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Multidimensional outcome measure of selective dorsal rhizotomy in spastic cerebral palsy

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Abstract

Background. One of the treatment option to reduce spasticity in [cerebral palsy](http://topics.sciencedirect.com/topics/page/Cerebral_palsy) children is [selective](http://topics.sciencedirect.com/topics/page/Selective_dorsal%20rhizotomy) [dorsal rhizotomy.](http://topics.sciencedirect.com/topics/page/Selective_dorsal%20rhizotomy) Several studies have demonstrated short and long term improvements in [gait](http://topics.sciencedirect.com/topics/page/Gait) and other activities after [rhizotomy](http://topics.sciencedirect.com/topics/page/Rhizotomy) but this surgery still remains a controversial procedure and patient outcome indicators measures are not uniform.

Aims. To describe our assessment and outcome evaluation protocol and to verify by this protocol short term results of rhizotomy.

Methods. We recruited 9 cerebral palsy children (mean age 7.9 years \pm 3.2) affected by mild to moderate spastic diplegia and operated by rhizotomy. Patients were studied preoperatively and at 12 months after surgery by the following clinical and instrumental measures correlated to the International Classification of Functioning: modified Ashworth Scale, passive Range of Motion, Medical Research Council Scale, Selective Motor Control Scale, 3D-motion analysis and energy cost of locomotion measurements (indicators of "body functions"); Gross Motor Functional Measure and Motor Functional Independence Measure (indicators of "activities and participation").

Results. Our data showed, after rhizotomy, reduction of spasticity specially in plantarflexors muscles $(p < 0.01)$, increase of strength of knee flexors/extensors and foot plantar/dorsiflexion muscles ($p < 0.01$), improvement of selective motor control ($p < 0.05$), more similar spatiotemporal parameters of [gait analysis](http://topics.sciencedirect.com/topics/page/Gait_analysis) to healthy subjects, reduced equinus foot and knees hyperflexion as energy cost.

Conclusion. The complementary use of multiple indicators may improve the evaluation of the results of dorsal rhizotomy. A beneficial outcome measured by these indicators has been found in our spastic diplegic children one year after rhizotomy.

Keywords: [Rhizotomy;](http://topics.sciencedirect.com/topics/page/Rhizotomy) [Cerebral](http://topics.sciencedirect.com/topics/page/Cerebral_palsy) palsy; Muscle spasticity; [Gait;](http://topics.sciencedirect.com/topics/page/Gait) Oxygen consumption

1. Introduction

Dorsal [rhizotomy](http://topics.sciencedirect.com/topics/page/Rhizotomy) was first performed in the humans for relieving spasticity by Foerster at the beginning of XX century.^{[1](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib1)} Due to the [comorbidity](http://topics.sciencedirect.com/topics/page/Comorbidity) associated with the extensive section of the roots proposed, the operation was almost abandoned for half a century. In the sixties Gros in France revaluated the procedure by performing partial [rhizotomies](http://topics.sciencedirect.com/topics/page/Rhizotomies) and then avoiding the excessive sensory loss produced by the Foerster's operation. The term of [Selective Dorsal Rhizotomy](http://topics.sciencedirect.com/topics/page/Selective_Dorsal%20Rhizotomy) [\(SDR\)](http://topics.sciencedirect.com/topics/page/SDR) was also introduced at that time.^{[2](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib2)} In the seventies Fasano^{[3](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib3) and [4](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib4)} in Italy refined the procedure by developing a set of criteria for choosing the dorsal rootlets to be cut, based on abnormality of evoked motor responses to their electrical stimulation. In the authors opinion these neurophysiological criteria could increase the selectivity of the rhizotomy, maintaining the effect on spasticity and further decreasing the sensory loss. Then Peacock^{[5](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib5)} in South Africa shifted the site of SDR from the conus [medullaris](http://topics.sciencedirect.com/topics/page/Conus_medullaris) region to the [cauda equina](http://topics.sciencedirect.com/topics/page/Cauda_equina) and popularized this procedure in the USA when he moved there. Many centres all over the word have utilized variations of the original technique, mainly regarding the approach to the [lumbo-sacral](http://topics.sciencedirect.com/topics/page/Lumbo-sacral) rootlets, that is done either at the intraforaminal or the juxtamedullary level.^{[6,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib6) [7](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib7) and [8](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib8)}

