

# Outcomes of Selective Dorsal Rhizotomy in Non-Walking Children with Spastic Cerebral Palsy

## *Resultados da Rizotomia Dorsal Seletiva em Crianças Não Deambuladoras com Paralisia Cerebral Espástica*

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### ABSTRACT

**Introduction:** There are divergent opinions on the use of selective dorsal rhizotomy (SDR) to treat spasticity in non-walking children. **Objective:** to investigate the neurological outcomes and perioperative complications after lumbar SDR in non-walking children with spastic cerebral palsy (CP). **Methods:** A total of 59 non-walking children with spastic CP, between 3 and 9 year-old, and submitted to lumbar SDR with surgical access to the medullary cone were submitted. The patients were followed-up by a multidisciplinary team and underwent intensive physical rehabilitation after surgery. Functional results were measured using the modified Ashworth and GMFM-88 scales 10 months after surgery. **Results:** The muscle relaxation documented in the lower limbs by the modified Ashworth scale resulted in significant functional improvement in dimensions A and B of the GMFM-88 scale. Peri-operative complications were present in 19 patients (32.2%), and consisted of urinary retention ( $n = 5$ ), delayed wound healing ( $n = 2$ ), fever of unknown cause ( $n = 1$ ), pain severe ( $n = 11$ ), and dysesthesias and spasms ( $n = 9$ ). **Conclusions:** Lumbar SDR in non-walking children with spastic CP promoted muscle relaxation in the lower limbs with significant improvement in gross motor functions, and mild and temporary perioperative complications.

**Keywords:** Cerebral palsy; Spasticity; Selective dorsal rhizotomy; Intraoperative neurophysiological monitoring; Physical rehabilitation

### RESUMO

**Introdução:** Há opiniões divergentes sobre o uso da rizotomia dorsal seletiva (RDS) para tratamento da espasticidade em crianças que não caminham. **Objetivo:** Investigar os resultados neurológicos e as complicações peri-operatórias após RDS lombar em crianças não

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*deambuladoras com paralisia cerebral espástica (PC). Métodos:* Foram incluídas no estudo 59 crianças não deambuladoras com PC espástica, entre 3 e 9 anos, submetidas a RDS lombar com acesso cirúrgico ao cone medular. Os pacientes foram acompanhados por equipe multidisciplinar e submetidos à reabilitação física intensiva após a cirurgia. Os resultados funcionais foram medidos com 10 meses de pós-cirúrgico, através das escalas de Ashworth modificada e GMFM-88. **Resultados:** O relaxamento muscular documentado nos membros inferiores pela escala de Ashworth modificada resultou em melhora funcional significativa nas dimensões A e B da escala GMFM-88. Complicações peri-operatórias estiveram presentes em 19 pacientes (32,2%) e consistiram em: retenção urinária (n = 5), cicatrização retardada (n = 2), febre de causa desconhecida (n = 1), dor intensa (n = 11) e disestesias e espasmos (n = 9). **Conclusão:** A RDS lombar em crianças não deambuladoras com PC espástica promoveu relaxamento muscular nos membros inferiores com melhora significativa da função motora grossa, com complicações peri-operatórias leves e temporárias.

**Palavras-Chave:** Paralisia cerebral; Espasticidade; Rizotomia dorsal seletiva; Monitoramento neurofisiológico intraoperatório; Reabilitação física

## INTRODUCTION

Cerebral palsy (CP) represents a broad spectrum of neurological changes from a non-progressive lesion on the immature brain, and is conceptually associated with disorders of motor development that lead to impairments in the acquisition of motor skills. A CP is classified according to the Gross Motor Function Classification System (GMFCS), levels I to V, and higher levels indicate worse physical functioning<sup>1</sup>.

Children with GMFCS levels IV or V have spastic tetraparesis, and unable to walk, and often have epilepsy and intellectual disability<sup>2</sup>. Spasticity is a common motor abnormality for these children, and responsible for the appearance of muscle-ligament shortening, joint deformities and structural changes in the spine<sup>3</sup>. Therefore, interventions are needed to promote muscle relaxation, in order to improve positioning, facilitate daily care, control pain and slow down the progression of musculo skeletal deformities. Therapeutic options include medications such as oral baclofen and botulinum toxin, which need to be used in high doses and can cause significant adverse effects<sup>4</sup>. The best results are associated with the use of continuous intrathecal baclofen (ITB) and the performance of selective dorsal rhizotomy (SDR).

