = 1), no sensory loss or awareness
Berman et al, 1990, ¹⁴² Cape Town	Pre-post DR cohort (4–14 months) Normal controls	29 (18 M, 11 F) 9.3 years (2–35 years) 9 (NR) 5.5 years	Spastic CP: ambulating (n = 18), nonambulating (n = 11)	Spasticity (1–5 score) Range of motion (1–4)	Crawling and 2-D gait MicronEye (computerized camera recording) Voluntary movement and functional movement (scale 1–5)	No
Peter et al, 1994, ¹²⁴ Cape Town	Pre-post cohort 2 years (minimum)	30 (19 M, 11 F) 12–15 years (n = 18) 16–20 years (n = 10) 21 years (n = 1) 26 years (n = 1)	Diplegia (n = 26) Quadriplegia (n = 4)	Spasticity (clinical observation, self-report) Muscle strength (clinical observation)	Sitting/standing (clinical observation) Ambulation (video, self-report) Patient satisfaction	Yes All reported sensory abnormalities immediately after DR Of 25 followed 2 years or more, 1 had persistent dysesthesia not severe or requiring medication Of 25 having post-operative radiographs, 4 developed asymptomatic spondylolysis
Sweden						
Nordmark et al, 2000, ¹⁰⁴ Lund	Pre-post cohort 12 months	18 (12 M, 6 F) 4.3 ± 1.0 years	Diplegia (n = 18: 10 ambulatory) GMFCS Level II (n = 3), Level III (n = 7), Level IV (n = 7), Level V (n = 1)	NR	GMFM Functional independence (PEDI)	No
Eliasson et al, 2000, ¹²⁸ Stockholm	Single pre-post family cohort	7 (5 M, 2 F) (4–8 years) 10 parents	Diplegia (n = 7:3 ambulatory (n = 3), nonambulatory (n = 4)	NR	Parents' perspectives (qualitatively analyzed interviews)	No
Taiwan						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Yang et al, 1996, ³¹² Taipei	Pre-post cohort 3 months	17 (10 M, 7 F) 5.1 years (3–8 years)	Diplegia (n = 16: ambulatory (n = 11), nonambulatory (n = 5) Quadriplegia (n = 1), nonambulatory (n = 1)	Spasticity (clinical observation)	Trunk stability and balance while sitting (Chattecx Balance System)	No
Wong et al, 2000, ³¹¹ Taipei	Pre-post cohort (Group 1) with matched control group (Group 2) 12 months	Group 1: 40 (24 M, 16 F) 6.6 years (3–16 years) Group 2: 18 (10 M, 8 F) 6.7 years (3.2–14.0 years)	Diplegia (n = 33: independent ambulatory (n = 12), dependent ambulatory (n = 21) Quadriplegia (n = 7, all nonambulatory)	NR	Gait pattern (computerized portable foot pressure system [dynography] and PEMG, ground force reaction)	No
Wong et al, 2005, ³¹⁰ Taipei	Pre-post cohort (Group 2) compared with active control groups, BTA (Group 1) and PT (Group 3) 12 months	62 (45 M, 17 F) Group 1: 13 M, 8 F 4.9 ± 2.0 years Group 2: 12 M, 8 F 5.4 ± 2.4 years Group 3: 11 M, 9 F 5.0 ± 2.8 years	Diplegia (n = 62: independent ambulatory (n = 30), dependent ambulatory (n = 32)	NR	Gait pattern (computer-assisted gait analysis foot pressure system [dynography])	No
United Kingdom						
Cole et al, 2007, ²⁷⁰ Oswestry	Pre-post cohort 18 months	19 (13 M, 6 F) 8 years, 7 months (5–10 years)	Diplegia (n = 17: 17 ambulatory (n = 17) Hemiplegia (n = 1, ambulatory) Hereditary spastic paraparesis (n = 1, ambulatory)	Spasticity (Ashworth Scale) Lower limb muscle strength (MRC scale)	Gait 3-D pattern Weight gain	Yes Transient numbness on anterior aspect of thigh (n = 1), urinary incontinence for 3 months (n = 1), asymptomatic but clinically detectable cutaneous sensory loss on leg (n = 2), mild vertebral prominence (n = 3), hip subluxation requiring reconstruction (n = 1), weight gain, crossing weight centile charts (n = 18)

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Ingale et al, 2016, ¹²⁷ Nottingham	Pre-post cohort after ITB failure or replacement 12 months	10 (8 M, 2 F) 12.1 years (5–16 years)	Spastic CP GMFCS Level IV (n = 3), Level V (n = 7)	Upper- and lower-limb spasticity (Modified Ashworth Scale) Range of motion	Bladder function Pain Treatment satisfaction Ease of care (parent/caregiver report) Patient mood (parental report)	Yes No deterioration in bladder function, incontinence improved (n = 2), some improvement of total incontinence (n = 1)
United States						
California						
Vaughan et al, 1988, ³⁰⁷ Los Angeles	Pre-post cohort 9 months (5–14 months)	14 (NR) 8 years (2–14 years)	Spastic CP, all ambulation abilities	--	Gait patterns MicronEye (2-D kinematic)	No
Vaughan et al, 1991, ³⁰⁸ Los Angeles	Pre-post cohort (Group 1) compared with normal controls (Group 2) 3 years	Group 1: 11 (NR) 8 years (2–14 years) Group 2: 9 (NR) 5 years	Spastic CP, all ambulation abilities	--	Gait patterns (2-D kinematic)	Yes No post-operative wound complications or bowel or bladder sphincter control problems, no sensory loss in area of affected dermatomes, increased sensitivity to light touch in all after surgery, resolved within 5–10 days
Adams et al, 1995, ²⁶⁵ Northridge	Pre-post cohort 8.3 months (6–14 months)	14 6.5 years (4.6–23.5 years)	Diplegia (n = 14: independent ambulatory (n = 7), assisted ambulatory (n = 7))	--	Gait kinematics, Rancho Stride Analyzer and foot-floor contact pattern and gait kinematics	No
Galarza et al, 2001, ²⁷⁸ Los Angeles	Pre-post cohort 3 months (1–6 months)	5 (4 M, 1 F) 7 ± 3 years (4–12 years)	Spastic CP, all independent ambulatory	Spasticity (Modified Ashworth Scale) Range of motion	Gait 3-D pattern, Peak Performance Technologies	No
Illinois						
Lewin et al, 1993, ²⁸⁹ Chicago	Pre-post cohort 12 months	36 (21 M, 15 F) 4.5 years (2–11 years)	Diplegia (n = 24) Quadriplegia (n = 12)	Upper-limb spasticity (Modified Ashworth Scale) Upper-extremity change (range of motion, grasp, release)	Self-help (RIC-FAS)	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Nishida et al, 1995, ¹¹⁶ Chicago	Pre-post cohort 2 years	96 (56 M, 40 F) 5.4 years (1.1–12.8 years)	Diplegia (n = 60): ambulatory (n = 52), nonambulatory (n = 8) Quadriplegia (n = 36): nonambulatory (n = 28), assisted ambulatory (n = 8)	Range of movement	Ambulatory status (clinical observation) Functional independence (self-care, mobility, sphincter control [WeeFIM])	No
Kentucky						
Sacco et al, 2000, ¹⁰⁰ Lexington	Pre-post cohort 12 months	10 (NR) 8.5 years (5–16 years)	Diplegia (n = 10)	Spasticity (Modified Ashworth Scale) Range of motion	GMFM 3-D gait analysis, Motion Analysis Corporation (kinematic and kinetic data)	No
Minnesota						
Boscarino et al, 1993, ²⁶⁸ St Paul	Pre-post cohort 12 months	19 (13 M, 6 F) 5.5 ± 1.2 years (independent ambulatory), 6.3 ± 1.5 years (dependent ambulatory)	Diplegia (n = 11): independent ambulatory (n = 11) Quadriplegia (n = 8) assisted ambulatory (n = 8)	Spasticity (Ely test, ankle clonus score) Passive range of motion	3-D gait analysis (kinematics [joint angle changes] at the pelvis, hip, knee, and ankle) EMG and kinematics (cadence, velocity, stride length, step length)	No
Schwartz et al, 2004, ³⁰² St Paul	Pre-post cohorts compared with orthopedic surgery 2 years (8–24 months)	135 (73 M, 62 F) 8.1 years (3.1–43.3 years) Group 1: 27 (16 M, 11 F) Group 2: 66 (34 M, 32 F) Group 3: 18 (12 M, 6 F) Group 4: 24 (11 M, 13 F)	Diplegia (n = 135, all ambulatory)	Lower-limb spasticity (clinical examination) Range of motion (clinical examination)	3-D gait (lower-extremity kinematics and kinetics) Gait efficiency (normalized oxygen consumption) Global gait pathology (NI) Community walking and activity function (Gilette FAQ)	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Trost et al, 2008, ³⁰⁶ St Paul	Pre-post cohort 18 months	136 (81 M, 55 F) 7 years, 3 months ± 2 years, 1 month	Diplegia (n = 107), quadriplegia (n = 10), triplegia (n = 19) GMFCS Level I (n = 6), Level II (n = 64), Level III (n = 59), Level IV (n = 7)	Spasticity (Ashworth Scale)	3-D gait profile Gait pathology (Gillette Gait Index) Gait efficiency (oxygen consumption) Functional walking (Gillette FAQ)	Yes Transient complications resolving 6 weeks after surgery: bowel and bladder (n = 11), skin related (n = 9), wound healing (n = 8), headache (n = 6), paresthesia (n = 5), weakness (n = 4), miscellaneous related (n = 5), miscellaneous unrelated (n = 3)
Missouri						
Montgomery et al, 1992, ¹²⁶ St Louis	Pre-post cohort, multisite 2 years	14 (9 M, 5 F) 80.3 months ± 38.6 months (29–170 months)	Diplegia (n = 5), quadriplegia (n = 8), hemiplegia (n = 1) Nonambulatory (n = 7)	Spasticity (clinical observation) Range of motion (clinical examination)	Upper-extremity function (parent report) Speech (parent report) Motor and ambulatory abilities (clinical observation) Treatment satisfaction (family report)	Yes Hypesthesia with decreased sensation (n = 8), 2 with sensory problems before DR; periodic leg or low back muscle spasms, often associated with fatigue (n = 8), bladder problems (n = 4: 1 incontinence, 3 bladder control), hip dislocation (n = 2), scoliosis requiring thoracic lumbosacral orthosis (n = 1)
Park et al, 1993, ²⁹⁸ St Louis	Pre-post cohort	66 (NR)	Spastic CP (n = 66): independent ambulatory (n = 21), assisted ambulatory (n = 27), nonambulatory (n = 18)	--	Ambulatory status (clinical observation)	Yes No patient developed post-operative complication of cerebrospinal fluid leak, infection, motor weakness persistent for more than a few weeks, neurogenic bladder, sensory loss, or other neurologic deficits indicating damage to the conus medullaris or the ventral spinal root

