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Short- and long-term effects of selective dorsal rhizotomy on gross motor function in ambulatory children with spastic diplegia

Clinical article

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Object. The primary aim of this prospective cohort study was to evaluate the short-term (1 year) and long-term (mean 6 years) effects of selective dorsal rhizotomy (SDR) on gross motor function and spasticity in ambulatory children with spastic diplegia. Secondary aims were to investigate side effects, additional treatment during follow-up (botulinum toxin type A injections or orthopedic surgery), and parental satisfaction.

Methods. Thirty-three children who had undergone SDR at a mean age of 6 years and 7 months (± 2 years) were included. There were 7 children at Gross Motor Function Classification System (GMFCS) Level I, 7 at Level II, and 19 at Level III. Gross motor function was assessed with the Gross Motor Function Measure–66 (GMFM-66). Spasticity was measured according to a modified Tardieu scale. Side effects, additional treatment, and parental satisfaction were recorded using a parental questionnaire and medical records.

Results. At 1-year follow-up, mean GMFM-66 scores improved significantly by 4.3 ± 4.1 points. Children at GMFCS Levels I and II showed significantly more improvement (7.2 points) on the GMFM-66 compared with children at GMFCS Level III (2.9 points). On long-term follow-up (mean 6 years ± 22 months), mean GMFM-66 scores improved significantly by 6.5 ± 5.9 points, without a difference between children at GMFCS Levels I and II and Level III. No relapse of spasticity was noted. Ten children (30%) needed orthopedic surgery and 13 children (39%) received botulinum toxin type A treatment after SDR. Twenty (91%) of the 22 parents who answered the questionnaire at long-term follow-up believed that their child's functioning had improved after SDR.

Conclusions. Selective dorsal rhizotomy resulted in short- and long-term improvements in gross motor function, without relapse of spasticity. However, the majority of the children still needed additional surgery or botulinum toxin A treatment. *(DOI: 10.3171/2011.2.PEDS10452)*

Key Words • **selective dorsal rhizotomy** • **gross motor function** • **spastic diplegia** • **botulinum toxin type A**

Cerebral palsy is the most common disorder causing physical disability in childhood. Cerebral palsy includes a group of disorders that affect the development of movement and posture, causing activity limitations that are attributed to nonprogressive disturbances that occurred in the fetal or infant brain.[15] In children with CP, spastic CP is the most common form. Spasticity is defined as hypertonia in which one or both of the following signs are present: 1) resistance to externally imposed movement increases with increasing speed of stretch, which varies with the direction of joint movement; and/or 2) resistance to externally imposed movement rises rapidly above a threshold speed or joint angle.[17,18] Spastic CP is often associated with discomfort, gait abnormalities, and functional limitations.[9] Treatment options for children with spastic CP consist of oral medication, intrathecal baclofen therapy, intramuscular botulinum toxin type A injections, or SDR.[9]

Selective dorsal rhizotomy is a neurosurgical procedure in which selected sensory nerves in the lumbar spine are transected to reduce the excitatory input entering the spinal cord via the lumbosacral posterior nerve roots.[21] The aim of SDR is to eliminate spasticity in the

Abbreviations used in this paper: CP = cerebral palsy; GMFCS = Gross Motor Function Classification System; GMFM-66 = Gross Motor Function Measure–66; ROM = range of motion; SDR = selective dorsal rhizotomy.

lower limbs, and thereby improve the walking ability of children with spastic diplegia at GMFCS[13] Levels I–III. A meta-analysis of 3 randomized controlled trials (90 total patients) demonstrated reduction of spasticity after SDR combined with physical therapy as compared with physical therapy only. In addition, gross motor function (as measured with the GMFM-66) improved 9–12 months after SDR.[9] Several studies reported long-term reduction in spasticity after SDR,[3,10] improved gait parameters,[3,22] improvements in functional movements,[10] mobility,[3] and self-care.[3,11] Spondylolisthesis[7] and scoliosis[6] are most frequently reported as adverse effects of SDR. In contrast, the decrease in spasticity after SDR could be associated with a reduction in the need for orthopedic procedures,[19] although reports on the frequency of additional treatments (such as botulinum toxin type A injections and orthopedic surgery) are unequivocal.[10,11]