After more than 30 years of experience using this surgical option, several studies^{[9,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib9) [10,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib10) [11](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib11) and [12](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib12)} have demonstrated that spasticity can be significantly and permanently reduced and improvements in [gait](http://topics.sciencedirect.com/topics/page/Gait) and other activities can be achieved after SDR. Nevertheless, this therapeutic option remains a controversial procedure, and patient selection criteria, surgical technique and outcome indicators are not uniform $1\overline{3}$

Furthermore, after the introduction of [Intrathecal](http://topics.sciencedirect.com/topics/page/Intrathecal) [Baclofen](http://topics.sciencedirect.com/topics/page/Baclofen) (ITB) administered by an implantable pump, many authors claimed that this procedure should be preferred because less invasive, reversible in its effects and more effective in tetraplegic patients, especially if [dystonia](http://topics.sciencedirect.com/topics/page/Dystonia) is associated with spasticity.^{[14](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib14) and [15](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib15)} The choice between ITB pump versus SDR is still under debate. All the authors agree that SDR is contraindicated when dystonia other than spasticity is the main disabling condition. In the other cases the prevalent opinion is that SDR can be an option (sometimes a second option) in spastic quadriplegic and in more severe diplegic children, but should be preferred in the ambulatory non-dystonic children. Actually the children with good level of voluntary mobility, good residual strength, and sufficient cognitive performances to be adequately rehabilitated after the operation achieve the greatest functional benefits from SDR, avoiding long-term management need and complications of ITB.^{[16,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib16) [17,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib17) [18](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib18) and [19](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib19)} On the other hand the selection of candidates for selective dorsal rhizotomy is sometimes difficult in this group of children, because the risk of inducing a deterioration of motor functions is always a concern. Also the evaluation of the results can be incomplete if an appropriate evaluation protocol is missed.

Many validated method for the pre- and post- operative assessment of children with spastic CP are currently used. Many studies focuses on one or few on them, leading to sometimes opposite conclusions. For example, clinical assessment of the short term effect on spasticity (1–3 years after rhizotomy) uniformly showed reduction of lower limbs spasticity, and increase in the motor function.^{[20,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20) [21](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21) and [22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22)} However when evaluating speed of locomotion, some studies reported an increase in the velocity^{[12,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib12) [20,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20) [21](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21) and [23](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib23)} whereas some others, reported no change or even a decrease.^{24,} ^{[25](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25) and [26](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib26)} When evaluating spatio-temporal parameters of locomotion, many studies showed increase of step length^{[25,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25) [26](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib26) and [27](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib27)} even when normalized by the legs length of the subjects^{[26](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib26)} or when the age was taken into account.^{[25](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25)} Also concerning lower limb joints range of motion during walking, there is a general consensus that after SDR it improves and the improvement is quite maintained over time.^{[10,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib10) [11](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib11) and [21](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21)} Some studies described also a joint [kinematics](http://topics.sciencedirect.com/topics/page/Kinematics) more similar to that of healthy children on a short term outcome.^{[20,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20) [22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22) and [25](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25)}

A systematic review by Grunt et coll^{[28](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib28)} looking at the long-term outcomes after SDR concluded that the studies are too few and the number of subjects is often too small to allow a definitive conclusion and recommendation. Moreover there is no evidence of long term significant benefits from SDR. In some works^{[29](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib29) and [30](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib30)} even a deterioration after the initial improvements has been observed, even though the effect on spasticity is maintained after up to 10 years. Improvement in range of motion is also maintained after rhizotomy and more similar to healthy subjects both during walking^{[27,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib27) [31](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib31) and [32](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib32)} and in passive condition.^{[10](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib10)} Effects on speed of walking differ in the short term (improvement) versus long term (not significantly different from no operated patients with CP).^{[27](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib27) and [31](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib31)}

Trost et al.^{[13](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib13)} described an increase in the economy of locomotion in half of their patients. Chan et al.[22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22) described a reduction of oxygen consumption one year after rhizotomy but without statistical significance.