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Craft et al, 1995, ²⁷¹ St Louis	Pre-post DR cohort (Group 1) compared with CP patients not undergoing DR (Group 2) and with normal age- and sex-matched controls (Group 3) 6 months	Group 1: 16 (NR) 7.7 years (5–12 years) Group 2: (NR) 7.5 years (5–12 years) Group 3: 24 (NR) 8.1 years (5–12 years)	Diplegia (n = 25)	--	Cognitive function (Woodcock-Johnson Psychoeducational Battery) Visual attention (Posner task)	No
Chicoine et al, 1996, ²⁶⁹ St Louis	Pre-post cohort 21 months (7–66 months)	90 (NR) 3.8 years (2–6 years)	Diplegia (n = 66) Quadriplegia (n = 22) Hemiplegia (n = 2)	Dorsiflexion tasks (clinical examination)	Gait (ambulatory score)	No
Engsberg et al, 1998, ³¹³ St Louis	Pre-post DR cohort (Group 1) compared with normal controls (Group 2) 8 months	Group 1: 19 (9 M, 10 F) 9 ± 4.2 years (4–16 years) Group 2: 6 (2 M, 4 F) 9 ± 4.5 years (4–17 years)	Diplegia	Spasticity Range of motion (clinical examination, Kin-Com dynamometer) Muscle strength	NR	No
Olree et al, 2000, ²⁹⁶ St Louis	Pre-post cohort (Group 1) compared with normal controls (Group 2) 8 months	Group 1: 27 (15 M, 12 F) 5.7 ± 3.7 years (2–16 years) Group 2: 7 (4 M, 3 F) 6.7 ± 5.3 years (3–17 years)	Diplegia (n = 27)	--	Synergistic movements of hip, knee, and ankle (2-D video system)	No
Ross et al, 2001, ³⁰⁰ St Louis	Pre-post cohort (Group 1) compared with normal controls (Group 2)	Group 1: 19 (9 M, 10 F) 9 ± 4.2 years (4–16 years) Group 2: 20 (10 M, 10 F) 9 ± 3.2 years (4–16 years)	Diplegia (n = 19): independent ambulatory (n = 9), assisted ambulatory (n = 9), nonambulatory (n = 1)	Muscle strength of quadriceps and hamstring (Kin-Com dynamometer)	Ambulatory status (clinical observation)	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Engsberg et al, 2002, ²⁷⁴ St Louis	Pre-post cohort (Group 1) compared with normal controls (Group 2) 8 months	Group 1: 24 (13 M, 11 F) 8 years, 5 months ± 4 years, 4 months (4–18 years) Group 2: 35 (19 M, 16 F) 8 years, 6 months ± 3 years, 1 month (4–15 years)	Diplegia (n = 24): independent ambulatory (n = 14), assisted ambulatory (n = 10)	Spasticity and muscle strength (Kin-Com dynamometer)	NR	No
Engsberg et al, 2004, ²⁷³ St Louis	Pre-post DR cohort (Group 1) compared with PT only (Group 2) and with age-matched normal controls (Group 3) 8 months	Group 1: 12 (8 M, 4F) 7.0 ± 4.7 years Group 2: 14 (7 M, 7 F) 7.4 ± 3.1 years Group 3: 20 (10 M, 10 F) 7.2 ± 3.4	46 (All ambulatory: 35 independent ambulatory, 11 assisted ambulatory) GMFCS Level I (n = 6), Level II (n = 8), Level III (n = 12)	Ankle range of motion, dorsiflexion, plantarflexion (Video PEAK 2-D sagittal plane)	NR	No
Abel et al, 2005, ²⁶⁴ St Louis	Pre-post DR cohort (Group 1) compared with untreated CP (Group 2) 10 months (9–12 months)	Group 1: 10 (NR) 8.5 years (3–14.9 years) Group 2: 10 (NR) 7.2 years (3–14.3 years)	Spastic CP (n = 20) GMFCS Level I (n = 2), Level II (n = 8), Level III (n = 10)	--	Gait 2-D biomechanical changes: joint positions, angular velocities, EMG response (6-VICON 370 camera motion analysis system) Kinematics (in-floor force measures)	No
Engsberg et al, 2006, ⁹⁴ St Louis	Pre-post DR cohort (Group 1) compared with CP cohort having PT only (Group 2) and with age-matched nondisabled children (Group 3) 2 years	Group 1: 37 (15 M, 16 F) 9 ± 5.3 years Group 2: 40 (19 M, 18 F) 9.7 ± 4.5 years Group 3: 40 (21 M, 19 F) 9.4 ± 3.4 years	Diplegia (all ambulatory) GMFCS Levels I–III (n = 77)	Spasticity, ankle-plantar reflex, knee flexor, hip adductor (Kin-Com dynamometer) Muscle strength (clinical examination)	Gross motor function (GMFM-66) Gait (6-camera KinTrak Motion software) temporal variables (speed, stride length, and cadence)	No

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Engsberg et al, 2007, ²⁷⁵ St Louis	Pre-post cohort 2 years	22 (10 M, 12 F) 8.5 ± 3.9 years	Diplegia (all ambulatory: independent (n = 19), assisted (n = 3) GMFCS Level I (n = 11), Level II (n = 5), Level III (n = 6)	Spasticity (Kin-Com dynamometer) Muscle strength (clinical examination)	Gross motor function (GMFM-66) Gait (6-camera HiRes system, KinTrak Motion software)	No
Reynolds et al, 2011, ⁹⁹ St Louis	Pre-post adult cohort 4 months	21 (15 M, 6 F) 26 years (18–39 years)	Diplegia (n = 21, all ambulatory) GMFCS Level I (n = 15), Level II (n = 28)	Spasticity (Modified Ashworth Scale) Passive range of motion (goniometer)	GMFM Ambulation (patient report) Basic ADL (Katz Basic ADL scale), Instrumental ADL (Lawton scale) Quality of life (patient report)	No
New York						
Moroto et al, 1995, ²⁹⁵ New York	Pre-post cohort Group 1: L2–S1 roots tested Group 2: L2–S2 roots tested Group 3: L2–S2 roots tested and modified sectioning 6 months	109 (NR) Group 1: 15 (NR) 6.1 years (3–26 years) Group 2: 62 (NR) 5.1 years (2–14 years) Group 3: 32 (NR) 3.9 years (2–8 years)	--	Spasticity gastrocnemius, hamstring (Modified Ashworth Scale)	--	No
Gigante et al, 2013, ²⁷⁹ New York	Pre-post cohort 16 months	42 (23 M, 19 F) 10.2 years (3–21 years)	Diplegia (n = 17), quadriplegia (n = 25) GMFCS Level I (n = 11), Level II (n = 5), Level III (n = 8), Level IV (n = 3), Level V (n = 9), unknown (n = 6)	Upper- and lower-extremity spasticity (Modified Ashworth Scale)	Functional change in upper limbs (family report)	No
Ohio						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Fukuhara et al, 2000, ²⁷⁷ Cleveland	Pre-post cohort	36 (27 M, 9 F) 6.5 years (2.3–16.2 years)	Spastic cerebral palsy (n = 36): ambulatory with or without assistance (n = 18), crawlers (n = 13), nonambulatory (n = 5)	Spasticity (Modified Ashworth Scale) Passive range of motion	--	Yes Wound infections requiring reclosure (n = 2), no persistent neurologic deficits but transient bilateral leg hyperesthesia (n = 1), prior urinary incontinence with transient worsening after DR (n = 1, pneumonia treated with intravenous antibiotics (n = 1)
Oregon						
Thomas et al, 1997, ³⁰⁵ Portland	Pre-post cohort 2 years	23 (13 M, 10 F) 6.4 years (3.7–10.9 years)	Spastic cerebral palsy (n = 23): independent ambulatory (n = 12), dependent ambulatory (n = 11)	Spasticity adductors, quadriceps, hamstrings, ankle-plantar flexors (Modified Ashworth Scale)	Gait 3-D patterns (5-camera VICON, joint kinematics) Hip stability	No
Buckon et al, 2004, ⁹³ Portland	Pre-post DR cohort (Group 1) compared with orthopedic surgery cohort (Group 2) (by parent choice) 2 years	Group 1: 18 (15 M, 3 F) 71.3 months (49–123 months) Group 2: 7 (4 M, 3 F) 78.6 months (95.1–132 months)	Group 1: Diplegia (n = 18): independent ambulatory (n = 11), dependent ambulatory (n = 7) Group 2: Diplegia (n = 7): independent ambulatory (n = 3), dependent ambulatory (n = 3), household ambulatory and community wheelchair (n = 1)	--	GMPM: 4 of the 5 GMFM scores Functional independence (PEDI)	No
Pennsylvania						
Albright et al, 1995, ²⁶⁶ Pittsburgh	Pre-post DR cohort (Group 1) compared with matched cohort undergoing continuous ITB therapy (Group 2) 12 months	Group 1: 38 (NR) Group 2: 38 (NR)	--	Upper-extremity spasticity (Ashworth Scale) Range of motion	Functional ability (parent report)	No
Texas						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Beck et al, 1993, ²⁶⁷ San Antonio	Pre-post cohort 12 months	14 (11 M, 3 F) (3–18 years)	Spastic CP (n = 14): independent ambulatory (n = 3), assisted ambulatory (n = 5), nonambulatory (n = 6)	--	Functional movement patterns (videotaped), fine motor skills (block building, bead threading, pegboard, writing, ball throwing), side sitting (score 1–5)	No
Utah						
Kan et al, 2008, ²⁸³ Salt Lake City	Pre-post DR cohort (Group 1) compared with age- and pre-operative motor function–matched ITB cohort (Group 2) 12 months	Group 1: 71 (NR) 5.6 years Group 2: 71 (NR) 4.0 years	Moderate or severe spastic CP GMFCS Levels III–V	Spasticity (Modified Ashworth Scale) Passive range of motion (hip abduction, knee flexion, ankle dorsiflexion)	GMFCS Treatment satisfaction (parent report) Subsequent orthopedic interventions	No
MacWilliams et al, 2011, ⁹² Salt Lake City	Pre-post DR cohort (Group 1) compared with age- and function-matched groups of children with CP receiving orthopedic surgery (Group 2) or not receiving surgery (Group 3)	Group 1: 8 (6 M, 2 F) 15 years, 4 months Group 2: (6 M, 3 F) 14 years, 6 months Group 3: (7 M, 2 F) 14 years, 10 months	Diplegia (n = 26) GMFCS Levels I or II	--	Gross motor function (GMFM-66), Functional gait (Gilette FAQ) Gait pattern 3-D motion analysis (10-camera VICON) Gait velocity, GDI, GVS Anthropometrics (body mass index)	No
Virginia						