Since 1998, 33 ambulatory children with spastic diplegia have been treated using SDR in the Netherlands. We reported short-term effects 1 year after SDR in the first 9 children,[23] showing reduced spasticity in the lower limbs and an improvement in gross motor function, self-care, and gait pattern. We recently showed the predictive value of brain MR imaging in a subset of patients of our cohort.[5]

The primary aim of the present study was to evaluate the short-term (1 year) and long-term (mean 6 years, range 3–8 years) effects of SDR on gross motor function and spasticity in ambulatory children with spastic diplegia, and investigate differences between children walking without (GMFCS Level I and II) or with ambulatory aids (GMFCS Level III). Secondary aims were to investigate the occurrence of side effects, necessity of additional treatment (botulinum toxin type A injections or orthopedic surgery), and parental satisfaction.

Methods

Study Participants

Between August 1998 and December 2005, 33 children underwent SDR treatment at the VU University Medical Center in the Netherlands. Two of these children had spastic familial paraparesis and 31 had CP. Inclusion criteria for SDR were as follows: spastic diplegia; 2.5 years of age or older; spasticity (defined as velocity-dependent resistance to passive stretch) in at least 6 groups of muscles in both legs; no contractures limiting function at the hip, knee, or ankle (< 20° flexion contracture in knee, popliteal angle < 80°, at least 0° dorsal flexion in the ankle possible); no structural bone deformities; at least able to crawl, sit independently > 10 seconds, maintain tall knee position (support for balance allowed), and squat 7 times; GMFCS Level I, II, or III; and good motivation and support after surgery from parents and the rehabilitation setting.

One child could not be contacted during follow-up due to an unknown change of address, therefore we used the last available data from a regular follow-up visit 3 years after SDR in our analyses. Another child did not show up for several appointments due to instable social circumstances, but regular follow-up data were available 1 year after SDR. At the long-term follow-up, only GMFCS data for this child were available from medical records.

In total, 6 children had only short-term follow-up data, and 27 children had undergone SDR more than 3 years previously (mean 6.0 years ± 22 months) at the time of their visit. The patient characteristics at baseline are presented in Table 1.

Surgical Technique

The operation was performed more or less according to the method described by Steinbok et al.[21] by the same neurosurgeon for all patients (W.J.R.O.). A laminotomy of L2–5 was performed and the dura was opened to expose the roots from L-2 to S-2. After separating the ventral from the dorsal roots at the foramina, each dorsal root was separated into 3–4 rootlets. Subsequently, each rootlet was stimulated separately to reveal abnormal, exaggerated responses of the muscles. The responses caused by electrostimulation were recorded by electromyography and muscle contractions were palpated in the lower extremities. A low threshold of muscle response and greater stimulation of the muscle responses than expected were used as criteria to select rootlets for the SDR. If stimulation of the penis or clitoris evoked an electrical response in a selected rootlet (S-1 or L-5), this rootlet was preserved to prevent bladder and sexual disturbances, respectively. Rhizotomy of selected dorsal rootlets was performed after registration of evoked responses at all investigated levels (so as not to interfere with spinal reflex circuits).

Follow-Up Procedure

All children were invited to participate in this follow-up study, which involved additional evaluations at their regular follow-up visits. One pediatric physiatrist (J.G.B.) and 2 pediatric physiotherapists (P.E.M.S. and M.S.) assessed the children during their visit to the outpatient clinic between April 2006 and April 2007. The assessments included the GMFCS, GMFM-66, physical examination for spasticity, and radiographs of the lumbar spine and hips. Parental questionnaires were used for estimation of side effects, additional treatment and surgery, and improve-

TABLE 1: Characteristics of the 33 patients at baseline

Variable	Value
boys/girls	21/12
age at op (yrs)	
mean ± SD	6.7 ± 2
range	2.9–12.1
GMFCS preop level	
I	7
II	7
III	19
GMFM-66 preop score*	
mean ± SD	58.6 ± 11.4
range	46.9–83.0

* Twenty-eight patients.

ment in the child's functioning after SDR. The GMFCS and GMFM-66 results at baseline and 1 year after SDR were reviewed retrospectively from medical records. For analysis, we grouped children with GMFCS Levels I and II and compared them with children with GMFCS Level III. The study protocol was approved by the local medical ethics committee. Informed consent was obtained from all parents and from children over 12 years of age.