The collaboration between the "E. Medea" Scientific Institute and the University of Turin started in 1994 and more than 30 patients underwent SDR in this lapse of time. After the first experience of rehabilitation treatment and of outcome assessment, in the last 3 years we identified an hopefully complete multidimensional protocol for selection and outcome assessment of these patients; we feel that this protocol, developed according the experience of previous studies,^{[13,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib13) [22,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22) [23](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib23) and [28](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib28)} could have proven to be practical and helpful in selecting the appropriate candidate for SDR as well as in providing the required training and the outcome monitoring of the operated children.

Aims of our study are i) to describe our clinical and instrumental assessment and outcome evaluation protocol in a population of children with spastic CP operated on by SDR, ii) to verify the efficacy of SDR at one year follow up using this protocol.

2. Material and methods

2.1. Population

Nine children (6 males and 3 females) with a mean age of 7.9 ± 3.2 years, mean weight of 23.0 ± 11.7 kg and mean height of 117.0 ± 16.8 cm underwent SDR in the last 3 years. All were spastic diplegic non-dystonic children, affected by CP. Seven of them were able to walk without aids (GMFCS level II). Two were able to walk only with aids (GMFCS level III). All the children had normal intelligence.

Selection for surgery was based on the following criteria: 1) diagnosis of spastic diplegia due to CP, as verified by history, clinical/instrumental examination and neuroimaging findings; 2) age between 4 and 12 years; 3) pure spasticity (no dystonia); 4) antigravitary muscle strength: mean lower limbs power >3 on Medical Research Council Scale (MRC)^{[33](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib33)}; 5) moderate to good motor control and movements selectivity; 6) ambulation ability with or without aids; 7) no previous orthopedic surgery; 8) absence of severe fixed joint deformity; 9) adequate cognitive function; 10) well motivated child with good compliance to the treatment; 11) good family/social support. Eligible patients were evaluated by a multidisciplinary team composed by neurodevelopment physiatrists, physiotherapists, rehabilitation engineers and neurosurgeon.

2.2. Evaluation protocol

Based on the literature^{[13,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib13) [22,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22) [23](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib23) and [28](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib28)} and on our past experience, we elaborated the following protocol to select the candidates for SDR, to obtain a preoperative baseline evaluation, and to assess the results:

- a) Measure of the passive Range of Motion (pRoM) of the joint of the lower limbs with a manual goniometry;
- b) Ashworth Scale modified by Bohannon and Smith (Ash), to measure the muscle tone^{[34](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib34)}:
- c) Medical Research Council Scale (MRC) to assess the muscular strength of the lower limbs^{[33](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib33)}:
- d) Selective Motor Control Scale (SMCS) to evaluate the selective motor control of the lower extremities 35 :
- e) Gross Motor Functional Measure $(GMFM)^{36}$ $(GMFM)^{36}$ $(GMFM)^{36}$ to evaluate the gross motor function and ability;
- f) Functional Independence Measure, children version (WeeFIM), 37 especially the motor component, to assess the functional level of mobility and the severity of motor disability;
- g) 3D-motion analysis of walking with a 6 cameras optoelectronic system working at 60 Hz (Smart-E™, BTS, Milan, Italy) to analyze spatio-temporal and kinematic parameters. The marker set of Davis protocol was used^{[38](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib38)}; each subject performed one standing trial and at least 8 trials of walking at self-selected speed along a pathway of 6 m;
- h) Energy cost measurements of walking by a breath-by-breath portable metabolimeter (Cosmed $K4b^{2TM}$, Rome, Italy) that measure the oxygen uptake and hence to calculate the energy cost of locomotion.^{[39](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib39)} The device was calibrated according to the constructor's recommendations. Cardiopulmonary parameters for each child were measured at rest and at different speeds (0.6, 1.2, 1.8, 2.4, 3.0, 4.2 and 6.0 km h^{-1}) until the one that the child could perform safely, on a motor driven treadmill (Galaxy MTC Climb, Runner, Italy). Then the average of the breath-bybreath values in the last minute of rest and at the steady state at each speed was calculated as in Marconi et al.^{[40](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib40)}