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
Feger et al, 2015, ⁹¹ Charlotteville	Pre-post DR cohort (Group 1) compared with 3 other groups: Group 2: orthopedic bone surgery (with or without soft tissue); Group 3: orthopedic soft tissue surgeries only; Group 4: CP but no surgery	Group 1: 8 (5 M, 3 F) 7.8 ± 4.5 Group 2: 11 (5 M, 6 F) 9.9 ± 3.6 Group 3: 37 (22 M, 15 F) 9.0 ± 3.6 years Group 4: 38 (22 M, 16 F) 8.4 ± 3.2	Diplegia (n = 68), hemiplegia (n = 22), other (n = 5) GMFCS Level I (n = 30), Level II (n = 34), Level III (n = 30)	--	GMFM 3-D gait (temporal, kinematic), kinetics (movement, power)	No
Washington						
Dudgeon et al, 1994, ¹¹¹ Seattle	Pre-post cohort 12 months	29 (NR) 8.1 ± 4.1 years (3.7–22 years)	Diplegia (n = 20) Quadriplegia (n = 9)	--	Functional upper limb, reach, and coordination (clinical examination) Self-care and functional mobility (PEDI)	No
McLaughlin et al, 1994, ¹⁰³ Seattle	Pre-post cohort 12 months	34 (21 M, 13 F) Diplegia 8.0 ± 3.9 years (4.7–21.3 years) Quadriplegia 7.2 ± 3.4 (3.3–13.0 years)	Diplegia (n = 24): independent ambulatory (n = 10), assisted ambulatory (n = 12), nonambulatory (n = 2) Quadriplegia (n = 10): assisted ambulatory (n = 2), nonambulatory (n = 8)	Upper- and lower-limb spasticity (Ashworth Scale) Range of motion Deep tendon reflexes (0–4 scale)	GMFM Ambulation (clinical observation)	Yes No post-operative complications, such as infection, bleeding, or neurologic loss. Mild transient paresthesia of 2 weeks or less (several), dysesthesia for more than 2 months (n = 2), post-operative bladder incontinence resolving with antibiotic treatment or urethritis (n = 1), hip or back pain of several weeks' duration between 6 and 9 months after DR

Author, Year, and Country	Study Design Follow-Up (Range)	Patient, n (M, F) Age, Mean ± SD (Range) ^a	Anatomic Type, Functional Class GMFCS, Ambulatory Status	Pathophysiology Outcomes	Other Outcomes	Complications Reported
McLaughlin et al, 1998, ⁷³ Seattle	Single-site RCT 2 years	Group 1: DR + PT 21 (11 M, 10 F) 6.4 ± 3.0 years (3.2–14.4 years) Group 2: PT 17 (12 M, 5 F) 7.2 ± 4.5 years (3.2–18.1 years)	Diplegia (n = 38) Group 1: ambulatory for 50 feet (n = 5), nonambulatory for 50 feet (n = 16) Group 2: ambulatory for 50 feet (n = 8), nonambulatory for 50 feet (n = 9)	Spasticity (Ashworth Scale and electromechanical torque measures) gastrocnemius/Achilles tendon unit	GMFM	Yes No major adverse events <i>In Group 1:</i> 53 complications in 20 children included back pain (n = 6), leg pain (n = 10), fatigue (n = 2), weakness (n = 4), urinary problem (n = 3), brace problem (n = 3), emotional/behavioural problems during PT (n = 6), other musculoskeletal issue (n = 3), other miscellaneous problems (n = 1), and sensory problem (n = 4) <i>In Group 2:</i> 48 complications in 17 children included back pain (0), leg pain (n = 16), fatigue (n = 7), weakness (n = 3), urinary problem (0), brace problem (n = 1), emotional/behavioural problems during PT (n = 6), other musculoskeletal issue (0), other miscellaneous problems (n = 1), and sensory problem (0)
Graubert et al, 2000, ²⁸⁰ Seattle	Single-site RCT Group 1: DR Group 2: PT 12 months	Group 1: 18 (NR) 6.5 years (3.3–14.5 years) Group 2: 11 (NR) 7.4 years (3.0–17.5 years)	Diplegia (n = 29) Group 1: 18: independent ambulatory (n = 5), assisted ambulatory (n = 5), nonambulatory (n = 8) Group 2: 11: 3 independent ambulatory (n = 3), 5 assisted ambulatory (n = 5), 3 nonambulatory (n = 3)	Range of motion (hip, knee, ankle) kinematics	3-D gait (5-camera Motion Analysis System, Orthotrak software) Ambulatory status (clinical observation)	No
West Virginia						
Kaufman et al, 1994, ¹²⁵ Morgantown	Pre-post cohort 12 months	19 (12 M, 7 F) (2–10 years)	Diplegia (n = 19)	Spasticity (Ashworth Scale) Range of motion (score 0–4) Self-care (score 1–7)	Trunk support (score 0–5) Transitional movement (score 0–5) Ambulatory ability (score 1–5) Treatment satisfaction (family report)	No

Abbreviations: 2-D, two-dimensional; 3-D, three-dimensional; ADL, activities of daily living; BAD Scale, Barry-Albright Dystonia Scale; BTA, botulinum toxin type A injection; CNS, central nervous system; COPM, Canadian Occupational Performance Measure; CP, cerebral palsy; DR, dorsal rhizotomy; EMG, electromyography; EPG, electrophysiologic guidance; F, female; FAQ, Functional Assessment Questionnaire; FLACC scale, Face, Legs, Activity, Cry, Consolability scale; GDI, gait deviation index; GMAE, Gross Motor Activity Estimator; GMFCS, Gross Motor Functional Classification System; GMFM, Gross Motor Function Measure; GMFM-66, 66-item modification of GMFM-88; GMFM-88, 88-item gross motor function measure; GP, group (?); GVS, gait variable score; IPT, interpersonal therapy; ITB, intrathecal baclofen; M, male; MRC, Medical Research Council; NI, Normality Index; NR, not reported; NT, not tested; Ortho, orthopedic surgery; PEDI, Pediatric Evaluation of Disability Inventory; PEMG, polyelectromyography; PT, physical therapy; QUEST, Quality of Upper Extremity Skills Test; RCT, randomized controlled trial; RIC-FAS, Rehabilitation Institute of Chicago's Functional Assessment Scale; SMCS, Selective Motor Control Scale; VAS, visual analogue scale; VMFM, Vancouver Motor Function Measure; WeeFIM, Functional Independence Measure for Children.

Table A3. Clinical Studies Evaluating Long-Term Effectiveness of Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Canada						
Gul et al, 1999, ¹³⁸ Vancouver	Pre-post cohort 5 years (Of the 80 original cohort, 33 completed 5-year follow-up) 5 years	33 (18 M, 15 F) 4.4 years (2.2–9.7 years)	Spastic diplegia (n = 10) Quadriplegia (n = 10)	Spasticity hip adductor abduction (Modified Ashworth Scale) Range of motion (goniometer) Muscle strength, quadriceps strength (MRC)	Gross motor function (VMFM) Adjunct orthopedic surgeries	No
Ailon et al, 2015, ¹³⁹ Vancouver	Pre-post cohort 14.4 years (Of 142 with DR performed 10 years earlier, 44 completed 6-month, 5-year, and 10-year follow-up) 14.4 years	44 (NR) 4.5 years (2.9–7.7 years)	GMFCS Level II (n = 8), Level III (n = 16), Level IV (n = 17), Level V (n = 3)	Spasticity (modified Ashworth Scale) Active and passive range of motion (goniometer) Muscle strength quadriceps (MRC)	Gross motor function (VMFM/GMFM) Adjunct orthopedic surgeries	No
Mittal et al, 2002, ¹⁴⁴ Montreal	Pre-post cohort 5 years (Of 57 completing 6-month and 1-year assessments, 41 completed 3-year and 30 completed 5-year assessments) 5 years	41 (20 M, 21 F) 4.8 years (3.0–7.5 years)	Spastic diplegia (n = 36) Quadriplegia (n = 3) Triplegia (n = 2) Ambulatory status: independent ambulators (n = 16), assisted ambulators, n = 18) nonambulators (n = 7)	--	Functional performance and caregiver assistance (PEDI) ADLs Caregiver assistance (PEDI) Adjunct orthopedic surgeries, botulinum toxin injections, surgical complications	Yes
Mittal et al, 2002, ²⁹³ Montreal	Pre-post cohort 5 years (Of 70 in original cohort, 45 at 3-year, 25 at 5-year)	45 (26 M, 19 F) 4.5 years (3.0–7.4 years)	Spastic diplegia (n = 38), quadriplegia (n = 4), triplegia (n = 3)	--	Upper-extremity fine motor skills (PDMS) Adjunct orthopedic	Yes