Outcome Measures

Gross motor function was measured with the GMFM-66, a measure that was specifically designed to assess the functional abilities of children with CP.[16] The GMFM-66 score, calculated with the Gross Motor Ability Estimator, addresses the linearity of the individual item scores across the entire range of GMFM-66 scores.[1]

Spasticity was assessed according to a modified Tardieu scale[2] in the rectus femoris, hamstring, short adductor, soleus, and gastrocnemius muscles of both legs by the pediatric physiatrist. In this modified Tardieu scale, spasticity is clinically assessed by passively moving the joint at specified velocities (slowly and rapidly) and rating the intensity and duration of the muscle reaction to stretch and the joint angle at which this muscle reaction is first felt. First, the muscle was stretched 3 times with very slow velocity (> 3 seconds) to record the maximum ROM of each muscle. Spasticity was assessed by stretching the muscle at a high velocity (full ROM within 1 second). If there was a clear catch within the ROM, with or without blocking further movement, this was recorded as spasticity.

A radiograph of the spine was obtained, and scoliosis was defined as a Cobb angle of 20° or more. At baseline and at long-term follow-up a radiograph of the pelvis was obtained. The anteroposterior pelvic radiographs were scored according to the Migration Index.[14] In the Netherlands, a Migration Index > 30% indicates increased lateralization (hip at risk), and a Migration Index > 50% is classified as subluxation. Age and disease progression are also considered before surgery: a stable lateralization of 30% in a child older than 4 years is not considered an indication for surgery.

The parental questionnaire was directed at side effects and additional treatment such as botulinum toxin type A or orthopedic surgery after SDR. In addition, all medical records were checked retrospectively for this information. One question was directed at the parents' rating of their child's overall improvement in functioning after SDR on a 7-point scale (no change, or slightly, moderately, or very much improved or worse).

Statistical Analyses

Statistical analyses were performed using SPSS version 15.0 for Windows. A paired t-test was used to analyze the difference between GMFM-66 scores at baseline and in the short-term (1 year) as well as the long-term (3–8 years) after SDR. The independent t-test was used to analyze the difference in short- and long-term improvements in GMFM-66 score between children at GMFCS Levels I and II and Level III. A probability value < 0.05 was considered statistically significant.

Results

Gross Motor Function Classification System

At baseline, 7 children were classified as GMFCS Level I, 7 as Level II, and 19 as Level III. At the 1-year follow-up, the GMFCS grades of 2 children increased 1 level (1 from Level III to II, 1 from Level II to I), and 1 child's grade dropped a level (from Level II to III), but recovered at the long-term follow-up. However, 30 (91%) of the 33 children remained at the same GMFCS level after SDR. At the long-term follow-up (27 total patients), 1 child's grade changed from Level III at the short-term follow-up to Level II at the long-term follow-up, and 1 child dropped from Level III at the short-term follow-up to Level IV at the long-term follow-up. All of the other 25 children's GMFCS grades remained stable.

Gross Motor Function Measure–66

Some GMFM-66 data were missing at baseline or at short- or long-term follow-up. At the short-term follow-up, the mean GMFM-66 scores for the entire group improved significantly by 4.3 points (range –2.53 to 12.31 points; 24 patients; $t = 5.18$, $p < 0.001$). The 8 children at GMFCS Levels I and II improved significantly more (mean improvement 7.2 points) than the 16 children at Level III (mean improvement 2.9 points; $t = 2.74$, $p = 0.012$). At the long-term follow-up, the mean GMFM-66 scores for the entire group improved significantly by 6.5 points (range −0.35 to 23.66; 23 patients; $t = 5.28$, $p < 0.001$). No significant difference in improvement was found between the children at GMFCS Levels I and II versus the children at GMFCS Level III (Table 2).

Spasticity

At short-term follow-up, 7 children had residual spasticity: in 2 children in the hamstring muscles in 1 leg, and in 5 children in the gastrocnemius muscle (in both legs in 3 children). In these 3 children only limited rhizotomy of S-1 could be performed, because the S-1 nerve rootlets were responsible for bladder or sexual function as tested perioperatively. Apart from the residual spasticity, at long-term follow-up spasticity did not relapse.

