SPINAL DEFORMITIES AFTER SELECTIVE DORSAL RHIZOTOMY

DEFORMIDADES DA COLUNA VERTEBRAL APÓS RIZOTOMIA DORSAL SELETIVA

DEFORMIDADES ESPINALES LUEGO DE RIZOTOMÍA DORSAL SELECTIVA

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ABSTRACT

Objective: Selective dorsal rhizotomy (SDR) used for spasticity treatment could worsen or develop spinal deformities. Our goal is to describe spinal deformities seen in patients with cerebral palsy (CP) after being treated by SDR. Methods: Retrospective study of patients operated on (SDR) between January/1999 and June/2012. Inclusion criteria: spinal Rx before SDR surgery, spinography, and assessment at follow-up. We evaluated several factors emphasizing level and type of SDR approach, spinal deformity and its treatment, final Risser, and follow-up duration. Results: We found 7 patients (6 males): mean age at SDR 7.56 years (4.08-11.16). Mean follow-up: 6.64 years (2.16-13), final age: 14.32 years (7.5-19). No patient had previous deformity. GMFCS: 2 patients level IV, 2 level III, 3 level II. Initial walking status: 2 community walkers, 2 household walkers, 2 functional walkers, 1 not ambulant, at the follow-up, 3 patients improved, and 4 kept their status. We found 4 TL/L laminotomies, 2 L/LS laminectomies, and 1 thoracic laminectomy. Six spinal deformities were observed: 2 sagittal, 3 mixed, and 1 scoliosis. There was no association among the type of deformity, final gait status, topographic type, GMFCS, age, or SDR approach. Three patients had surgery indication for spinal deformity at skeletal maturity, while those patients with smaller deformities were still immature (Risser 0 to 2/3) although with progressive curves. Conclusions: After SDR, patients should be periodically evaluated until they reach Risser 5. The development of a deformity does not compromise functional results but adds morbidity because it may require surgical treatment.

Keywords: Cerebral palsy; Muscle spasticity; Rhizotomy; Scoliosis.

RESUMO

Objetivo: A rizotomia dorsal seletiva (SDR) para tratamento da espasticidade pode piorar ou desenvolver deformidades da coluna vertebral. Nosso objetivo é descrever as deformidades da coluna observadas em pacientes com paralisia cerebral (PC) depois de serem submetidos à SDR. Métodos: Avaliação retrospectiva de pacientes operados (SDR), entre janeiro/1999 e junho/2012. Critérios de inclusão: Radiografias vertebrais prévias, espinografia e avaliação no acompanhamento. Foram avaliados vários fatores com ênfase no nível e tipo de abordagem da SDR, deformidade da coluna vertebral e seu tratamento, Risser final e tempo de acompanhamento. Resultados: Encontramos 7 pacientes (6 do sexo masculino): média de idade à SDR 7,56 anos (4,08-11,16). Acompanhamento médio: 6,64 anos (2,16-13); idade final: 14,32 anos (7,5-19). Nenhum paciente tinha deformidade anterior. GMFCS: 2 pacientes com nível IV, 2 com nível III, 3 com nível II. Estado deambulatório inicial: 2 deambuladores comunitários, 2 deambuladores domiciliares, 2 deambuladores funcionais, 1 Não deambulador; no acompanhamento, 3 melhoraram e 4 mantiveram seu estado. Foram encontradas 4 laminotomias TL/L, 2 laminectomias L/LS, 1 torácica. Foram detectadas 6 deformidades da coluna: 2 sagitais, 3 mistas e 1 escoliose. Não houve nenhuma associação entre o tipo de deformidade e o estado de deambulação final, tipo topográfico, GMFCS, idade nem abordagem da SDR. Três pacientes tinham indicação de cirurgia à maturidade esquelética, mas as deformidades menores eram ainda imaturas (Risser 0 a 2/3), apesar das curvas evolutivas. Conclusões: A coluna deve-se ser avaliada periodicamente depois da SDR até Risser 5. O desenvolvimento de uma deformidade não afeta o resultado funcional, mas sim acrescenta morbidade, pois pode exigir tratamento cirúrgico.

Descritores: Paralisia cerebral; Espasticidade muscular; Rizotomia; Escoliose.