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
	follow-up) 5 years		Ambulatory status: independent ambulators (n = 13) assisted ambulators (n = 22) nonambulators (n = 10)		surgeries, botulinum toxin injections	
Mittal et al, 2002, ¹³³ Montreal	Pre-post cohort (Of 93 meeting study criteria who completed 1-year post-operative assessment, 71 completed 3-year and 50 completed 3-year and 5-year assessments)	71 (43 M, 28 F) 5.2 years (3.0–10.7 years)	Spastic diplegia (n = 57), quadriplegia (n = 10), triplegia (n = 4) Ambulatory status: independent ambulators (n = 22), assisted ambulators (n = 27), nonambulators (n = 22)	Spasticity hip adductor (Modified Ashworth Scale) Range of motion (goniometer)	Developmental positions and transitional movements (Rusk/NYU form) Gross motor function (GMFM-88) Adjunct orthopedic surgeries Botulinum injections	Yes
Dudley et al, 2013, ¹³⁰ Montreal	Pre-post cohort 15 years (Of original cohort of 105, 102 had formal post-operative assessments, and 97, 62, 57, and 14 patients completed the 1-, 5-, 10-, and 15-year assessments, respectively) 15 years	105 (65 M, 40 F) 5 years (3.0–10.5 years)	Spastic diplegia (n = 65), triplegia (n = 5), quadriplegia (n = 11) GMFCS (n = 52): Level I (n = 11) Level II (n = 22) Level III (n = 14) Level IV (n = 5)	Spasticity (Ashworth Scale)	Gross motor function (GMFM-88) Functional benefit (PEDI) Adjunct orthopedic surgeries, Botulinum injections	No
Finland						

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Maenpaa et al, 2003, ²⁹⁰ Helsinki	Pre-post cohort (Group 1) compared with control group receiving PT (Group 2) 5 years (Of original cohorts of 44, 42 in Group 1 and 38 in Group 2 achieved their 3-year and 5-year post-procedural assessments) 5 years	44 (31 M, 13 F) Group 1: 22 (16 M, 6 F) 6 years (3–11 years) Group 2: 21 (15 M, 6 F) 6 years (3–14 years)	Group 1. Spastic diplegia (n = 14), spastic quadriplegia (n = 4) Group 2. Spastic diplegia (n = 17), spastic quadriplegia (n = 4)	Spasticity (Ashworth Scale): hip flexors, hip rotators, hip adductors, knee flexors, plantar flexors Muscle strength (Peacock's criteria)	Gross motor function classification (GMFCS) and Illinois–St Louis Scale	No
Italy						
Fasano et al, 1980, ²⁷⁶ Torino	Pre-post DR cohort (2–7 years)	80 (NR) Age NR	--	Spasticity Range of motion Reflexes and clonus	Functional effects on standing reaction, straightening reaction, equilibrium in sitting, standing and walking positions (clinical examination)	Yes
Netherlands						
Grunt et al, 2010, ¹³¹ Amsterdam	Pre-post cohort (Of 32 with MRI, 19 had follow-up assessments) 5 years, 4 months (1 year, 9 months–9 years)	36: 19 had MRI and follow-up (13 M, 6 F) (3 years, 11 months–10 years, 1 month)	Spastic diplegia (n = 19)	--	Gross motor function (GMFM-66)	No

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Van Schie et al, 2011, ¹³⁶ Amsterdam	Pre-post cohort 6 years ± 22 months	33 (21 M, 12 F) 6.7 ± 2 years (2.9–12.1 years)	Spastic diplegia (n = 33): ambulators (n = 14), nonambulators (n = 19) GMFCS Level I (n = 7), Level II (n = 7), Level II (n = 19)	Spasticity (Modified Tardieu scale)	Gross motor function (GMFM-66) Parental satisfaction Additional interventions (botulinum injections, orthopedic surgery)	Yes (long term)
Bolster et al, 2013, ¹²⁹ Amsterdam	Pre-post cohort 5 years and 10 years (28 completed 5-year and 20 completed 5- and 10-year assessments) 5 years and 10 years	29 (18 M, 11 F) 6 years, 4 months (2 years, 10 months–12 years, 1 month)	Spastic diplegia (n = 29): ambulators (n = 11), nonambulators (n = 18) GMFCS Level I (n = 7) Level II (n = 4) Level III (n = 18)	--	Gross motor function (GMFM-66) Additional interventions (orthopedic surgery, botulinum injections, casting)	Yes
South Africa						
Arens et al, 1989, ¹⁴⁷ Cape Town	Pre-post cohort (Of original 60, 53 had long-term assessments; parents of 35 children were interviewed) (3–7 years)	53 (38 M, 14F) (20 months–14 years)	Spastic cerebral palsy (n = 51) Dystonic athetoids (n = 2)	Spasticity (clinical examination) Muscle strength (clinical examination)	Gross motor function classification (Peabody criteria) Overall treatment benefits (physiotherapist report, parent report, children's report) Upper limb function (clinical examination) Subsequent orthopedic surgeries	Yes

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Peter et al, 1993, ¹⁴⁸ Cape Town	Pre-post DR cohort (extension of 1989 cohort); of 168 eligible, 110 patients country-wide had long-term follow-up	110 (80 M, 30 F) < 12 years (n = 80): 2–5 years (n = 41), 6–12 years (n = 39), ≥ 12 years (n = 30)	Spastic diplegia (n = 76) Spastic quadriplegia (n = 34)	Spasticity (clinical examination)	Gross motor function: sitting, standing, locomotion (clinical examination) Treatment satisfaction (physiotherapist report, parent report, children's report) Subsequent orthopedic surgeries	Yes
Subramanian et al, 1998, ³⁰⁴ Cape Town	Pre-post DR cohort (Group 1) compared with age-matched healthy children (Group 2) (from Vaughan et al cohort of 14 ambulatory children of original 29-child cohort) 10 years	Group 1: 11 (NR) 7.8 years at surgery (2.5–13.2 years), 18.4 years at gait assessment (12.9–24.1 years) Group 2: 12 (NR) 19 years	Spastic diplegia (n = 11, all ambulators)	Range of motion joint kinematics of hip and knee	Gait, 2-D evaluation, (temporospatial parameters [cadence, step length, velocity]) Subsequent orthopedic surgeries	No
Langerak et al, 2007, ²⁸⁴ Cape Town	Pre-post DR cohort (Group 1) compared with age-matched healthy children (Group 2) (from Vaughan et al cohort of 29 children, 14 ambulators) 10 years	Group 1: 11 (NR) 7.8 years At surgery (2.5–13.2 years) At gait assessment 18.4 years (12.9–24.1 years) Group 2: 12 (NR) 19 years	Spastic diplegia (n = 11, all ambulators)	Range of motion	Gait, 2-D evaluation (temporospatial parameters [cadence, step length, velocity])	No

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Langerak et al, 2008, ²⁸⁵ Cape Town	Pre-post DR cohort (Group 1) compared with age-matched healthy controls (Group 2) (from Vaughan et al cohort of 29 children, 14 ambulators) 20 years	Group 1: 13 (8 M, 5 F) 27.3 years (22–34 years) Group 2: 12 (7 M, 5 F) 27.8 years (22–34 years)	Spastic diplegia (n = 13): independent ambulatory (n = 11), assisted ambulatory (n = 2)	Range of motion of hip and knee	Gait, 2-D (DCR-TRV80E digital video camera) Gait temporal distance parameters Patient satisfaction Work, living, ADL status (self-report)	No
Langerak et al, 2009, ¹⁴⁰ Cape Town	Pre-post cohort 20 years	14 (8 M, 6 F) 27 years (22–33 years)	Spastic diplegia (n = 14), all ambulators) GMFCS Level I (n = 7), Level II (n = 3), Level III (n = 3), Level IV (n = 1)	Spasticity, joint stiffness Voluntary movement (selective motor control, muscle strength, and joint range of motion)	Gross motor function (Berman scale) Adjunct interventions (orthopedic surgeries, botulinum toxin, ITB pump, antispasmodic medication)	No
Langerak et al, 2011, ¹³⁷ Cape Town	Post cohort (47 patients were eligible; 31 of 37 contacted agreed to participate) 20 years	31 (18 M, 13 F) Age at surgery 5.2 years (2–27 years) Current age 26.8 years (21–44 years)	Spastic diplegia GMFCS Level I (n = 15), Level II (n = 11), Level III (n = 5)	--	Daily activities and social role (Assessment of Life Habits) Performance mobility FMS Patient satisfaction	No
Langerak et al, 2012, ²⁸⁶ Cape Town	Pre-post DR cohort (Group 1) compared with normal adults without history of orthopedic or neurologic pathology (Group 2) (of 47 eligible patients, 31 of 37 contacted agreed to participate) 21.2 years (17–26 years)	Group 1: 31 (18 M, 13 F) Age at surgery 5.2 years (2–27 years) Current age 26.8 years (21–44 years) Group 2: 43 (24 M, 19 F) 28.3 years (21–45 years)	Spastic diplegia (n = 31) GMFCS Level I (n = 15) Level II (n = 11) Level III (n = 5)	--	Gait 3-D (8-camera optoelectric system VICON) Subsequent interventions: spinal and orthopedic surgeries, ITB, intramuscular botulinum toxin	No
South Korea						

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Kim et al, 2001, ¹⁵⁴ Seoul	Pre-post cohort 4.2 years (1–9 years)	208 (NR) 5.9 years (2–23 years)	Spastic cerebral palsy (n = 198): independent ambulators (n = 43), assisted ambulators (n = 34), nonambulators (n = 131)	Spasticity (Ashworth Scale) Passive range of motion	Gross motor function classification (Peacock's criteria) Gait (videotape and electromyography kinematic motion analysis) Upper extremity function Trunk control	Yes
Sweden						
Nordmark et al, 2008, ¹³⁴ Lund	Pre-post cohort 5 years	35 (24 M, 11 F) 4.5 years (2.5–6.6 years)	Spastic diplegia (n = 35) GMFCS Level I (n = 1), Level II (n = 8), Level III (n = 10), Level IV (n = 15), Level V (n = 1)	Spasticity (Modified Ashworth Scale) Passive joint range of motion in lower extremities (goniometer)	Gross motor function (GMFM-88, GMFM-66) Skills, activities and caregiver assistance (PEDI) Subsequent orthopedic surgeries	Yes
Josenby et al, 2012, ¹³² Lund	Pre-post cohort (of the original 35 patients, 29 participated in 10-year follow-up) 10 years	29 (20 M, 9 F) Age at surgery 4.5 years (2.5–6.6 years) Current age 15 years	Spastic diplegics and quadriplegics GMFCS Level I (n = 1), Level II (n = 8), Level III (n = 7), Level IV (n = 12) Level V (n = 1)	Spasticity (Modified Ashworth Scale) Range of motion (goniometry)	Gross motor function (GMFM-66) Subsequent interventions (botulism toxin A and orthopedic surgery)	No
Josenby al, 2015, ¹⁴³ Lund	Pre-post cohort (24 Of the original 35 patients, 24 completed post-operative follow-up: 6 at 5-year, 24 at 10-year follow-up) 10 years	24 (19 M, 5 F) 4.1 years (2.5–6.4 years)	Spastic diplegia GMFCS Level I (n = 1), Level II (n = 7), Level III (n = 4), Level IV (n = 11), Level V (n = 1)	--	Functional skills, caregiver assistance for self-care, mobility (PEDI) Environmental adaptations (MAE scale)	No