Long-Term Side Effects

Three children experienced long-term spinal side effects after SDR (Table 3). One child experienced spontaneous fusion of spinous processes at L2–5, 1 child had spondylolysis and listhesis from L-3 to L-4, and 1 child had scoliosis (Cobb angle 21°). No other side effects were found in the medical records.

Additional Treatment

Thirteen children (39%) received botulinum toxin type A treatment of the gastrocnemius muscle or hamstrings muscles because of muscle shortening. Seven patients received 1 treatment, 5 received 2 treatments, and 1 received 3 treatments. Nine of these children were GMFCS Level III and 4 were Level II. The botulinum toxin type A treatment took place between 1 and 8 years (mean 3.5 years) after SDR.

TABLE 2: Short and long-term effects of SDR on GMFM-66*

GMFCS Group (no. of patients)	Preop GMFM-66 Score ± SD	Follow-Up GMFM-66 Score ± SD	Mean Improvement ± SD	p Value (dependent t value)	p Value† (independent t value)
short-term follow-up					
total (24)	56.6 ± 10.5	60.9 ± 12.4	4.3 ± 4.1	<0.001 (5.18)	
Level I & II (8)	69.0 ± 8.9	76.2 ± 8.5	7.2 ± 4.0		0.012 (2.74)
Level III (16)	50.4 ± 3.0	53.3 ± 3.9	2.9 ± 3.4		
long-term follow-up					
total (23)	57.5 ± 11.4	64.0 ± 12.8	6.5 ± 5.9	<0.001 (5.28)	
Level I & II (7)	73.0 ± 7.3	79.0 ± 8.5	6.0 ± 4.4		NS
Level III (16)	50.7 ± 3.0	57.4 ± 7.8	6.7 ± 6.6		

* Values are presented as means ± SDs unless otherwise indicated. Abbreviation: NS = not significant.
† Difference in GMFM-66 scores between Levels I and II and Level III was significant at short-term follow-up (p = 0.012; t = 2.74), but not at long-term follow-up.

Ten children (30%) required orthopedic surgery between 2 and 7 years after the SDR (mean 4 years and 4 months ± 16 months) to maintain walking ability (18 total operations; Table 3). Nine children had severe pes planovalgus and needed a subtalar extraarticular arthrodesis operation for realigning the foot, sometimes combined with soft tissue surgery (medial hamstring muscle lengthening and/or gastrocnemius muscle myotenotomy). Three children (2 of whom also underwent subtalar arthrodesis) developed hip subluxation (Migration Index > 50%) at 1, 2, and 7 years after SDR, respectively, and underwent surgery. Eight of these children were GMFCS Level III and 2 were Level II.

Parental Opinion of Improvement

The parents of 6 children with only short-term follow-up data rated the functioning of their child as moderately improved (3 children) or very much improved (3 children).

TABLE 3: Side effects, surgery, and additional treatment after SDR*

Case No.	Age at SDR (yrs, mos)	GMFCS Level	Spinal Deformities (yrs after SDR)	Type of Op (yrs after SDR)	Botulinum Toxin A Treatment (yrs after SDR)
1	9, 11	III	fusion of spinous processes at L2–5 (6)		none
2	5, 8	III	spondylolysis & listhesis L3–4 (8)	subtalar arthrodesis both feet & re-arthrodesis lt (5, 7)	5
3	8, 9	III	scoliosis (Cobb angle 21°) (5)		none
4	3, 8	III	none	subtalar arthrodesis & FDO (2) for hip subluxation (1)†	none
5	4, 1	III	none	DVIO (5) for hip subluxation (2) both hips	none
6	7, 3	II	none	subtalar arthrodesis (3) & hip op for progressive dysplasia (6)	4
7	4, 11	III	none	subtalar arthrodesis & TEO (5)	7
8	5, 2	III	none	subtalar arthrodesis & TEO (3, 5)	1, 3
9	2, 9	II	none	subtalar arthrodesis & GM (5)	2, 5
10	6, 5	III	none	subtalar arthrodesis (3)	none
11	5, 2	III	none	subtalar arthrodesis (4)	1, 3, 4
12	5, 0	III	none	subtalar arthrodesis & GM (5)	1, 4
13	6, 0	III	none	none	1
14	10, 11	II	none	none	1, 2
15	8, 1	III	none	none	3
16	5, 7	III	none	none	5
17	6, 11	III	none	none	7
18	5, 9	II	none	none	4, 8

* DVIO = derotational varus intertrochanteric (femur) osteotomy; FDO = femur derotational osteotomy; GM = gastrocnemius myotenotomy; TEO = tibia endorotational osteotomy.
† Preoperative radiographs showed a subluxation.