All these evaluations were repeated before (within two weeks, T0) and 12 months (T1) after SDR in all children. To the two children with GMFCS level III, 3D-motion analysis of walking were not applied.

The evaluation procedure was studied in order to cover all components of the child functioning following the bio-psycho-social model embedded in the International Classification of Functioning (WHO 2001),^{[41](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib41)} as in previously study of Langerak et coll.^{[42](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib42)} Passive range of motion, strength and resistance to passive movement (spasticity), energy expenditure and gait pattern are indicators of body functions; GMFM and FIM are indicators of activities and participation [\(Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig1) 1).

Fig. 1. Representation of the proposed assessment protocol in the ICF-model.

2.3. Neurosurgical procedure

All children were operated on by the same neurosurgeon at the Neurosurgery Division of the University of Turin (Italy). The original juxtamedullary approach introduced by Fasano was utilized.^{[43](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib43)} Each patient received general anesthesia, and was then placed in a prone position with bolsters under the chest and pelvis. Needle electromyographic recording electrodes were placed in major muscle groups of both the lower extremities (ileo-psoas, rectus femoris, biceps femoris, adductors, tibialis anterior, gastrocnemius medialis), and in the anal sphincter to monitor the lumbosacral rootlets. Only a limited laminectomy/laminotomy of the first lumbar vertebra (L1) was performed. After opening the dura, the dorsal lumbo-sacral rootlets were exposed. Small bundles of 3–5 rootlets were isolated and a 0.5 Hz train of stimuli was applied to recognize the investigated muscles. Then a 50 Hz train of stimuli was applied at the threshold intensity for muscular contraction. The electromyographic and clinical muscular responses were recorded on a form and classified as normal, slightly abnormal (tetanic), and markedly abnormal (tetanic and diffuse). The dorsal nerve rootlets associated with a normal response were left intact, whereas those associated with a markedly abnormal response were completely cut. Rootlets with slightly abnormal electromiographic responses were cut for 1/2 - 1/3 if the more spastic muscles were involved. Following this criteria from 18% to 53% (mean 41%) of the stimulated rootlets have been sectioned.

2.4. Postoperative rehabilitation treatment

The main goal of postoperative rehabilitation program was to stretch the muscles and to promote the acquisition of a new modality of movement without or with less spasticity. The children needed to strengthen their muscles and started using them in a previously unfamiliar way; so children needed motor learning. The rehabilitation focused firstly on basic skills like correct alignment on the wheelchair, trunk control, stretching at lower limbs, strengthening unloaded exercises for lower limbs and trunk. Following this initial approach exercises focused on crawling, standing, walking

and performing self-care activities (such as washing and dressing/undressing), the most complex motor activities were introduced gradually. After fourth week of rehabilitation treatment, in patients who presented still loss of strength of one or two muscle (MRC scale \lt 3), neuromuscular- electrostimulation was used to strength the muscle (especially gluteus, quadriceps, dorsiflexors muscles) for about 30 min a day, until muscle strength recovery. Generally from the fourth week of rehabilitation treatment, the children started to use an orthopedic bicycle and to walk with anklefoot orthosis, if necessary, with a walker or tripods or crutches. If the children started advanced activities too soon, there was the risk of reverting to abnormal movement patterns, used before surgery. We usually performed six-eight weeks (from the 2nd to the $7th$ -9th week after SDR) of inpatients intensive rehabilitation (2 sessions/day); then the rehabilitation was continued for about a year in outpatient's modalities, in order to achieve maximum functional outcome.