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
Tedroff et al, 2011, ¹³⁵ Stockholm	Pre-post cohort 10 years	19 (15 M, 4 F) 4 years, 7 months ± 1 year, 7 months	Spastic diplegia (all ambulators)	Spasticity (Modified Ashworth Scale) Passive joint range of motion of hip, knee, and ankle	GMFM-88 Ambulatory status (Wilson Gait score) Subsequent orthopedic surgeries	Yes
United Kingdom						
McFall et al, 2015, ²⁹² Oswestry	Pre-post cohort Follow up at 10 and 16 years of age for girls and at 12 and 18 years of age for boys	17 (11 M, 6 F) (5–10 years)	Spastic diplegia (n = 15) and quadriplegia (n = 2) GMFCS Level II (n = 6) Level III (n = 9), Level IV (n = 2)	Range of motion, hip, knee, and ankle Muscle strength (MRC)	Gait 3-D (6- or 12-camera VICON motion analysis system) Gait Profile Score Subsequent orthopedic surgeries and botulinum toxin injections Weight gain (BMI)	No

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
United States						
Marty et al, 1995, ²⁹¹ Chicago	Pre-post DR cohort with or without subsequent orthopedic soft-tissue procedures (Group 1) compared with orthopedic soft-tissue surgery only cohort (Group 2) Average follow-up 4 years with 1-6 years follow-up Follow-up of 1–6 years for Group 1 and 1–7 years' follow-up for Group 2	Group 1: 50 (28 M, 22 F) 5 years (3–12 years) Group 2: 50 (29 M, 21 F) 5 years (1–13 years)	Spastic diplegia (n = 100) Group 1 community ambulators (n = 7), independent household ambulators (n = 10), assisted ambulators (n = 28), nonambulators (n = 5) Group 2: independent community ambulators (n = 17), independent household ambulators, (n = 4), assisted ambulators (n = 24) nonambulators (n = 5)	Passive range of motion (abduction and ankle dorsiflexion)	Ambulation classification status (clinical examination) Gait (videotape)	No
O'Brien et al, 2004, ¹⁴⁹ St Louis	Pre-post cohort 7.5 years (2-14 years)	52 (NR) (2–14 years) Group 1 aged 2–5 years (n = 35) Group 2 aged 6–14 years (n = 16)	Spastic quadriplegia (n = 77)	--	Gait score Parental treatment satisfaction	Yes

Author, Year, Country	Study Design Follow-Up Mean (Completed)	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Cerebral Palsy Classification Gross Motor Functional Status	Impairment Pathophysiology Outcomes	Other Outcomes	Adverse Outcomes Reported
O'Brien et al, 2005, ¹⁵⁰ St Louis	Pre-post cohort 7.5 years (5–9 years)	158 (NR) Age group 1: 2–3 years (n = 59) Age group 2: 4–7 years (n = 73) Age group 3: 8–14 years (n = 26)	Spastic diplegia (n = 158, all ambulatory independent or assisted)	--	Parental views on DR Subsequent orthopedic surgery	Yes
Hurvitz et al, 2013, ¹⁵¹ Ann Arbor, Mich, and Chicago	Post-DR cohort survey (of 271 eligible patients [adults > 21 years, older adolescents 16–20 years], 88 completed follow-up assessments) Follow-up at site 1, 21.8 ± 1.4 years and at site 2, 18.2 ± 3.0 years	88 (51 M, 37 F) Age at surgery 6.0 ± 3.9 years Age at follow-up 25.6 ± 4.8 years	GMFCS Level I (n = 6), Level II (n = 16), Level III (n = 20), Level IV (n = 32) Level V (n = 14)	--	Upper-extremity function (MACS) Gross motor function classification (GMFCS) Pain (NRS, 0–10) ADLs (self-report) Healthy perception (SF-36) Treatment satisfaction (patient report) Global or overall life satisfaction (Diener SWLS) Subsequent additional interventions for spasticity (orthopedic surgeries, baclofen pump, botulinum toxin or phenol injections, oral spasticity medications) and for scoliosis	No

Abbreviations: 3-D, three-dimensional; ADLs, activities of daily living; BMI, body mass index; CP, cerebral palsy; DR, dorsal rhizotomy; F, female; FMS, functional mobility scale; GMFCS, Gross Motor Function Classification System; GMFM, Gross Motor Function Measure; ITB, intrathecal baclofen; M, male; MACS, Manual Ability Classification System; MAE, Movement Analysis and Education Strategies; MRC, Medical Research Council; MRI, magnetic resonance imaging; NR, not reported; NRS, numerical rating scale; NYU, New York University; PDMS, Peabody Developmental Motor Scale; PED1, Pediatric Evaluation of Disability Inventory; PT, physical therapy; SD, standard deviation; SF-36, short-form 36; SWLS, Satisfaction with Life Scale; VMFM, Vancouver Motor Function Measure.

Table A4. Clinical Studies Evaluating Safety of Lumbrosacral Dorsal Rhizotomy for Spastic Cerebral Palsy

Author, Year, Town, Country	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Study Population	Study Follow-Up Duration Mean ± SD (Range) ^a	Surgical Intervention Period	Study Safety Objective (Primary, Secondary)
Canada					
Houle et al, 1998, ¹⁶¹ Montreal	40 (25 M, 15 F) 5.4 ± 2.1 years	Spastic cerebral palsy	6 months	January 1992 to September 1995	(Primary) To evaluate bladder function after DR
Golan et al, 2007, ¹⁶⁷ Montreal	98 (60 M, 38 F) 5.1 years (3.0–11 years)	Ambulatory and nonambulatory SCP NY grade I (n = 32), grade II (n = 52)	5.8 years (1.1–11.5 years)	September 1991 to June 2001	(Primary) To evaluate frequency of spinal deformities after DR and their risk factors
Steinbok et al, 1998, ¹⁵³ Vancouver	158 (NR) 5.3 ± 2.9 years	Spastic diplegia (n = 86), quadriplegia (n = 66), hemiplegia (n = 2), triplegia (n = 4)	30 ± 21 months	1987 to 1996	(Primary) To evaluate frequency and type of complications from DR occurring intra-operatively, peri-operatively, and after discharge
Hicdonmez et al, 2005, ²²⁹ Vancouver	82 (47 M, 35 F) 5.2 years (2.7–14.6 years)	Spastic diplegia (n = 44), quadriplegia (n = 35), triplegia (n = 2), hemiplegia (n = 1)	4 ± 2.7 years (0–12.1 years) 9 ± 3.2 years (3.9–16.2 years)	1987 to 2001	(Primary) To determine incidence of hip subluxation and risk factors after DR
Steinbok et al, 2005, ^{168,175} Vancouver	105 (56 M, 49 F) 5.2 years (2.7–14.6 years)	Spastic diplegia (n = 62), quadriplegia (n = 34), quadriplegia and intellectual delay (n = 9)	4.3 years (1.0–13.6 years)	1987 to 2001	(Primary) To evaluate incidence of scoliosis, kyphosis, and hyperlordosis after DR
Steinbok et al, 2009, ⁹⁵ Vancouver	22, 22	Spastic cerebral palsy	12 months	--	(Primary) To evaluate utility of electrophysiologically guided versus non-electrophysiologically guided DR on number of dorsal rootlets cut, outcomes, and complications
China					
Li et al, 2008, ¹⁶⁹ Guangzhou	61 (42 M, 9 F) 6.9 years (3–20 years)	--	6.3 years (5–9 years)	October 1992 to December 2002	(Primary) To evaluate incidence of lumbar spinal deformity after DR
France					
Parise et al, 1997, ¹⁶⁶ Lyon	10 (6 M, 4 F) (5–16 years)	Spastic diplegia	6 months	--	(Primary) To evaluate effect of DR on somatosensory function of lower limbs
Germany					

Author, Year, Town, Country	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Study Population	Study Follow-Up Duration Mean ± SD (Range) ^a	Surgical Intervention Period	Study Safety Objective (Primary, Secondary)
Floeter et al, 2014, ²²⁶ Berlin	33 (19 M, 14 F) 6.7 ± 2.4 years (2.7–11.8 years)	GMFCS Level I (n = 11), Level II (n = 16), Level III (n = 6)	18 ± 6 months	--	(Primary) To evaluate changes in hip geometry after DR
Funk et al, 2016, ⁶⁷ Berlin	132 7.2 ± 2.9 (2.7–17.1 years)	Ambulatory spastic cerebral palsy GMFCS Levels I–III	33 months (12–81 months)	January 2007 to February 2015	(Primary) To evaluate effect of single-level DR on development of scoliosis in ambulatory children with cerebral palsy
Hong Kong					
Chan et al, 2013, ²²⁵ Hong Kong	53 (30 M, 23 F) 7.9 ± 2.2 years	Spastic diplegia (n = 45), triplegia (n = 2), quadriplegia (n = 3) GMFCS Level I (n = 15), Level II (n = 9), Level III (n = 33), Level IV (n = 5), Level V (n = 2)	5.3 years	2003 to 2010	(Primary) To evaluate effect of DR and other factors on hip development in Asian children
Chiu et al, 2014, ¹⁵⁹ Hong Kong	54 7.7 years (4–15 years)	Spastic diplegia (n = 49), triplegia (n = 2), quadriplegia (n = 3)	8.4 ± 3.6 months	June 2003 to August 2010	(Primary) To evaluate effect of DR on bladder function and urodynamic findings
Netherlands					
Van Schie et al, 2011, ¹³⁶ Amsterdam	33 6.7 ± 2 years	Ambulatory cerebral palsy	3 years	August 1998 to December 2005	(Secondary) To investigate occurrence of side effects, such as spinal deformity and hip migration, need for additional treatment (botulinum type A or orthopedic surgery), and parental satisfaction
South Africa					
Peter et al, 1990, ¹⁷⁵ Cape Town	55 6.5 years (2–16 years)	Spastic diplegia (n = 39), quadriplegia (n = 14), severe dystonia (n = 2)	4.5 years (1–7 years)	--	(Primary) To determine incidence of spinal deformity after multiple-level laminectomy for DR
Peter et al, 1993, ¹⁷⁴ Cape Town	99	Spastic diplegia and quadriplegia	--	Since 1981	(Primary) To determine incidence of spondylolysis and spondylolisthesis after 5-level DR
Peter et al, 1993, ¹⁴⁸ Cape Town	110	Spastic diplegia (n = 76) and quadriplegia (n = 34)	10 years	1981 to 1991	(Secondary) To report on complications including tone, sensory disturbance, and spinal abnormalities
Langerak et al, 2009, ¹⁷³ Cape Town	30 (17 M, 13 F) 5.2 ± 5 years (2–27 years)	Spastic diplegia	4 ± 2 years (1–8 years)	1981 to 1991	(Primary) To evaluate incidence of spinal abnormalities in spastic diplegia after DR