At long-term follow-up, 22 parents answered the question about their child's overall functioning. Twenty (91%) of these 22 parents believed that their child's functioning had improved since baseline (slightly in 5 children, moderately in 11 children, and very much in 4 children), and 2 parents (9%) believed that their child's functioning was worse (slightly worse in 1 child and moderately worse in 1 child).

Discussion

In the present study we evaluated the short- and long-term effects of SDR on gross motor function and spasticity, and compared the effects between children walking without and with ambulatory aids. In addition, we investigated the occurrence of long-term side effects, necessity of additional treatment and surgery, and parental opinion about improvement in their child's functioning after SDR.

The children in our cohort improved significantly in gross motor function in the short-term (1 year) and long-term (mean 6 years) after SDR. This result is similar to the results of other studies.[3,4] This improvement in gross motor function resulted in only a few children receiving a change in GMFCS level. This stability in GMFCS level is in agreement with the findings of Chan et al.[3] who reported that only 1 of 21 children changed GMFCS level 1 year after SDR.

We found a larger improvement in GMFM-66 score 1 year after SDR (4.3 points) than McLaughlin et al.[9] The difference with our results might be explained by the inclusion criteria of the children. In our study we only included children at GMFCS Levels I, II, and III. Our finding that children at GMFCS Levels I and II improved significantly more than children at GMFCS Level III during the first year of follow-up is in contrast to the results of the meta-analysis[9] in which GMFCS classification was not related to outcome. However, it can be hypothesized that children at GMFCS Levels I and II have better initial neuromuscular control and can benefit from the release of spasticity faster than children at GMFCS Level III. At the long-term follow-up, mean GMFM-66 scores changed significantly from baseline in the entire study group, with no difference in change in GMFM-66 score between children at GMFCS Levels I and II and Level III, indicating that children at Level III can achieve a similar improvement as children at Levels I and II.

As in other studies,[10,22] we found that spasticity had disappeared completely in the majority of the children, and no relapse of spasticity occurred in the long term. However, abnormal movement patterns such as walking on toes and with internal rotation of the hips in the lower extremities persisted in some children, indicating that spasticity is only one component of the spastic motor disorder. Abnormal muscle activation due to the brain damage causing CP, demonstrated by muscle weakness and impaired motor coordination, is obviously persistent after SDR, although spasticity has disappeared.

In our study, a low percentage of children (9%) developed spinal deformities after SDR, compared with other studies in which percentages of 12%–20% for spondylolysis or Grade I spondylolisthesis[6,7] and 24%–55% for scoliosis[6,20] were reported. An explanation for this low percentage of orthopedic adverse effects might be the selection of only ambulatory children in our study.

Our finding that 39% of the children received botulinum toxin type A treatment after SDR is consistent with reports in the literature.[10,11] Treatment with botulinum toxin type A after SDR was only partly used to decrease residual spasticity. For most children, the reason for botulinum toxin type A treatment in combination with serial casting was to maintain ROM of the ankle joint.

The percentage of children who underwent orthopedic surgery after SDR in our study (30%) was also within the range of percentages in previous studies,[7,8,10,12] and is low in comparison with the 65% reported by Steinbok.[19] The subtalar extraarticular arthrodesis was the most frequently performed operation, although foot deformity also frequently develops in children with CP who do not undergo SDR treatment. Of the children who underwent orthopedic surgery, 80% were GMFCS Level III. This data is consistent with the results of an earlier study in which it was found that children who walked independently after SDR underwent fewer orthopedic operations than children who walked with assistance.[12]

Conclusions

This study showed a significant improvement in gross motor function and a reduction or disappearance of spasticity after SDR at short- and long-term follow-up. However, the disappearance of spasticity did not prevent secondary orthopedic deformity. Ongoing follow-up and treatment is necessary to maintain mobility after SDR, especially in children using walking aids.

Disclosure

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