2.5. Data analysis

We investigated the normal distribution of clinical and instrumental parameters and we verified the violation of this assumption for clinical and kinematic data. We used a robust nonparametric Fisher's sign test for data analysis on SCALE, MRC, Ashworth and FIM scores, between paired observations. We therefore proceeded to a nonparametric analysis of the different kinematic parameters evaluated at T0 and T1 through the use of two-tailed sign test. Otherwise for energy cost the normal distribution has been verified and we used a parametric two-tailed t-test. Significance was set at a *p* values <0.05 in both. At least eight walkings for each patient were recorded. We analyzed before the mean between the right and left side for each walk performed and lastly the mean of all walks for each patient. For the kinematic data, all the curves were normalized with respect to 100% of the gait cycle duration. For the comparison at T0 and T1, the statistical analysis of joint rotation was performed both on the entire cycle and on eight specific subphases, 44 as suggested by Del Din et al.^{[45](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib45)} The subdivision was customized on each patient gait pattern basing on the succession of contact and lifting of each foot from the floor. We considered as parameters of interest for the entire cycle range of motion, maximum and minimum of the curve. For each subphase we choose to consider range of motion, mean angular amplitude and the value of the starting angle.

Regarding the oxygen consumption, for all the patients energy cost of locomotion has been plotted as function of the different velocities. Then for each child, the data of energy cost of locomotion as function speed have been interpolated by a second-degree equation. Finally, by equaling to zero the first derivative of the quadratic equation, the minimum of the curves was found and it was considered to correspond to the optimal speed.

This study was approved by our Institutional Review Board.

3. Results

3.1. Clinical evaluation

The results of the statistical analysis of the clinical evaluation are presented in [Table](http://www.sciencedirect.com/science/article/pii/S1090379814000907#tbl1) 1 and in [Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig2) 2. After SDR there is an improvement of all the studied clinical parameters. We found a statistically significant reduction of spasticity, as measured by the modified Ashworth scale, especially in the plantarflexors muscles $(p < 0.01)$, a statistically significant increase of the strength of the knee flexors/extensors and foot plantar/dorsiflexion muscles $(p < 0.01)$ as measured by MRC Scale, a statistically significant improvement of the selective motor control $(p < 0.05)$. There were not statistically different between pre and post treatment in the muscle tone and muscle strength of the hip adductors, hip extensors, hip flexors muscles. Also pRoM showed a general improvement, statistically significant as far as knee extension ($p < 0.001$ with hip at 90 $^{\circ}$ of flexion) and foot dorsiflexion ($p = 0.019$ with knee in neutral position; $p = 0.044$ with knee at 90° of flexion).

Results of statistical analysis of clinical evaluations, obtained by Selective Control Assessment of the Lower Extremity (SCALE), Gross Motor Functional Measure (GMFM), Functional Independence Measure for Children (Wee-FIM) (total and motor scale), passive range of motion (pRoM) of knee and ankle, muscular strength with Medical Research Council Scale (MRC) and muscle tone with Ashworth Scale (Ash) parameters, measured before and after selective dorsal rhizotomy (SDR). Highlighted cells indicate significant differences (at least $p < 0.05$).

Fig. 2. Boxplots of clinical evaluations, obtained by: Selective Control Assessment of the Lower Extremity (SCALE) total score; Gross Motor Functional Measure (GMFM); Ashworth Scale for muscle tone: knee extensors (KE), knee flexors (KF), foot plantaflexors with knee at 0° (FPF K0°), foot plantaflexors with knee at 90° (FPF K90°); Functional Independence Measure (FIM): total, motor scale; Medical Research Council Scale (MRC) for muscular strength: knee extensors (KE), knee flexors (KF), foot plantaflexors (FPF), tibialis anteriors (TA), foot lateral dorsiflexors (FLDF). All the evaluations was performed before (white boxes) and after selective dorsal rhizotomy (gray boxes). Significant differences between before and after treatment are marked with a symbol (p value < 0.05).