Author, Year, Town, Country	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Study Population	Study Follow-Up Duration Mean ± SD (Range) ^a	Surgical Intervention Period	Study Safety Objective (Primary, Secondary)
			21.4 ± 3 years (17-26 years)		
South Korea					
Kim et al, 2001, ¹⁵⁴ Seoul	208 5.9 years (2–23 years)	Spastic cerebral palsy	4.2 years (1-9 years)	1990 to 1999	(Secondary) To evaluate effects of DR on functional ability and adverse effects
Kim et al, 2002, ¹⁵⁵ Seoul	200 (NR) 5.9 years (2–23 years)	Spastic cerebral palsy	4 years (1–9 years)	1989 to 1998	(Primary) To evaluate effects of various rootlet sectioning techniques on adverse outcomes including hypotonia, bladder dysfunction, spinal instability, and hip migration
United States					
California					
Van De Wiele, 1996, ¹⁵⁸ Los Angeles	102 5.2 years (3–15 years)	Spastic diplegia (n = 62), quadriplegia (n = 32), other paraplegia (n = 6) Ambulator (n = 76), nonambulator (n = 26)	Peri-operative	1986 to 1991	(Primary) To evaluate incidence and clinical relevance of adverse peri-operative events and their potential risk factors after DR
Florida					
Turi et al, 2000, ¹⁷² Gainesville	47 (28 M, 19 F) 6.8 years	Ambulators and nonambulators	5.3 years (2–9 years)	1987 to 1995	(Primary) To evaluate type and rates of spinal deformities after DR, origin of deformities, and suggested treatment
Massachusetts					
Mooney et al, 1999, ¹⁷⁸ Boston and Salem, NC	6	Spastic quadriplegia (n = 5), diplegia (n = 1) Nonambulator (n = 5), ambulator (n = 1)	--	--	(Primary) To describe cases of spinal deformity occurring after DR
Michigan					
Silva et al, 2012, ²³¹ Ann Arbor	69 (40 M, 29 F) 6 years, 11 months (30–220 months) 50 (27 M, 23 F) 9 years, 8 months (37–222 months)	Spastic quadriplegia, nonambulator (n = 69)	130 months 65 months	1988 to 2002	(Primary) To compare hip dislocation and containment procedures between DR and ITB in nonambulators
Minnesota					

Author, Year, Town, Country	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Study Population	Study Follow-Up Duration Mean ± SD (Range) ^a	Surgical Intervention Period	Study Safety Objective (Primary, Secondary)
Spiegel et al, 2004, ¹⁷¹ Minneapolis	79 (47 M, 32 F)	Spastic diplegia (n = 54), triplegia (n = 12), quadriplegia (n = 13) Community ambulator (n = 64), household ambulator (n = 8), ambulator (n = 6), nonambulator (n = 1)	5.8 years	Since 1991	(Primary) To evaluate frontal and sagittal spinal alignment to determine prevalence of scoliosis, thoracic hyperkyphosis, lumbar hyperlordosis, and spondylolisthesis in ambulatory cerebral palsy after DR
Missouri					
Montgomery et al, 1992, ¹²⁶ St Louis	14 (9 M, 5 F) 80.3 ± 38.6 months (29–170 months)	Spastic quadriplegia (n = 8), spastic diplegia (n = 5), spastic hemiplegia (n = 1)	27.5 ± 11.6 months (14 – 59 months)	--	(Secondary) To evaluate changes in sensory status, bladder function, orthopedic surgeries, and family perception of DR
Park et al, 1994, ²³⁰ St Louis	67 (2–11 years) Younger (2–4 years), older (5 – 11 years)	Spastic diplegia (n = 67)	6–10 months (n = 20) 15–46 months (n = 46)	July 1987 to April 1990	(Primary) To evaluate effect of DR on hip stability
Heim et al, 1995, ²²⁸ St Louis	45 (22 M, 23 F) 5 years, 1 month (2–9 years)	Spastic quadriplegia	20 months (7 – 50 months)	August 1987 to November 1990	(Primary) To evaluate changes in lateral hip migration after DR and relationship to age and baseline measure of lateral migration
New York					
Abbott et al, 1992, ¹⁵⁶ 1993, ¹⁵⁷ New York	250	Spastic diplegia, quadriplegia	--	Since 1986	(Primary) Peri-operative and long-term complications after DR and any protective measures implemented
Deletis et al, 1992, ¹⁶⁰ New York	31 (20 M, 11 F) 4.3 years (2–17 years)	Spastic cerebral palsy	Peri-operative	--	(Primary) To evaluate effect of intraoperative monitoring technique to reduce risk of bladder dysfunction
Huang et al, 1997, ¹⁶² New York	114 (72 M, 42 F) 3.8 years	Spastic cerebral palsy	Peri-operative	1991 to 1995	(Primary) To evaluate effectiveness of intraoperative technique to minimize risk of bowel, bladder, and sexual dysfunction
Lang et al, 1994, ¹⁶³ New York	85 4.8 ± 0.2 years	Spastic cerebral palsy	6 months	1986 to 1991	(Primary) To evaluate residual spasticity and bladder function with and without inclusion of S2 dorsal rootlets

Author, Year, Town, Country	Patients, n (F, M) Age, Mean ± SD (Range) ^a	Study Population	Study Follow-Up Duration Mean ± SD (Range) ^a	Surgical Intervention Period	Study Safety Objective (Primary, Secondary)
Sweetser et al, 1995, ¹⁶⁴ New York	34 (19 M, 15 F) (3.0–9.3 years)	Spastic diplegia (n = 24), quadriplegia (n = 9), hemiplegia (n = 1)	--	November 5, 1990, to January 20, 1994	(Primary) To evaluate effects of DR on urinary tract symptoms and bladder function
North Carolina					
Greene et al, 1991, ²²⁷ Chapel Hill	6 3.9–3.8 years	Spastic diplegia (n = 1), quadriplegia (n = 5)	--	--	(Primary) To describe progression of hip subluxation after DR
Oregon					
Johnson et al, 2004, ¹⁷⁰ Portland	34 6 years (3.7–10.9 years)	Ambulatory spastic diplegia	5 years	January 1990 to July 1997	(Primary) To assess incidence of spinal deformity in ambulatory spastic diplegia and whether laminoplasty offered any advantages over laminectomy
Texas					
Crawford et al, 1996, ¹⁷⁶ Dallas	2 (2 M) 13-year-old 7-year-old	Spastic quadriplegia	1 year 8 months, 5 years-	--	(Primary) To describe cases of severe lumbar lordosis occurring after DR
Utah					
Gooch et al, 1996, ¹⁷⁷ Salt Lake	2 (2 F) 8-year-old, 11-year-old	Spastic diplegia	4 years, 3 years	1989 to 1990	(Primary) To describe cases of spinal stenosis associated with abnormal gait after DR
Washington					
McLaughlin et al, 2005, ¹⁶⁵ Seattle	Cerebral palsy (n = 62) 9.1 ± 4.0 years Normal control (n = 65) 7.8 ± 3.9 years	Spastic diplegia (n = 34), quadriplegia (n = 14), left hemiplegia (n = 6), right hemiplegia (n = 4), athetoid quadriplegia (n = 3), hypotonia (n = 1)	--	--	(Primary) To evaluate feasibility of lower extremity sensory testing in children and comparing sensory changes after DR with children with cerebral palsy not treated by DR and with normal controls

Abbreviations: DR, dorsal rhizotomy; f, female; m, male; n, number; NR, not reported; SCP NY, Surveillance of Cerebral Palsy in New York; SD, standard deviation

Appendix 3: Evidence Quality Assessment

In considering the quality of evidence, our first consideration was study design; we started with the assumption that RCTs are high quality, whereas observational studies are low quality. We then took into account five additional factors—risk of bias, inconsistency, indirectness, imprecision, and publication bias. Limitations in these areas resulted in downgrading the quality of evidence. Finally, we considered three main factors that may raise the quality of evidence: the large magnitude of effect, the dose-response gradient, and any residual confounding factors. For more detailed information, please refer to the latest series of GRADE articles.³¹⁴

As stated by the GRADE Working Group, the final quality score can be interpreted using the following definitions:

High	We are very confident that the true prognosis (probability of future events) lies close to that of the estimate
Moderate	We are moderately confident that the true prognosis (probability of future events) is likely to be close to the estimate, but there is a possibility that it is substantially different
Low	Our confidence in the estimate is limited: the true prognosis (probability of future events) may be substantially different from the estimate
Very Low	We have very little confidence in the estimate: the true prognosis (probability of future events) is likely to be substantially different from the estimate

Table A5: GRADE Evidence Profile for Lumbosacral Dorsal Rhizotomy for Spastic Cerebral Palsy

Number of Studies (Design) [Initial Grade]	Risk of Bias ^a	Inconsistency	Indirectness	Imprecision	Publication Bias	Upgrade Considerations	Quality
Gross Motor Function, Short Term ≤ 2 years							
3 RCTs (DR vs Dr + PT) [High]	No serious limitations	No serious limitations	No serious limitations	Serious limitations (-1) ^b	Unevaluated	NA	⊕⊕⊕ Moderate
9 observational pre-post cohort studies [Low]	No serious limitations See Table 5a	No serious limitations	No serious limitations	No serious limitations	Unevaluated	Statistically significant large magnitude of effect (+1) ^c	⊕⊕⊕ Moderate
Gross Motor Function, Long Term > 2 years							
10 observational pre-post longitudinal cohort studies [Low]	No serious limitations See Table 5b	No serious limitations	No serious limitations	No serious limitations	Unevaluated	Statistically significant very large magnitude of effect (+1) ^d	⊕⊕⊕ Moderate
Functional Independence, Short Term ≤ 2 years							
10 observational pre-post cohort studies [Low]	No serious limitations See Table 5c	No serious limitations	No serious limitations	No serious limitations	Unevaluated	Statistically significant large magnitude of effect (+1) ^e	⊕⊕⊕ Moderate
Functional Independence, Long Term > 2 years							
4 observational pre-post longitudinal cohort studies [Low]	No serious limitations See Table 5d	No serious limitations	No serious limitations	Serious limitations (-1) ^f	Unevaluated	Statistically significant very large magnitude of effect (+1) ^g	⊕⊕ Low

Abbreviations: DR, dorsal rhizotomy; GRADE, Grading of Recommendations Assessment, Development, and Evaluation; NA, not applicable; PT, physical therapy.