Intrathecal baclofen involves surgical placement of a catheter through a small opening in the lumbar dura, with subsequent catheter connection to an implanted baclofen pump, usually placed in the subcutaneous tissue of the abdomen. Although offering an effective way to continuously deliver baclofen directly to the central nervous system, the use of ITB has the following

disadvantages: high risk of complications, high cost of treatment, and need for frequent visits to the hospital to make adjustments to the equipment<sup>5</sup>. ITB has historically been reserved for non-walking children with spastic tetraparesis, usually with the goal of reducing spasticity as a way to improve comfort and decrease the caregiver burden<sup>6</sup>.

Selective dorsal rhizotomy is a surgical procedure that allows partial sectioning of lumbar and sacral spinal roots, with consequent muscle relaxation by reducing the peripheral sensory stimulation conducted to the spinal cord by these dysfunctional roots. In the last decades, this surgery has been established as the best choice for the treatment of spasticity in walking children with GMFCS, levels II or III, when the objective is to improve the walking of these children<sup>7,8</sup>. In the surgical technique developed by Warwick Peacock (1986) the lumbar SDR is performed through laminotomy of the L2 to L5 vertebrae with laminectomy of the S1 and S2 vertebrae, and identification of the spinal roots through direct visualization in their respective spinal foramina<sup>9</sup>. Unfortunately, some studies have associated this SDR technique with an increased incidence of spinal deformities<sup>10-12</sup>. In 1991, Park and Jonhston reintroduced lumbar SDR with surgical access at the level of the medullary cone. In this technique, the authors performed laminectomy of the L1 or L2 vertebrae, to access the spinal roots of the cauda equina. Also, the intraoperative electromyographic study is used to identify the segmental level of each root, through differences existing in the neurophysiological parameters of these delicate roots<sup>13</sup>. The authors also improved the process of quantifying changes in the excitability of the dorsal spinal roots proposing a classification scale for the reflex motor response that facilitated the decision-making during surgery<sup>14</sup>. In this less invasive surgical approach, the incidence of spinal deformities

was comparable to the natural history of children with spastic CP who received only outpatient care<sup>15</sup>.

Recent publications support the choice of SDR in non-walking children with spastic CP. A study carried out in children with GMFCS, levels IV and V, reported a greater reduction in spasticity and greater functional gain with SDR when compared to the use of ITB<sup>16</sup>.

The purpose of this article is to investigate the neurological outcomes and perioperative complications after lumbar SDR in non-walking children with spastic CP.

## MATERIAL AND METHODS

The authors performed lumbar SDR with surgical access at the level of the medullary cone followed by laminoplasty at the operated vertebral level, and used the resources of intraoperative neurophysiological monitoring and intraoperative behavioral assessment to identify and classify the excitability of the dorsal spinal roots. The children are accompanied by a multidisciplinary team (Neurosurgery, Neurology, Orthopedics, Physiotherapy and Occupational Therapy) and undergo intensive physical rehabilitation after surgery.

Parental/guardian free and informed consents were signed, and the research protocol was approved by the local Research Ethics Committee.

### *Functional evaluation*

Pre- and postoperative functional evaluation were performed by a team of physical therapists and occupational therapists. They used a standardized technique for clinical quantification of spasticity using the modified Ashworth scale<sup>17</sup>, and for the classification and measurement of motor function using the GMFCS and GMFM – 88 scales<sup>1</sup>.

### *Surgical procedure*

All procedures were performed under total intravenous anesthesia. When necessary, succinylcholine was used as a short-acting neuromuscular blocker to avoid the use of long-acting muscle

relaxants that could interfere with the performance of the intraoperative neurophysiological monitoring.