Regarding the functional outcome, there was a significant reduction of the disability measured by the WeeFIM ($p < 0.05$) and a non significant increase in the score of GMFM.

3.2. 3D motion analysis

One year after SDR all spatio-temporal parameters were more similar to the data corresponding to healthy subjects but only cadence resulted significantly different at T1 compared to T0 (*p* < 0.001).

The results of the statistical analysis of kinematic data are presented in [Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig3) 3. Statistically significant differences from T0 were on the foot progression, on the plantar-dorsiflexion and on the knee flexion/extension: the equinus foot was reduced especially in terminal stance and initial swing phase and the hyperflexion of the knees were in the stance phase.

Fig. 3. Kinematic gait curves Gait analysis was performed before selective dorsal rhizotomy (mean between subjects, continuous line) and after selective dorsal rhizotomy (mean between subjects, dashed line). Data are compared with ones of healthy subjects provided by the equipment constructor (grey band). For each gait subphase significant differences between before and after selective dorsal rhizotomy (*p* value < 0.05) are marked in terms of mean amplitude (black x), range of motion (black o), and starting angle (black +).

3.3. Energy cost and optimal speed

[Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig4) 4 shows the gross metabolic cost values as a function of speed, recorded at 0.6, 1.8, 3.0, 4.2 km h⁻¹ tested speed on treadmill. The highest tested speed was 6.0 km h⁻¹ but no one patient reached 6.0 km h^{-1} in the pre-SDR phase and we decided not to considerate such speed in data analysis. [Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig4) 4 shows that for each tested speed energy cost of locomotion after SDR are lower than before SDR, in mean values and for each patient.

Fig. 4. K4 data for: energy cost of walking (Cw) as function of speed (v) for selected walking speeds, with characteristic U-shape patterns (A); second grade polynomials as interpolating functions of the Cw data as a function of speed (B); Heart rates (HR) as function of speed (C); Oxygen uptakes (VO2⋅kg⁻¹) as function of speed (D). K4 was performed before selective dorsal rhizotomy (mean value between subjects marked with black square and continuous line) and after (mean value between subjects marked with black circle and dashed line. Uncertainty bars are drown in the same format). Significant differences between before and after treatment are marked with a symbol (p value < 0.05).

For all the patients energy cost of locomotion has been plotted as function of the different velocities. Energy cost data were fitted with 2nd order polynomial functions, both before and after the SDR (before: $C(v) = 2.2297v^2 - 15.631v + 34.411$, $R2 = 0.9892$; after SDR: $C(v) = 1.8353v^2$ $12.776v + 27.936$, $R2 = 0.9997$). These functions present the typical energy cost "U shape pattern" and show minimums at 7.02 km*h-1 and at 5.70 km*h-1, before and after SDR respectively. These minimums can be considered to correspond to the optimal speed. Furthermore before SDR energy cost of locomotion tends to decrease more monotonically with speed than after SDR. At 0.6 km*h-1 heart rate and oxygen uptake resulted statistically significant reduced after surgery.

4. Discussion

In our study an assessment and outcome evaluation protocol for spastic children with CP operated on by SDR is presented.

The protocol contains the following validated clinical and instrumental measures, commonly used in the neurorehabilitation units: pRoM, modified Ash scale, MRC scale, SCALE, WeeFIM, GMFM. They explore the different components of the ICF:

- body structure and function, by pRoM, Ash, MRC, SCALE, 3-D gait analysis, energy cost measurements;
- activities/participation, by GMFM and FIM.

These measures allow a more comprehensive representation of child's functioning. They permit a more firmly grounded evaluation of the effectiveness of SDR.