RESUMEN

Objetivo: La rizotomía dorsal selectiva (SDR) para el tratamiento de la espasticidad podría empeorar o desarrollar deformidades espinales. Nuestro objetivo es describir deformidades espinales observadas en pacientes con parálisis cerebral (PC) luego de someterse a SDR. Métodos: Evaluación retrospectiva de pacientes operados (SDR) entre enero/1999 y junio/2012. Criterios de inclusión: Rx raquídeas previas, espinograma y evaluación al seguimiento. Se evaluaron diversos factores con énfasis en nivel y tipo de abordaje de SDR, deformidad espinal y su tratamiento, Risser final y tiempo de seguimiento. Resultados: Encontramos 7 pacientes (6 varones): edad promedio a la SDR 7,56 años (4,08 – 11,16). Seguimiento promedio: 6,64 años (2,16 - 13); edad final: 14,32 años (7,5 – 19). Ninguno tenía deformidad previa. GMFCS: 2 pacientes nivel IV, 2 nivel III, 3 nivel II. Estado ambulatorio inicial: 2 Deambuladores Comunitarios, 2 Domiciliarios, 2 Funcionales, 1 No Deambulador; al seguimiento 3 mejoraron y 4 lo mantuvieron. Hubo 4 laminotomías TL/L, 2 laminectomías L/LS y 1 torácica. Se detectaron 6 deformidades espinales: 2 sagitales, 3 mixtas y 1 escoliosis. No hubo asociación entre tipo de deformidad y estado ambulatorio final, tipo topográfico, GMFCS, edad ni abordaje para la SDR. Tres pacientes tuvieron indicación de cirugía; pero estos llegaron a la madurez esquelé-

Study conducted at the Centro "Nicolás Andry", Monteagudo 201, Resistencia, Argentina. Correspondence: Centro "Nicolás Andry", Monteagudo 201. (3500) Resistencia, Chaco, Argentina. manzonepatricio@hotmail.com tica, mientras que los de deformidades menores aún son inmaduros (Risser 0 a 2/3) aunque con curvas evolutivas. Conclusiones: Se debe evaluar periódicamente la columna después de SDR hasta Risser 5. La aparición de una deformidad no afecta el resultado funcional pero sí agrega morbilidad pues puede requerir tratamiento quirúrgico.

Descriptores: Parálisis cerebral; Espasticidad muscular; Rizotomía; Escoliosis.

INTRODUCTION

Rhizotomy of the dorsal or posterior root to treat spasticity was performed for the first time at the beginning of the 20th century, based on the hypothesis that the rigidity would disappear after section of the dorsal roots.

The surgical procedure can be accomplished using the techniques popularized by Peacock, in which laminectomies of L1 to L5 are performed, and by Fazano, which consists of a single laminectomy of T12 and L1.² A third variant is that of Park^{3,4} involving less removal of bone.

Although the procedure is recognized as useful and approved for use, ⁵⁻⁷ there is not yet a universal consensus around patient selection criteria. ^{8,9} Furthermore, little is known about the effect of rhizotomy on the development or the progression of spinal deformity. The treatment of spasticity by selective dorsal rhizotomy (SDR) could worsen or develop spinal deformities.

Our objective in this study was to describe the spinal deformities observed in a group of patients diagnosed with cerebral palsy, after having undergone selective dorsal rhizotomy.

METHODS

We conducted a retrospective evaluation of patients diagnosed with cerebral palsy, who had undergone laminectomies or laminotomies for selective dorsal rhizotomy between January 1999 and June 2012 (13 and a half years). The critical selection criteria were the existence of pre-rhizotomy spinal radiographs and spinography and a complete final follow-up assessment. (Figure 1)

The following parameters were evaluated for each case: etiology, sex, topographical and physiopathological type of cerebral palsy, GMFCS classification, presence of pre-rhizotomy extraspinal orthopedic deformities, ambulatory state pre-rhizotomy and at the last control visit, age at the completion of surgery, pre- and post-rhizotomy spinal deformity, level of the laminectomy, post-rhizotomy spinal deformity treatment, appearance of orthopedic deformities following the rhizotomy, degree of bone maturation (according to the Risser sign) at the last control visit, and post-rhizotomy follow-up time. The Phelps, ¹⁰ Hoffer et al., ¹¹ and Palisano et al. ¹² classification systems were used.

Because this was an observational investigation with no linkable data and no patient risk, since only clinical records and complementary studies were used, and because the absolute protection of the privacy and total confidentiality of the subjects and their data were guaranteed, neither an approval by the Institutional Review Board nor a signed informed consent form were required by the institution where the study was conducted.

RESULTS

We identified 8 patients, of whom 7 could be completely evaluated: 1 female, 6 males, with an average age of 7 years 7 months at the time of surgery (ranging from 4 years 1 month to 11 years 2 months).