The medullary cone was localized by preoperative study with magnetic resonance imaging of the lumbar spine. Laminotomy of the L1 or L2 vertebrae was performed, with access to the dura mater. After exposure of the cauda equina, each spinal root was submitted to electromyographic study, in order to provide fundamental information for surgical procedure, such as: the neurophysiological identification of dorsal and ventral spinal roots; and the quantification of changes in the excitability of the dorsal spinal roots. From L1 to S2, the dorsal roots were identified and then separated into 4 to 6 radicles, being sectioned in specific percentages according to the criteria established by the surgical team. The dura mater was sealed tightly. Anatomical restoration of the spine was performed through laminoplasty at the operated vertebral level.

### *Neurophysiological procedure*

In the anesthetized patient, a sterile technique was used to insert pairs of disposable needle electrodes into specific muscles innervated by spinal roots of surgical interest: iliac muscle, magnus adductor muscle, vast medial muscle, anterior tibial muscle, semitendinosus muscle, gastrocnemius muscle, adductor hallucis muscle and external anal sphincter muscle. The clinical presentation of the reflex motor response was observed and palpated by a physical therapist, with particular attention to contractions in muscle groups other than those monitored by the clinical neurophysiologist.

The intraoperative electromyographic protocol was based on the neurophysiological study originally described by Fasano et al.<sup>18</sup>, and modified by Philips and Park<sup>14,19</sup>, consisting of two stages: electromyography stimulated at 1 Hertz or threshold stimulation, and electromyography stimulated at 50 Hertz or tetanus stimulation. Electrical stimulation was performed with a 0.5 cm bipolar probe between the poles.

The classification of the intensity of the reflex motor response of the dorsal radicles followed the classification scale proposed by Philips and Park<sup>14</sup>: grade 0 (when there was no reflex motor response); grade 1 (when there was a reflex motor response only at the segmental level corresponding to stimulation); grade 2 (when there was a reflex motor response at the segmental level corresponding to stimulation, with the propagation of this

response to the adjacent segmental level); grade 3 (when there was a reflex motor response in the entire lower limb ipsilateral to stimuli); and grade 4 (when there was a reflex motor response in both lower limbs). The choice of dorsal radicles for surgical section was based on three principles: the electromyographic response; the behavioral response recorded by the physical therapist; and the clinical and functional objectives that were established for the patient in the preoperative multidisciplinary evaluation. In general, the dorsal roots that showed responses were not submitted to section: grade 0, grade 1 and grade 2. Dorsal roots with grade 3 and grade 4 responses were sectioned from 52.5% to 74.9% of their transverse area, varying according to the segmental level of each root.

### Postoperative physical rehabilitation

In a multidisciplinary environment, composed of physical therapists and occupational therapists, an intensive physical rehabilitation protocol is based on learning and motor recovery. Post-operative rehabilitation was performed during the first week, and training was given to relearn previously performed motor activities, such as rolling and dragging. From the second week onwards, treatment consisted of functional electrostimulation followed by assisted active exercise, pilates adapted to neurofunctionality, treadmill training with partial weight support, sensory integration, restriction and movement induction training, bimanual training, neuroevolutionary Bobath method, resources of the Therasuit method (except the suit in this initial phase of rehabilitation), quality training of movements, and guidance on the use of daily parapods at home<sup>20</sup>.

### Statistical analysis

Assessment using the distribution of normality was carried out with Kolmogorov-Smirnov test, and the Wilcoxon test compared the values obtained on the modified Ashworth and GMFM-88 scales in the pre- and post-lumbar SDR moments. It was adopted a p-value <0.05 as statistically significant.

## RESULTS

The sample consisted of 59 non-walking children with spastic CP, 43 boys and 16 girls, between 3 and 9 years of age. Other data is described in Table 1.

The GMFCS scale is an ordinal measure of five levels that establishes, in decreasing order, the level of independence and functionality of these children. In this study, 31 children were classified as GMFCS level IV (child with limited mobility and frequent use of a wheelchair) and 28 children were classified as level V (child without walking and unable to sit independently)<sup>1</sup>.

The functional results achieved with the surgery were quantified of the modified Ashworth and GMFM-88 scales, from pre-SDR and about 10 months post-SDR. It was observed a significant reduction in the values of the modified Asworth scale between the pre- and post-SDR moments (Wilcoxon test,  $p < 0.001$ ) in all joint movements studied. Reflecting a significant reduction in lower limb spasticity 10 months after surgery (Table 2).