A multidimensional assessment approach has already been adopted by previous early studies and controlled clinical trials.^{[46,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib46) [47](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib47) and [48](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib48)} With this approach, comparing diplegic children that underwent SDR plus intensive physiotherapy with similar children treated only with physiotherapy, a significant improvement in gross motor function was found in SDR by Steinbock et al. 46 46 46 and Wright et al.^{[48](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib48)} On the other hand, the study of Mac Laughlin et coll.^{[47](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib47)} did not found this difference, and concluded that SDR may not be more efficacious than intensive physiotherapy alone in children with mild spastic diplegia.

In our study 7 out of 9 children were affected by mild spastic diplegia (GMFCS level II): in these children we observed a positive improvement in the clinical measured data and the gait analysis results. Therefore we found that SDR for this group of high functioning children is beneficial in their gait training and gait performance. These findings concurred the previous studies of short term improvement for high functioning GMFCS level II patients^{[22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22)} maintained at long term.^{[32](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib32)}

In conclusion, if at the earlier days SDR was recommended for GMFCS level III patients, nowadays, with more collected data, it seems to help also the higher function CP children in terms of mobility gain. Our protocol is quite simple and met good compliance with the patients and their family (and the operators!).

Finally our experience showed that the contextual factors, personal and environmental, play an important role. Particularly the motivation of the child to improve his condition, his good compliance to the surgical and rehabilitative treatment, and a strong support by the family are fundamental to achieve the best functional results. These factors should be considered, as we did, among the eligible criteria of a candidate for SDR.

As far as the results at one year follow-up, our study corroborates the results of other previous reports, showing for all patients undergoing SDR a significant motor and functional improvement. $20, 21$ $20, 21$ and 22

Resistance to passive movement, as measured by the modified Ash scale, demonstrated a significant reduction, mainly in the hamstrings and in the triceps surae; moreover pRoM were larger than before SDR mainly in the knee and in the hip. These data are similar to those reported in most of the studies, allowing Steinbock in his 2007 review^{[16](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib16)} to conclude that there is very strong evidence that SDR provides a significant improvement of lower limbs spasticity. Spasticity reduction represents the first aim of this intervention and this aim has to be considered as achieved. Moreover on the bases of some long-term previous studies we expect that this reduction of spasticity would be maintained for at least $5-20$ years,^{[10,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib10) [12](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib12) and [49](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib49)} then possibly all over the life time.

The muscle strength on MRC Scale for knee flexor and extensor and foot plantar flexors and dorsal flexors muscles are significantly different, showing a higher score after SDR. This has been observed also by Gul et al.,^{[10](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib10)} who found that the quadriceps strength was significantly increased 1 year and 5 years after SDR. Also Engberg^{[20](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20)} documented significant strength gains in children with spastic CP treated by SDR plus physiotherapy compared to children treated only by physiotherapy.

One-year after SDR, there is a statistically significant improvement of the selective motor control (on the SCALE evaluation). These data also are confirmed by other studies.^{[20,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20) [21](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21) and [22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22)}

We could hypothesize that the increase in muscle strength and the improvement of the selective motor control are due to the decrease of spasticity, to the decrease of opposition to agonists exerted by the spastic antagonist, and thus be related to the reduced co-contractions between agonists and antagonists muscles. With reducing of spasticity, the weak muscles without the persistent hyperexcitability allows the training to regain the muscle strength through intensive exercise and motor learning.

Moreover there is great interest for the data reported from the functional MRI findings, showing an important rearrangement of the sensory-motor areas in the brain after SDR.^{[50](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib50)} All these observations strongly support the benefits of reducing spasticity also in terms of improving voluntary movements and rearranging brain areas belonging to the motor system.

We found significant change in WeeFIM evaluation, as in Cole, $2¹$ but not in GMFM. This may be explained by the good gross motor ability the patients showed before SDR (thus revealing a ceiling effect): all the children before SDR had a GMFCS level of II or III, so we could image that the results depends by the no-use of the aids after SDR. The results could be related to the long time needed for some GMFM items to show changes.