Given the small sample, it was impossible to draw statistically valid conclusions from the analyses. We were only able to give our impressions and identify certain trends.

Four patients had spastic diplegia, one had spastic triplegia, one had spastic quadriplegia, and one had mixed diplegia. None of the patients presented any spinal deformity prior to the rhizotomy.

Six of the 7 patients had suffered perinatal anoxia-hypoxia. According to the GMFCS, 2 patients were level IV, 2 were level III, and 3 were level II. The pre-rhizotomy ambulatory states of the patients were 2 community ambulatory (CA), 2 household ambulatory (HA), 2 functional ambulatory (FA), and 1 non-ambulatory (NA). At the end of follow-up, 3 patients had improved their ambulatory status and 4

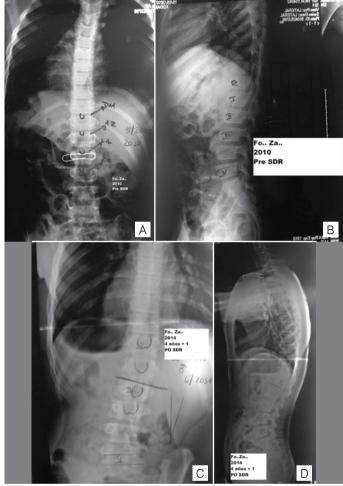


Figure 1. Case 7 (see Table 1). (A): Pre-rhizotomy frontal spine x-ray; note the marks of the planned laminotomy sites and the absence of prior deformity. (B): Pre-rhizotomy profile spine x-ray. (C): Frontal x-ray more than 4 years after rhizotomy surgery; right lumbar curve of 15°. Note the marks of the laminotomy sites. (D): Lateral spinography more than 4 years after rhizotomy surgery; thoracolumbar kyphosis (T12-L1) of 25°.

had maintained theirs: 4 patients as CA, 1 as HA, and 2 as FA (1 HA progressed to CA, one FA progressed to CA, and 1 NA progressed to FA). (Tables 1 and 2) All had associated pre-rhizotomy extraspinal orthopedic deformities, but 5 also had deformities that appeared later.

Four patients underwent rhizotomies via thoracolumbar and upper lumbar laminectomies (T11, T12, L1, and L2), 2 patients via lumbar/lumbosacral laminectomies, and 1 via a mid-thoracic laminectomy.

The average post-rhizotomy follow-up was 6 years and 8 months (ranging from 2 years 2 months to 13 years) and the average age at end of follow-up was 14 years and 4 months (ranging from 7 years 6 months to 19 years).

At the end of follow-up, we detected significant spinal deformities in 6 of the 7 patients and an insignificant frontal curve (9°) in 1 of the 7 patients. Two patients had strictly sagittal deformities (one lumbar hyperlordosis of 90° and one thoracolumbar hyperkyphosis of 75°), 3 patients had mixed deformities (3 thoracolumbar kyphoscolioses [TL kyphosis of 80° + right lumbar scoliosis of 69°] [TL kyphosis of 25° + atypical right lumbar scoliosis of 15°] [TL kyphosis of 35° + right

Table1. Case series.

Case number	Age at end of follow-up (years + months)	Sex	Type of impairment	Type of CP	GMFCS	Pre-rhizotomy ambulatory state	Age at rhizotomy (years + months)	Spinal deformity prior to rhizotomy	Level of surgical approach	Post-rhizotomy deformity	Treatment of deformity	Current ambulatory state	Post-rhizotomy follow-up (years + months)	Risser at last control
1	17	М	Quadri	S	IV	NA	4 + 1	NO	L5-S1	HL	Surgery	FA	13	4-5
2	17 + 4	F	Spa Dip	S	III	НА	11	NO	T6-T7-T8	KS	Surgery	CA	6 + 4	5
3	19	М	Spa Dip	S	Ш	НА	11 + 2	NO	T12-L1-L2	НК	TLSO (*)	НА	7 + 10	5
4	12	М	Spa Trip	S	Ш	CA	7	NO	T12-L1	S	TLSO	CA	5	1
5	7 + 6	М	Spa Dip	S	IV	FA	5 +4	NO	L1-L2	KS	N/T	FA	2 + 2	0
6	14	М	Spa Dip	S	Ш	FA	6 + 2	NO	L4-L5-S1	Smaller curve	N/T	CA	8	2-3
7	14 + 3	М	Diplegia	М	III	CA	10 + 2	NO	T11-T12-L1-L2	KS	N/T	CA	4 + 1	0-1

Quadri: quadriplegia. Spa Dip: Spastic Diplegia. Spa Trip: Spastic Triplegia. S: spastic. M: mixed. NA: Non-ambulatory. HA: household ambulatory. CA: community ambulatory. FA: functional ambulatory . HL: Hyperlordosis. HK: hyperkyphosis. S: scoliosis KS: kyphoscoliosis. (*): in planning for surgical treatment. N/T: no treatment.