The GMFM-88 (Gross Motor Function Measure) is an instrument developed specifically to measure and monitor gross motor function in children with CP over time. A total of 88 items is evaluated, equally weighted and grouped into 5 dimensions: A. lying and rolling; B. sitting; C. crawling and kneeling; D. standing; and E. walking, running and jumping. The scores for each

**Table 1.** Sample aspects of the patients who underwent lumbar SDR (n = 59).

|                                | n (%)     | Mean $\pm$ SD   |
|--------------------------------|-----------|-----------------|
| <b>Age (years)</b>             |           | 5.11 $\pm$ 1.7  |
| <b>Gender</b>                  |           |                 |
| Male                           | 43 (72.9) |                 |
| Female                         | 16 (27.1) |                 |
| <b>GMFCS scale</b>             |           |                 |
| level IV                       | 31 (52.5) |                 |
| level V                        | 28 (47.5) |                 |
| <b>Dorsal root section (%)</b> |           |                 |
| L1                             |           | 73.9 $\pm$ 5.1  |
| L2                             |           | 74.1 $\pm$ 5.6  |
| L3                             |           | 73.4 $\pm$ 5.4  |
| L4                             |           | 70.2 $\pm$ 5.6  |
| L5                             |           | 73.8 $\pm$ 5.1  |
| S1                             |           | 74.9 $\pm$ 5.7  |
| S2                             |           | 52.5 $\pm$ 24.8 |
| <b>Follow-up (months)</b>      |           | 10.3 $\pm$ 1.15 |

SD = standard deviation.

**Table 2.** Comparison of data from modified Ashworth scale to assess the spasticity of patients undergoing lumbar SDR (n = 59).

| Movement        | pre SDR   | post SDR  | <i>p</i> -Value <sup>1</sup> |
|-----------------|-----------|-----------|------------------------------|
|                 | Mean±SD   | Mean±SD   |                              |
| Hip flexion     | 1.37±0.69 | 0±0       | <0.001                       |
| Hip adduction   | 2.29±0.63 | 0.35±1.12 | <0.001                       |
| Knee extension  | 1.21±0.79 | 0.21±0.72 | <0.001                       |
| Knee flexion    | 1.71±0.72 | 0±0       | <0.001                       |
| Plantar flexion | 2.83±0.39 | 0±0       | <0.001                       |
| Halux flexion   | 1.11±0.87 | 0±0       | <0.001                       |

<sup>1</sup>Wilcoxon test. SD = standard deviation.

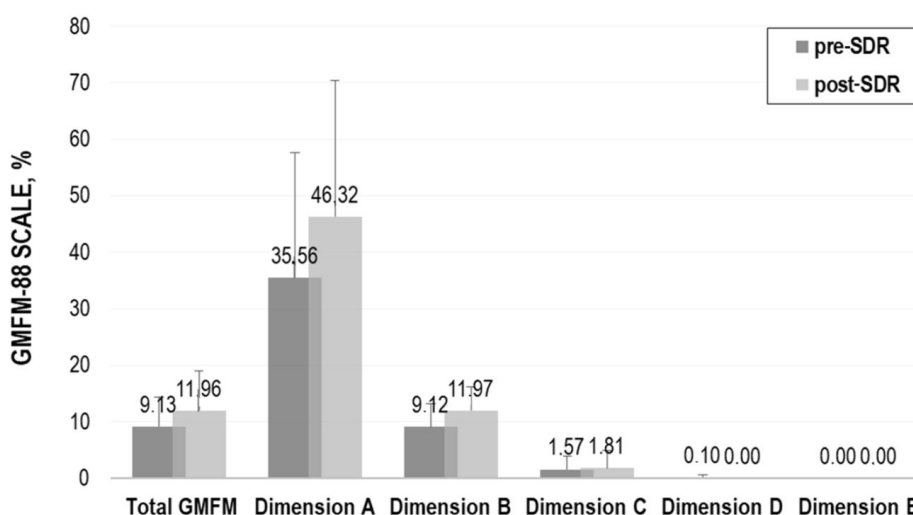


**Figure 1.** Comparison of data from GMFM-88 scale to assess gross motor function of patients who underwent lumbar SDR (n = 59).