^aBias was assessed in individual observational pre-post studies for several criteria at pre-intervention, intervention, and post-intervention stages. Among criteria considered were prospective study design, eligibility criteria, interventions and co-interventions, outcome measurement, and follow-up. Although reporting of these factors varied in individual studies, overall data in reports were sufficient (most information was from studies of low risk) to evaluate bias as not having serious limitations.

^bMeasures of gross motor function in RCTs were limited by imprecision because of small sample sizes and restriction to higher-functioning subgroup of cerebral palsy patients with spastic diplegia.

^cUpgrade considerations for gross motor function in short term evaluated in observational studies were based on statistically and clinically significant improvements: large magnitude of effect with validated outcome measures and temporal relationship of improvements after surgery when mobility increased after lower-limb spasticity decreased.

^dUpgrade considerations for gross motor function in long term evaluated in observational studies were based on statistically and clinically significant improvements: very large magnitude of effect with validated outcome measures and temporal relationship of improvements after surgery when mobility increased with continued reduction in lower limb spasticity.

^eUpgrade considerations for functional independence in short term were based on statistically and clinically significant improvements: large magnitude of effect with validated outcome measures.

^fImprecision limitations with functional independence measures in long term were due to the few studies and small number of patients being followed.

^gUpgrade considerations for functional independence in long term were based on statistically and clinically significant improvements: very large magnitude of effect with validated outcome measures.

^hUpgrade considerations for caregiver assistance in short term were based on statistically and clinically significant improvements: large magnitude of effect with validated outcome measures.

Table A6: Risk of Bias in Observational Uncontrolled Pre-Post Intervention Studies

Table A6a: Short-Term Gross Motor Function

	Confounding	Selection or classification bias	Measurement bias	Follow-up bias	Missing data	Reporting bias	Other bias
GMFM							
Carraro et al, 2014 ⁹⁶	N	N	Y	N	N	N	UN
Chan et al, 2008 ⁹⁷	N	N	N	N	N	N	UN
Funk et al, 2015 ⁹⁸	N	N	N	N	N	N	UN
Hodgkinson et al, ¹⁰²	N	N	N	N	N	N	UN
McLaughlin et al, 1994 ¹⁰³	N	N	N	N	N	N	UN
Nordmark et al, 2000 ¹⁰⁴	N	N	N	N	N	N	UN
Reynolds et al, 2011 ⁹⁹	N	N	N	Y	N	N	UN
Sacco et al, 2000 ¹⁰⁰	N	Y	Y	N	N	N	UN
Van Schie et al, 2005 ¹⁰¹	N	N	N	N	N	N	UN

Abbreviations: GMFM, Gross Motor Function Measure; N, no; UN, unknown; Y, yes.

Table A6b: Long-Term Gross Motor Function

	Confounding	Selection or classification	Measurement bias	Follow-up bias	Missing data	Reporting bias	Other bias
Ailon et al, 2015 ¹³⁹	N	Y	N	Y	Y	N	UN
Bolster et al, 2013 ¹²⁹	N	N	N	N	N	N	UN
Dudley et al, 2013 ¹³⁰	N	N	N	N	Y	N	UN
Gul et al, 1999 ¹³⁸	N	N	Y	N	N	N	UN
Josenby et al, 2012 ¹³²	N	N	N	Y	N	N	UN
Langererak et al, 2009 ¹⁴⁰	N	Y	Y	N	N	N	UN
Mittal et al, 2002 ¹³³	N	N	N	N	Y	N	UN
Nordmark et al, 2008 ¹³⁴	N	Y	N	N	Y	N	UN
Tedroff et al, 2011 ¹³⁵	N	Y	N	N	N	N	UN
Van Schie et al, 2011 ¹³⁷	N	N	N	N	Y	N	UN

Abbreviations: GMFM, Gross Motor Function Measure; N, no; UN, unknown; Y, yes.

Table A6c: Short-Term Functional Status

	Confounding	Selection or classification bias	Measurement bias	Follow-up bias	Missing data	Reporting bias	Other bias
PEDI							
Buckon et al, 2004 ⁹³	N	Y	N	N	N	N	UN
Chan et al, 2008 ⁹⁷	N	N	N	N	N	N	UN
Dudgeon et al, 1994 ¹¹¹	N	Y	N	N	Y	N	UN
Nordmark et al, 2000 ¹⁰⁴	N	N	N	N	N	N	UN
Van Schie et al, 2005 ¹⁰¹	N	N	N	N	N	N	UN
WeeFIM							
Carraro et al, 2014 ⁹⁶	N	N	Y	N	N	Y	UN
Loewen et al, 1998 ¹¹⁷	N	N	Y	N	Y	N	UN
Nishida et al, 1995 ¹¹⁶	N	N	N	Y	N	N	UN
Steinbok et al, 2009 ⁹⁵	N	Y	Y	N	Y	N	UN
Tichy et al, 1995 ¹¹⁸	N	Y	N	N	Y	N	UN

Abbreviations: N, no; PEDI, Pediatric Evaluation of Disability Inventory; UN, unknown; Wee FIM, Functional Independence Measure for Children; Y, yes.

Table A6d: Long-Term Functional Status

	Confounding	Selection or classification bias	Measurement bias	Follow-up bias	Missing data	Reporting bias	Other bias
PEDI							
Dudley et al, 2013 ¹³⁰	N	Y	N	N	Y	N	UN
Josenby et al, 2012 ³¹⁵	N	N	N	Y	N	N	UN
Mittal et al, 2002 ¹⁴⁴	N	N	N	N	Y	N	UN
Nordmark et al, 2008 ¹³⁴	N	Y	N	N	Y	N	UN

Abbreviations: N, no; PEDI, Pediatric Evaluation of Disability Inventory; UN, unknown; Y, yes.

Table A7: McHarm Quality Assessment of Safety Based on Observational Studies

	Definitions	Collection period	Collection method	Reporting	Sampling	Follow-up	Overall Quality
Peri-operative							
Abbott et al, 1992 ¹⁵⁶	Y	Y	N	N	Y	Y	Low
Kim et al, 2002 ¹⁵⁵	Y	Y	N	Y	Y	Y	Mod
Steinbok et al, 1999 ¹⁵³	Y	Y	N	Y	Y	Y	Mod
Van De Wiele et al, 1996 ¹⁵⁸	Y	Y	N	Y	Y	Y	Mod
Bladder Dysfunctions							
Chiu et al, 2014 ¹⁵⁹	Y	Y	Y	Y	N	Y	Mod
Deletis et al, 1992 ¹⁶⁰	Y	Y	Y	Y	Y	N	Low
Houle et al, 1998 ¹⁶¹	Y	Y	N	N	N	Y	Low
Huang et al, 1997 ¹⁶²	Y	Y	Y	N	N	Y	Low
Kim et al, 2002 ¹⁵⁵	Y	N	N	N	Y	N	Low
Lang et al, 1994 ¹⁶³	Y	Y	Y	N	N	Y	Low
Montgomery et al, 1992 ¹²⁶	N	Y	N	Y	N	Y	Low
Sweester et al, 1995 ¹⁶⁴	Y	Y	Y	Y	N	Y	Mod
Steinbok et al, 1998 ¹⁵³	Y	Y	N	Y	Y	Y	Mod
Sensory Abnormalities							
McLaughlin et al, 1993 ¹⁶⁵ , 2005	Y	Y	Y	Y	N	Y	Mod
Montgomery et al, 1992 ¹²⁶	Y	Y	N	Y	N	Y	Low
Parise et al, 1997 ¹⁶⁶	Y	N	Y	Y	Y	N	Low
Peter et al, 1993 ¹⁴⁸	Y	Y	N	Y	N	N	Low
Steinbok et al ¹⁵³ , 1998	Y	Y	N	Y	Y	Y	Mod
Hip Instability							
Chan et al ²²⁵ , 2013	Y	Y	Y	N	Y	Y	Mod
Floeter et al ²²⁶ , 2014	Y	Y	Y	Y	N	Y	Mod
Greene et al ²²⁷ , 1991	Y	N	Y	N	N	N	Low
Heim et al ²²⁸ , 1995	Y	Y	Y	Y	N	Y	Mod
Hicdonnez et al ²²⁹ , 2005	Y	Y	Y	N	N	Y	Low
Kim et al, ¹⁵⁵ 2002	Y	Y	Y	N	N	Y	Low
Park et al ²³⁰ , 1994	Y	Y	Y	Y	N	Y	Mod
Silva et al ²³¹ , 2012	Y	Y	Y	N	N	N	Low

Abbreviations; Mod, moderate; N, no (more bias); Y, yes (less bias)