Table 2. Ambulatory state pre-rhizotomy and at end of follow-up.

Preoperative	Ambulatory Status at End of Follow-up									
Ambulatory Status	Non- Ambulatory	Functional Ambulatory	Household Ambulatory	Community Ambulatory						
Non- Ambulatory		1								
Functional Ambulatory		1		1						
Household Ambulatory			1	1						
Community Ambulatory				2						

lumbar scoliosis of 17°]), one patient had a left lumbar scoliosis of 31° , and the last patient had a right TL curve of 9° . We found no obvious association between the type of deformity and the ambulatory state at the end of follow-up, or the topographical type of CP, or the GMFCS level, or the age at the time of the SDR, or the technique of approach for the SDR, or the level of the SDR approach.

Two patients had to be treated surgically, 2 were treated with TLSO (one failed and was indicated for surgery and the other is still at Risser 1), and 3 received no treatment other than rehabilitation (patients with Risser 0 to 2/3). We found no association between age at the time of the rhizotomy and the type of curve, but the degree of bone maturation seemed to influence the degree of curve severity. Thus, if we compare the severity of the deformities (using the need for surgery as the criterion) against the degree of bone maturation (according to the Risser sign), we see that the 3 patients with severe deformities were rated Risser 4 or 5, while the remaining patients with less serious deformities were still skeletally immature (Risser 0 to 2/3), though 2 of these 3 had curves in a state of evolution. (Table 3)

DISCUSSION

Although selective dorsal rhizotomy (SDR) has been effective in reducing spasticity in children with spastic cerebral palsy,^{5,13-17} its long-term effects on the musculoskeletal are still unknown,¹⁸ a fact

Table 3. Relationship between age at performance of rhizotomy, severity of the deformity, and Risser at the end of follow-up.

	Severity of the Deformity											
Age at time of SDR	Sagittal or mixed deformity with surgical indication			Sagittal or mixed deformity without surgical indication			Scoliosis with surgical indication			Scoliosis and curves without surgical indication		
4 to 6 years			1	1*								
6 to 8 years										1		
8 to 10 years										1*		
10 to 12 years		1	1	1*								
	< 0 =3	4	5	< 0 =3	4	5	< 0 =3	4	5	< 0 =3	4	5
	Risser at the end of Follow-up											

*Curves still evolving.

that gains importance due to the permanence of the neurological change produced by the rhizotomy. As regards to the spine, hyperlordosis, scoliosis, spondylolysis, and spondylolisthesis have been documented following a rhizotomy. Hyperlordosis, present in one of our patients, is referenced as the most common and most difficult to manage secondary deformity. Figure 2)

It is not clear whether the prevalence of these conditions is greater than in children with spastic cerebral palsy who have not undergone rhizotomy, 22 but several retrospective 19,23 and prospective 21 studies appear to suggest it.

Several studies have reported the appearance of scoliosis following rhizotomy, and they have stated that accelerated progression of already existing deformities might also be a reason for concern. ^{19,23-25} Turi and Kalen²³ reported scoliosis in 12 out of 42 patients, most of whom were non-ambulatory, and none of whom had a preexisting deformity. In our study, we observed spinal deformity in 6 out of 7 patients, all ambulatory, strikingly different from other studies. ²² In our cases, sagittal deformities were the most common, whether standalone or associated with others. (Figure 3) Only one case presented a scoliotic curve without a more serious sagittal deformity. Like the Johnson et al. series, ²¹ there were no differences between the patients who underwent more or less conservative procedures (laminotomies versus laminectomies).