dimension are expressed as a percentage of the maximum score to be achieved for that dimension. The total score of the scale is obtained by averaging the percentage scores of the 5 dimensions. In the pre-SDR assessment, the best scores on this scale were obtained in dimensions A and B, due to the marked motor impairment of these children<sup>1</sup>. After SDR, an important improvement in the gross motor function was observed, demonstrated by an increase of 2.83% in the values of the total GMFM-88 between the pre- and post-SDR moments (Wilcoxon test,  $p < 0.05$ ). As expected, this improvement occurred in dimension A (10.76% increase; Wilcoxon test,  $p < 0.05$ ) and in dimension B (2.85% increase; Wilcoxon test,  $p < 0.05$ ). These dimensions represent the gross motor functions related to the trunk, being partially impaired in tetraparetic children with spastic CP (Figure 1).

Peri-operative complications were present in 19 patients (32.2%), and consisted of a urinary retention, requiring intermittent catheterization ( $n = 5$ ), delayed wound healing ( $n = 2$ ), fever of unknown cause ( $n = 1$ ), severe pain ( $n = 11$ ), and dysesthesias and spasms ( $n = 9$ ). All complications were mild and temporary (Table 3).

## DISCUSSION

A large number of scientific publications advocate the use of lumbar SDR to treat spasticity in walking children with mild to moderate CP. A multicenter study conducted in England by Summers et al.<sup>8</sup>, confirmed the results of previous studies on the

**Table 3.** Incidence of complications of patients who underwent lumbar SDR (n = 59).

|                                    | Number    | %           |
|------------------------------------|-----------|-------------|
| <b>Perioperative complications</b> | <b>19</b> | <b>32.2</b> |
| Pain, severe                       | 11        | 18.6        |
| Delayed wound healing              | 2         | 3.4         |
| Fever of unknown cause             | 1         | 1.7         |
| Urinary retention                  | 5         | 8.5         |
| Dysesthesias and spasms            | 9         | 15.3        |
| Sensory alteration                 | 4         | 6.8         |

benefit of lumbar SDR for the walking of children with spastic CP. The results served as a basis for the implementation of a public policy to offer this surgery by the National Health Service of England for walking children with spastic CP, GMFCS levels II and III, and aged from 3 to 9 years<sup>21</sup>. On the other hand, ITB is often the conventional treatment for non-walking children with spastic CP<sup>4</sup>. However, some studies have shown benefits with the use of lumbar SDR in them. Kan et al.<sup>22</sup>, showed in a group of 71 children classified as GMFCS levels III, IV and V that lumbar SDR was more effective in reducing the degree of spasticity and improving gross motor function than ITB. Ingale et al.<sup>23</sup>, evaluated spasticity and functional outcome in 10 children with CP, GMFCS levels IV and V one year after lumbar SDR. These children were previously treated with ITB. Spasticity was reduced after SDR, and 90% of the parents felt that functional outcome was improved compared to ITB.

In the present study, the authors observed a significant improvement in the two functional assessment scales: modified Ashworth and GMFM-88 (Table 2 and Figure 1). The muscular relaxation documented in the lower limbs by the modified Ashworth scale resulted in significant functional improvement in dimensions A and B of the GMFM-88 scale, which are associated with the thick motor functions of the trunk, such as: lying down, rolling and sitting. This beneficial functional effect of lumbar SDR on thoracic, and even cervical, spinal segments has also been documented by other authors<sup>24,25</sup>. In one of these studies, lumbar SDR showed a greater reduction in spasticity of the upper and lower limbs when compared to the use of intrathecal baclofen<sup>9</sup>. The mechanisms by which this upper limb relaxation occurs are still not clear, but it is believed to be related to the action of interneurons on ascending spinal cord pathways<sup>26</sup>.