Regarding 3D-motion analysis, our post- SDR kinematic data at the various phases of the stride (displacement of ankle, knee flexion and extension movement, foot progression) are often significantly different from the preoperative ones. [Fig.](http://www.sciencedirect.com/science/article/pii/S1090379814000907#fig3) 3 shows that after SDR the walking pattern of patients with CP becomes more similar to the controls, as previously suggested in other studies.^{[21,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21) [22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22) and [25](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25)} The reduction of spasticity is also reflected in pRoM and in the kinematic data, as observed in previous studies.^{[9,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib9) [24,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib24) [26,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib26) [32](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib32)} and [51](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib51) Indeed pROM of the knee and hip are significantly larger after SDR. Thomas et al.[25](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib25) did not find an increased range of motion but they described an increase dorsiflexion in the midstance phase. $Gul¹⁰$ $Gul¹⁰$ $Gul¹⁰$ found an increase in the range of motion of hip (abduction), knee (extension) and ankle (dorsiflexion). pRoM modifications seems to be maintained till 10 years after SDR as reported by Langerak et al.^{[31](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib31)}

The optimal speed and the relative energy cost of locomotion did not change significantly, as in Chan.^{[22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22)} However optimal speed showed a trend towards increase, and energy cost of locomotion towards decrease, suggesting a trend (even if not significant) for the improvement in speed and economy of locomotion. Decreased energy cost, not only during locomotion but all over daily activities of children with spastic CP, is an objective measure of the shift of metabolic resources from spastic muscles to the rest of the body (including brain). Speed of locomotion and related variables such as energy cost of locomotion can be easily influenced also by anthropometry, and consequently by the age. These problems can be resolved by means of normalization procedure,

often employed in case of people of different size such children at different ages. Since no one of our patients showed significant changes in height before and after SDR, we did not consider necessary any normalization procedure.

Good results of a surgical treatment usually mean good selection criteria. This is why we hope that this protocol could help to better select the candidates for SDR, mainly in the more problematic group represented by children in GMFCS level II.

Finally our experience shown that patients should be continuously and systematically monitored by the same multidisciplinary team to meet their needs during their development.

4.1. Study limitations

There was no formal control group for this study, since each patient was control to himself. The sample size was really small. The short-term design was specifically aimed at describing our assessment and outcome evaluation protocol and evaluating the effects of SDR. In this study the inpatient rehabilitation is standardized, but variations inevitably exist. We cannot exclude that the effects here described are due to the presence of the surgical intervention plus the rehabilitation sessions. It should nevertheless be noted that all the treated children were not rehabilitation naïve, and their pre- SDR regimen was the same as that followed after the first 8 weeks post surgery.

5. Conclusion

We think that the key finding of the present paper is that a multidimensional clinical and instrumental assessment of patients with CP allowed us to precisely measure the benefit one year after SDR. Our set up and method was simple and the study met with a good compliance by the patients and their family. The clinical scales used are internationally validated and commonly utilized in rehabilitation units. Nowadays three-dimensional motion analysis is highly diffuse in clinical settings, and it allows quantitative and objective evaluation of biomechanics parameters while subjects are performing specific tasks. Energy cost measurements document how the beneficial shift of metabolic resources from spastic muscle to the entire organism.

According to other previous studies^{[20,](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib20) [21](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib21) and [22](http://www.sciencedirect.com/science/article/pii/S1090379814000907#bib22)} and clinical trials, our study findings show that a multidimensional clinical and instrumental assessment is essential to evaluate the results of SDR. By evaluating the indicators of various dimensions of human functioning SDR, followed by intensive physiotherapy, seems to be an effective treatment for CP children with spastic diplegia, especially for those with high motor function (GMFCS level II) and normal intelligence. In this group of patients SDR leads to a beneficial outcome regarding quantitative gross motor function, performance of functional skills and activities, as well as increased independence in self-care and mobility.

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