Scoliosis seems to develop in 16%-17% of cases, 22,25 being appar-

ently more common in quadriplegics.^{24,25} Similarly to the Spiegel series,²² none of our 7 patients had a deformity prior to the rhizotomy. (Figure 1)

Steinbok et al. ¹⁹ reports a greater than 38% incidence of kyphotic deformity following SDR. Other authors confirm this. ²³ Apparently, this deformity is more likely to develop when a Fazano-type thoracolumbar laminectomy is used. ² We identified thoracolumbar kyphosis in 4 (57%) of the 7 patients evaluated, either isolated (1 case) or associated with coronal deformity (3 cases). (Figure 3) Curiously, the sagittal deformity was always the most significant, even though 3 patients underwent a thoracolumbar/high lumbar approach and one a mid-thoracic approach. (Table 1)

On the contrary, lumbar sagittal deformities, including hyperlordosis and spondylolisthesis, were reported following rhizotomy with laminectomy. ^{25,26} Although Spiegel et al. ²² found no significant difference between postoperative lordosis and lordosis at the end of follow-up regardless of the technique used, lordosis greater than 50° following SDR was found in from 7% to 17% of patients according to these series. ^{23,25} Lumbar hyperlordosis is usually more frequent in non-ambulatory and/or quadriplegic patients. ^{20,24} Crawford et al. ²⁰ described two quadriplegic patients who developed progressive, rigid hyperlordotic deformities. One of our patients corresponded exactly to that pattern, developing hyperlordosis of 120°. (Figure 2)

Incidences of from 6% to 24% spondylolisthesis and of 14% isolated spondylolysis have been reported, ^{21,22,25} although the highest rate occurs in spastic diplegics. ²¹ However, we did not encounter this condition in our group.

As already mentioned, we found no closer association of the presence of post-rhizotomy deformities with any particular technique of approach for the rhizotomy. However, nor did we find any association between the degree of severity of the CP and any ambulatory state having a higher incidence of these deformities, unlike the other authors.²⁷ Age had already been described as an important trigger

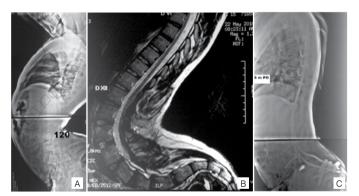


Figure 2. Case 1 (see Table 1). (a): Lateral spinography at more than 11 years following selective rhizotomy surgery. Gross sequelary lumbar hyperlordosis of 120°. (b): MRI of lumbosacral spine corresponding to spinography. (c): Lateral spinography 6 months following the surgery for correction of the hyperlordosis.

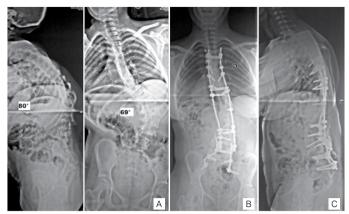


Figure 3. Case 2 (see Table 1). (a): Frontal and lateral spinographies 3 years and 9 months following selective rhizotomy, showing a mixed deformity (kyphoscoliosis) with right frontal lumbar curve of 69° and thoracolumbar kyphosis of 80°. Note the mark of the mid-thoracic laminectomy (T6-T7-T8) in front (b): Late postoperative frontal spinography of the surgical correction of the deformity. (c): Late postoperative lateral spinography of the surgical correction of the deformity.

factor in the development of deformities following multi-level laminectomies, regardless of the underlying pathology.²⁸ In this study and as also described by others,² age and the time remaining to skeletal maturity in children with CP who underwent rhizotomy were important in the development of spinal deformities. (Table 3)

While our post-rhizotomy follow-up is similar to that of other series, ^{19,21,22,25-27} we agree with Johnson that the frequency of spinal deformities in patients who undergo SDR is higher than expected in this group of patients, ²¹ and that the controls should be prolonged until well into skeletal maturity. ²⁴ (Table 3)

CONCLUSIONS

Periodic evaluation of the alignment of the spine following a rhizotomy is recommended. Follow-up of these patients should be conducted until these reach skeletal maturity. The cause of this incidence of deformity is not clear and more studies are warranted. The appearance of deformity does not seem to affect the functional ambulatory outcome of the SDR, but it does increase morbidity since it may require surgical treatment.

Faced with the spinal deformities associated with it, dorsal rhizotomy should be evaluated carefully and with the expectation of the possible need for future spinal stabilization. Considering stabilization at the time of the rhizotomy could be justified in selected patients.

All the authors declare that there are no conflicts of interest regarding this article.

CONTRIBUTIONS OF THE AUTHORS: Each author made significant individual contributions to the development of the manuscript. PPM, MSVA, EO, MD, and EMA participated in the treatment and clinical follow-up of the patients and collaborated in data collection for the study. PPM and MSVA participated in the statistical analysis. PPM, MSVA, EO, MD, EMA, M LCI, and JG worked on the bibliographical research. All the authors contributed to the intellectual concept of the study.

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