Thus, with the control of spasticity of the lower limbs, trunk and even the upper limbs, a reduction in the incidence of orthopedic

deformities can be observed. Children with spastic CP have a higher incidence of scoliosis. A cohort study, that followed 962 children with CP for 20 years, found a 75% incidence of scoliosis in patients with GMFCS level V<sup>4</sup>. Muquit et al.<sup>27</sup>, demonstrated that intrathecal baclofen, at an ideal dose, was unable to reduce spasticity and prevent the progressive worsening of thoraco-lumbar scoliosis in a patient with spastic CP. This patient underwent lumbar SDR and scoliosis correction at the same surgical time, with a significant improvement in spasticity after lumbar SDR. Besides that, some studies have shown that lumbar SDR is also associated with a reduction in the number of orthopedic surgeries to correct musculoskeletal disorders in children with spastic CP, such as: muscle-ligament shortening and hip dislocation<sup>28,29</sup>. Another study evaluating the rate of orthopedic surgery after SDR showed that in all age groups 25% of independent walkers and 44% of assisted walkers required orthopedic surgery over a 9-year follow-up<sup>30</sup>. Those undergoing SDR at a young age demonstrated the lowest requirement for orthopedic surgery after SDR<sup>31</sup>. In addition, orthopedic deformities in non-walking children with spastic CP cause pain, make positioning and daily care difficult. It was observed that lumbar SDR promotes pain reduction, facilitates daily care and improves the quality of life of these children<sup>32</sup>.

All patients in this study underwent postoperative physical rehabilitation, because several studies have shown that the lumbar SDR combined with physical rehabilitation has better results than physical rehabilitation alone. In the Toronto study, evaluation at 12 months showed significant improvements in GMFM scores, knee and ankle tone, passive ankle range of motion, soleus EMG reflex activity on forced dorsiflexion and foot-floor contact pattern<sup>33</sup>. In the Vancouver trial, significant improvements were observed at 1 year in GMFM, spasticity and range of movement in the group undergoing SDR combined with physical therapy<sup>34</sup>.

The risk of structural instability of the spine is a frequent concern when performing this surgery. It has been suggested that SDR may increase the chance of developing progressive spinal deformity requiring surgical fixation, and that this risk is higher in non-walking children<sup>35</sup>. Therefore, the authors performed lumbar SDR with surgical access at the level of the medullary cone followed by laminoplasty at the operated vertebral level. A large patient series has shown that limited laminectomies at the level of the conus are not associated with long-term spinal deformity<sup>15</sup>. The authors also sectioned between 50% and 70% of the dorsal

roots, as it is shown that lower section values did not result in functional improvement<sup>36</sup>.

As in previous studies, this study presented a low rate of serious adverse events attributable to the intervention<sup>32</sup>. Transient urinary retention occurred in a few patients, as a consequence of transient bladder hypotonia after the immediate reduction in spasticity, or due to a possible contusion of the spinal roots in the cauda equina during surgical manipulation. To reduce the risk of incontinence a intraoperative electromyographic study used the differences in neurophysiological parameters between the adductor hallucis and the external anal sphincter muscles to delimit the dorsal root section S2.

Pain is a common feature of the immediate postoperative recovery period. A significant number of patients who underwent SDR experienced moderate to severe pain in the first 48-72 hours after surgery. Concomitantly, almost half of these patients also had transient dysesthesia in the lower limbs, often accompanied by muscle spasms associated with involuntary flexion of the hip and lower limbs. However, permanent changes in sensitivity, such as hypoesthesia, were uncommon and clinically discrete.

The results of this study suggest that lumbar SDR with surgical access at the level of the medullary cone followed by laminoplasty is a valid therapeutic option for non-walking children with spastic CP, GMFCS levels IV and V, whose therapeutic objective is not associated with improved gait, as in children with GMFCS levels II and III. It aims to reduce diffuse spasticity, decrease pain and prevent spinal deformities, among other musculo skeletal disorders, resulting in greater comfort in carrying out daily care and improving the quality of life of these children.

## CONCLUSIONS

Lumbar SDR with surgical access at the level of the medullary cone in non-walking children with spastic CP promoted muscle relaxation in the lower limbs with significant improvement in gross motor functions, and the perioperative complications were mild and temporary.

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