	Definitions	Collection period	Collection method	Reporting	Sampling	Follow-up	Overall Quality
Scoliosis							
Funk et al, 2016 ⁶⁷	Y	Y	Y	Y	N	Y	Mod
Golan et al, 2007 ¹⁶⁷	Y	Y	Y	Y	N	N	Low
Johnson et al, 2004 ¹⁷⁰	Y	Y	Y	Y	N	Y	Mod
Kim et al, 2002 ¹⁵⁵	Y	Y	N	N	N	Y	Low
Langerak et al, 2009 ¹⁷³	Y	Y	Y	N	N	N	Low
Li et al, 2008 ¹⁶⁹	N	Y	Y	Y	N	N	Low
Peter et al, 1990 ¹⁷⁵	Y	Y	Y	N	N	N	Low
Spiegel et al, 2004 ¹⁷¹	Y	Y	Y	Y	N	Y	Mod
Steinbok et al, 2005 ¹⁶⁸	Y	Y	Y	N	N	Y	Low
Turi et al, 2000 ¹⁷²	Y	Y	Y	N	N	Y	Low
Van Schie et al, 2011 ¹³⁶	Y	Y	N	N	N	Y	Low
Kyphosis/Lordosis							
Golan et al, 2007 ¹⁶⁷	Y	Y	Y	N	N	Y	Low
Johnson et al, 2004 ¹⁷⁰	Y	Y	Y	N	N	Y	Low
Kim et al, 2002 ¹⁵⁵	Y	Y	Y	N	N	N	Low
Langerak et al, 2009 ¹⁷³	Y	Y	Y	N	N	N	Low
Li et al, 2008 ¹⁶⁹	Y	Y	Y	N	N	N	Low
Peter et al ¹⁷⁵ , 1990	Y	Y	Y	N	N	Y	Low
Spiegel et al ¹⁷¹ , 2004	Y	Y	Y	Y	N	Y	Mod
Steinbok et al ¹⁶⁸ , 2005	Y	Y	Y	N	N	Y	Low
Turi et al ¹⁷² , 2000	Y	Y	Y	N	N	N	Low
Spondylolysis/Spondylolisthesis							
Golan et al ¹⁶⁷ , 2007	Y	Y	Y	N	Y	Y	Mod
Johnson et al ¹⁷⁰ , 2004	Y	Y	N	Y	Y	Y	Mod
Kim et al ¹⁵⁵ , 2002	Y	Y	N	N	N	Y	Low
Langerak et al ¹⁷³ , 2009	Y	Y	Y	N	N	N	Low
Li et al ¹⁶⁹ , 2008	Y	Y	Y	N	Y	Y	Mod
Peter et al ¹⁷⁴ , 1993	Y	Y	Y	N	N	N	Low
Spiegel et al ¹⁷¹ , 2004	Y	Y	Y	N	Y	Y	Mod
Turi et al ¹⁷² , 2000	N	Y	Y	N	N	N	Low
Van Schie et al ¹³⁶ , 2011	Y	Y	N	Y	N	N	Low

Appendix 4: Budget Impact Analysis Inputs

Table A8: Total Net Cost of Dorsal Rhizotomy per Year Sensitivity Analyses

Variable	Cost per Patient(\$)	Number of Patients	Total Cost
Sensitivity analysis: 12 patients receiving dorsal rhizotomy per year			
Patients receiving dorsal rhizotomy	148,657 ^a	12	1,783,890
Patients assessed but ineligible to receive dorsal rhizotomy	620	7	4,338
Total cost			1,788,227
Sensitivity analysis: 6-week inpatient rehabilitation			
Patients receiving dorsal rhizotomy	76,815	9	691,337
Patients assessed but ineligible to receive dorsal rhizotomy	620	7	4,338
Total cost			695,674
Sensitivity analysis: postsurgical rehabilitation protocol same as standard care			
Patients receiving dorsal rhizotomy	14,901	9	134,111
Patients assessed but ineligible to receive dorsal rhizotomy	620	7	4,338
Total cost			138,449

^aNet cost of dorsal rhizotomy less standard care (\$151,470–\$2,813)

Appendix 5: Letter of Information and Consent Form



LETTER OF INFORMATION

SUMMARY:

Health Quality Ontario (HQQ) is conducting a formal assessment of **Selective Dorsal Rhizotomy**, to better understand whether this treatment should be funded by the healthcare system. An important part of this assessment involves speaking to families of patients with Cerebral Palsy, who may or may not be considering SDR. Having had this procedure is not necessary to be involved in this review. Our goal is to illuminate the lived-experience of patient and families with CP, their existing therapies, and the context around the SDR procedure – what families view as its possibilities, implications, and availability.

WHAT DO YOU NEED FROM ME?

- ✓ Willingness to share your story
- ✓ 30-60 minutes of your time for a phone or in-person interview
- ✓ Permission to audio- (not video-) record the interview

WHY DO YOU NEED THIS INFORMATION?

Health Quality Ontario (HQQ) is conducting a [Health Technology Assessment of Dorsal Rhizotomy for Lower Extremity Spasticity in Children with Cerebral Palsy](#). As part of HQO's core function to promote health care supported by the best evidence available, established scientific methods are used to analyze the evidence for a wide range of health interventions, including diagnostic tests, medical devices, interventional and surgical procedures, health care programs and models of care. These analyses may be informed and complemented by input from a range of individuals, including patients and clinical experts, and serve as the basis recommendations about whether health care interventions should be publicly funded or not.

The perspective that you share will be useful to help provide context to the day-to-day realities of families experiencing cerebral palsy and the decisions they face in terms of therapies. The ultimate goal of the project is to provide recommendations to the Ontario Health Technology Assessment Committee who advises the Ontario Ministry of Health and Long-term Care on the appropriateness of funding.

WHAT YOUR PARTICIPATION INVOLVES

If you agree to enroll, you will be asked to participate in an interview or focus group conducted by HQO staff. The interview or focus group will likely last 30-60 minutes. The session will be conducted in a private location and will be audio-taped. The interviewer will ask you questions about your lived experience with cerebral palsy, existing therapies, and potentially dorsal rhizotomy, if your child has undergone this procedure or may be considering it.

Participation is voluntary. You may refuse to participate, refuse to answer any questions or withdraw before your interview. Withdrawal will in no way affect care you receive.

CONFIDENTIALITY

All information collected for the review will be kept confidential and privacy will be protected except as required by law. The results of this review will be published, however no identifying information will be released or published. Any records containing information from your interview will be stored securely.

RISKS TO PARTICIPATION:

There are no known physical risks to participating. Some participants may experience discomfort or anxiety after speaking about their lived experience. If this is the case, please contact any staff.

HEALTH QUALITY ONTARIO STAFF:

Mark Weir

Senior Program Analyst, Patient, Family and Public Engagement

Tel: (416) 323-6868 x. 653, Email: Mark.Weir@hqontario.ca

David Wells

Program Analyst, Patient, Family and Public Engagement

Tel: (416) 323-6868 x710 Email: David.Wells@hqontario.ca

Consent and Release Form

This form is to be read and completed in accordance with the following instructions before it can be signed.

1. I, _____ allow Health Quality Ontario (Ontario Health Quality Council) to use to inform the development of an evidence based review:

Check off all appropriate boxes:

- a) a recording of my voice
- b) a quotation or summary of my opinion that I expressed during an interview
- c) name & contact information

2. Please read the following paragraphs before affixing your signature under section 3.

- a) Personal information collected pursuant to, and on this form, will be used for purposes described on this form and for no other purpose. Health Quality Ontario (Ontario Health Quality Council) acknowledges that you have provided this personal information freely and voluntarily. If you have any questions about this collection of this personal information, contact:

Amy Lang
 Director, Patient, Caregiver and Public Engagement
 Tel: (416) 323-6868, ext. 610, E-mail: amy.lang@hqontario.ca

- b) By signing this form as indicated below, you agree to hereby release and forever discharge the Health Quality Ontario (Ontario Health Quality Council), its officers, employees, agents and representatives from any and all claims, demands, expenses, actions, causes of action and for any and all liability howsoever caused, arising out of, or in any way related to the collection, use and disclosure of information, recordings and images authorized to be collected pursuant to, or on this form.
- c) By signing this form as indicated below, you agree to forever waive any and all rights that you may have to the use of information and recordings that are authorized to be collected pursuant to, or on this form; and you acknowledge that all information, recordings and images shall hereafter remain the exclusive property of the Health Quality Ontario (Ontario Health Quality Council).

3. Signature is to be affixed in the appropriate space provided below.

I have read this form after it was completed, I understand and agree to be bound by its contents, and I am eighteen (18) years of age or over.

Signature _____

Print name _____

Date _____

Appendix 6: Interview Guide



Interview for Dorsal Rhizotomy HTA

Intro

Explain HQO purpose, HTA process, and purpose of interview
History of CP diagnosis and various treatments (general only)

Lived-Experience

What is the day-to-day routine, quality of life?
What is the impact on caregiver (parent) on caring for child?
What is the impact on siblings?
How much self-care is involved, independence?

Current Therapies

What current therapies/treatments does the child with CP undergo and their impact on child + caregiver?
Is accessibility to therapies an issue (are you able to take advantage of all potential therapies)?
What are the expectations for these therapies longterm?
Are there any side-effects or risks with the therapies that you have experienced? Was it difficult to weigh up potential benefits and risks when deciding on which therapies to go with?
Are there any other therapies you are considering? If so, how do you go about making a decision?

Dorsal Rhizotomy

What have you heard of SDR?
Do you feel you have enough information to understand risk/benefits and make a decision?
What are your expectations of the procedure?
What was the procedure like? What was the challenges to getting it done?
What is your understanding of the therapies needed afterwards? Is this different than you expected?
Are there any unexpected consequences from the procedure?

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About Health Quality Ontario

Health Quality Ontario is the provincial advisor on the quality of health care. We are motivated by a single-minded purpose: **Better health for all Ontarians.**

Who We Are.

We are a scientifically rigorous group with diverse areas of expertise. We strive for complete objectivity, and look at things from a vantage point that allows us to see the forest and the trees. We work in partnership with health care providers and organizations across the system, and engage with patients themselves, to help initiate substantial and sustainable change to the province's complex health system.

What We Do.

We define the meaning of quality as it pertains to health care, and provide strategic advice so all the parts of the system can improve. We also analyze virtually all aspects of Ontario's health care. This includes looking at the overall health of Ontarians, how well different areas of the system are working together, and most importantly, patient experience. We then produce comprehensive, objective reports based on data, facts and the voice of patients, caregivers and those who work each day in the health system. As well, we make recommendations on how to improve care using the best evidence. Finally, we support large scale quality improvements by working with our partners to facilitate ways for health care providers to learn from each other and share innovative approaches.

Why It Matters.

We recognize that, as a system, we have much to be proud of, but also that it often falls short of being the best it can be. Plus certain vulnerable segments of the population are not receiving acceptable levels of attention. Our intent at Health Quality Ontario is to continuously improve the quality of health care in this province regardless of who you are or where you live. We are driven by the desire to make the system better, and by the inarguable fact that better has no limit.

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Health Quality Ontario
130 Bloor Street West, 10th Floor
Toronto, Ontario
M5S 1N5
Tel: 416-323-6868
Toll Free: 1-866-623-6868
Fax: 416-323-9261
Email: EvidenceInfo@hqontario.ca
www.hqontario.ca

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